

**A GT study of parent adjustment after the birth of a  
child with a facial birthmark.**

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## **1. Abstract**

Little is known about the experiences of parental adjustment following the birth of a child with a facial birthmark due to a scarcity of literature. The literature within the fields of cleft lip and palate (CL/P) and cranio-facial abnormality (CFA) suggest that there are many factors involved in the process of parental adjustment. The aim of this project was to explore how parents adjust following the birth of their child with a facial birthmark. In-depth interviews were conducted with 13 parents of children aged between birth and two years old. The data was analysed using a Constructivist Grounded Theory approach and a model of adjustment was co-constructed from the data. It appears that parents move through a process of adjustment, that begins with coping with the initial shock and uncertainty before moving on to understanding the birthmark and treatment options, making sense of the birthmark, developing coping strategies and adjusting to birthmark. This process is affected by the availability of resources and the extent of social challenges experienced by the parents. The findings of this study indicate that psychology should be routinely involved with the assessment of parental psycho-social well-being and psycho-social support should be promoted for these families. Furthermore, greater training about birthmarks for medical professionals is recommended.

## **2. Introduction to the researcher**

### 2.1 My interest in the field

This study was inspired by my personal experience of having a congenital facial birthmark. I was born with a sebaceous nevus, which is an uncommon type of birthmark. I experienced a degree of social stigma, including bullying from peers and reactions from adults such as stares, remarks and sometimes more invasive responses such as strangers making attempts to wipe it off with the assumption that it was food. In my experience, having strong family and social support makes it much easier to cope. I underwent multiple surgeries throughout my childhood, adolescence and early twenties. The majority of these were attempts to manage the aesthetic appearance of the lesion although the potential skin cancer risk was also a consideration. I felt lucky to grow up in a family where I was able to talk about the challenges I faced and to have support when I had another unpleasant social experience or further surgery.

I have come across a great deal of research literature and personal accounts of people living with visible difference over the years, but I have rarely seen accounts of the experiences of those caring for someone with a visible difference. Through my clinical experiences I have come to appreciate how much 'problems' do not just impact an individual, but affect those around them as well. This sparked an interest to understand more about the difficulties parents face when adjusting to having a child born with a facial birthmark. From the experience of my parents, some of the issues they faced were dealing with the shock of my unexpected appearance at birth; the processes of trying to make the right choices when they had little medical knowledge coupled with contradicting medical advice and supporting me to overcome the challenges of growing up with a visible difference.

### 2.2 My epistemological position

Within current qualitative research methods there is an epistemological diversity. It has been helpful to view the various epistemological positions as a

continuum between positivist and social constructionist viewpoints. It is important to acknowledge my epistemological stance to bring transparency to the enquiry and provide the reader with a framework to understand and evaluate it.

I reject the notion of the existence of a singular human reality as I believe that an individual's experience and understanding is developed in relation to their culture, language, interpersonal interactions and social and historical background. This position is aligned to a social constructionist position (Gergen, 1999). I consider that my identity has been shaped through my experiences and that my experience is not universal to others. Gergen (1999) suggests that people's understanding and identity is constructed in accordance with the social world. Social constructionists are influenced by the way knowledge is named, described and negotiated between people in particular contexts and at particular times (Raskin, 2002). I align myself with this position as I believe that "subjectivity is inseparable from human existence" (Charmaz, 2006, p.14).

### 2.3 The use of the language & terminology

Firstly, it is important to define what is meant by the term 'facial disfigurement' as it is used widely in the literature. Historically the term facial disfigurement was used to describe visible difference. Deconstructing this term into its component parts, facial is defined as 'of or affecting the face' and 'disfigurement' is defined as 'the action of spoiling the appearance of something or someone; defacement' (Oxford Dictionary, 2015). However, I would argue that this terminology is socially constructed as is the concept of what 'spoils' appearance. There has been much discussion about the use of appropriate language to define this phenomenon and Rumsey & Harcourt (2004, 2005, 2012) argue that much of the terminology used has a predominant negative focus as these terms are usually derived from a biomedical and pathological context. Alternative terms including 'visible difference,' 'visible distinction' and others have not been met with universal approval and at present do not have the advantage of evoking the shared

understanding that occurs when the term 'disfigurement' is used (Rumsey & Harcourt, 2004). Nevertheless, I agree with Rumsey & Harcourt's (2004) view that disfigurement has a negative connotation and can have a stigmatising effect. Therefore, the term 'visible difference' as used by Rumsey & Harcourt, (2004) will be used in this paper to refer broadly to congenital conditions such as craniofacial abnormalities (CFA), maxillofacial (MF) conditions such as cleft lip and palate (CL/P) or birthmarks. Where the specific type of facial visible difference is defined it will be referred to by name.

As this study is interested specifically with birthmarks, birthmarks will now be briefly introduced. There are many different types of birthmarks that can present anywhere on the body and vary in colour, texture and aetiology. The most common types of birthmark are either vascular (relating to blood vessels) such as port wine stains (PWS) and haemangiomas or may be related to skin pigmentation (e.g.. café au lait and melanocytic nevi). Some less common types may develop from the malformation of different types of skin cells (e.g.. sebaceous nevi, epidermal nevi). Birthmarks are usually present at birth or develop within the first month of life and are benign although some carry a higher risk of malignancy later in life.

Birthmarks do not only present as a visible difference, there are a number of associated health concerns, for example, facial PWS birthmarks are associated with Sturge Webber Syndrome and, if the birthmark occurs over or near to the eye Glaucoma may develop. Sturge Webber syndrome occurs when the blood vessels in the brain are abnormal and can cause epilepsy or learning difficulties (Enjolras, Riche, & Merland, 1985). Haemangiomas are rarely obvious at birth but begin to develop within the first few days or weeks of life. They typically start as a red spot and then grow to become enlarged, bulbous and can ulcerate. Typically, this type of birthmark can be treated and usually reduces in size eventually reaching the stage of involution (fade away) but sometimes leaving permanent discolouration or scarring and the medications in use have side effects (Tanner, Dechert & Frieden, 1998). Sebaceous nevi are an uncommon type of birthmark that are most often found on the scalp and involve malformation of the sebaceous glands, overgrown

epidermis (upper layers of the skin) and affects the hair follicles and apocrine glands. This type of birthmark has an associated syndrome, Sebaceous Nevus Syndrome. The 'syndrome' is diagnosed when the birthmark is implicated in the development of problems such as epilepsy and learning difficulties (Kaye, 1980). Giant Congenital Melanocytic Nevi can affect the whole body, have a higher risk of malignancy later in life and research has shown that the presence of the birthmark on the face or head is associated with an increased risk of learning disabilities or epilepsy (Koot, De Waard-van der Spek, Peer, Mulder & Oranje, 2008).

### **3. Reviewing the literature about visible difference**

In order to fully understand the context in which this study is situated, I began by reading books, articles and personal accounts of people living with visible difference with particular focus on key authors in the field such as Rumsey, Harcourt, Lansdown and Bradbury. Following consultation with Dr Nicola Stock (Research Fellow) at the Centre for Appearance Research in Bristol, additional papers were sourced, however the focus of much of this literature was in the field of CL/P. Birthmark specific literature is scarce therefore I have chosen to introduce the reader to the broader field of visible difference literature. It is hoped that this will provide the reader with an appropriate context to make sense of the project. The introduction will consider the challenges of living with a visible difference, adjustment processes and how infant/parent attachment may be influenced by a visible difference. Following this, the results of a systematic literature search related to parental adjustment to facial visible difference will be presented.

#### **3.1 Living with a visible difference**

The charity Changing Faces estimates that over 400,000 people in the UK alone have a visible difference (Changing Faces, 2001). In 2004, Rumsey & Harcourt conducted a literature review addressing the psycho-social implications of living with a visible difference and highlighted that it is a multi-

faceted and complex issue that involves social, physical, cultural and psycho-social factors. Thompson & Kent (2001) conducted a comprehensive literature review and concluded that individuals with a facial visible difference are subjected to social stigma based on their appearance and experience negative reactions from others, including stares and in some cases rude or intrusive comments that cause distress. Lansdown, Lloyd & Hunter (1991) describe a visible difference to be a life-long stressor which may, at times, exacerbate pressures associated with certain developmental stages.

Culture is thought to determine ideals of beauty and responses to visible difference and the extent of identification with cultural norms is a crucial factor in measuring how much pressure one feels to match up to cultural ideals (Fallon, 1990). Across cultures, birth defects, including facial visible difference or physical illness that results in visible differences result in significant social stigma (Shaw, 1981; Strauss, 1985; Brazil & Sargent, 1982 cited in Fallon, 1990).

Rumsey & Harcourt, (2004) describe how, within cultures, there are gender differences relevant to appearance and in relation to the pressures experienced by the individual to conform to the societal norms. In Western cultures the beauty and appearance ideals are thought to be shared through modelling within the family and more widely within society through the media. Therefore, it is considered that the culture that someone living with a visible difference experiences can impact on their psycho-social wellbeing (Rumsey & Harcourt, 2004).

### 3.2 Adjustment

Given the social significance of the face and the social prejudices towards visible difference, it is not surprising that people with visible differences experience difficulties in social encounters and the reactions of others. A number of factors are known to influence an individual's ability to adjust to living with a visible difference and these will now be discussed. This discussion will be underpinned by a model of adjustment.

### *3.2.1 Models of adjustment*

A model of adjustment was proposed by Wallander, Varni, Babini, Banis, DeHaan & Wilcox (1989) and has been used to conceptualise parental and family adjustment within the field of paediatric chronic health disorders (Nguyen, Pertini, & Kettler, 2013; Brown, Doepke & Kaslow, 1993; Malik & Koot, 2009; Lutz, Barakat, Smith-Whitley & Ohene-Frempong, 2004). This model captures a multitude of factors that are involved in family adjustment and this model is conceptualised as a risk-resistance framework. It is considered that this model provides a framework to explore the factors relating to adjusting to visible difference.

The model describes risk factors that include the type of illness, functional independence and psycho-social stressors while the resilience factors include intrapersonal factors, stress processing and social ecological factors (Wallander et al., 1989). It is suggested that when risk factors are high and resistance factors are low, there are likely to be greater difficulties in physical, emotional and psycho-social functioning and lower rates of adjustment (Brown, Doepke & Kaslow, 1993). Within the disease/disability parameters diagnosis, severity, disease duration, visibility and potential for impact on cognitive functioning are considered to play a role. Psycho-social stressors include daily stresses, as well as negative life events and socio-economic disadvantage. Illness related stressors can include medical appointments, treatment and medication regimes. Illness related stressors have been found to be a risk factor for poor adjustment and higher rates of emotional stress in mothers of children with chronic illness (Cadman, Rosenbaum, Boyle & Offord, 1991).

Resistance factors include personal factors such as personality, problem solving skills and temperament. Stress processing and coping strategies are important for positive adjustment (Wallander & Varni, 1992). Families use a range of coping strategies but planning and problem solving is seen to be one of the most adaptive strategies. Seeking out help, advice and social support are also positive coping strategies that are recognised in the chronic health literature to be used by parents (Azar & Solomon, 2001). Socio-ecological

factors such as having positive relationships within the family, high levels of support, greater higher marital satisfaction and a good social support network have been found to be features of positive adjustment to chronic health problems (Cadman et al., 1991) and it is considered that these factors may also be important for parents of children with visible differences.

### *3.2.2 Severity and visibility*

Visibility and severity are recognised to be potential risk factors by Wallander et al. (1989). It is considered that those people who experience greater distress as a result of their visible difference may find it harder to adjust to their visible difference. Although, some research has demonstrated that the severity of the visible difference is not associated with the severity of the distress or adjustment difficulties, instead, this research suggests that distress is mediated by psycho-social factors such as perceived reactions, pre-occupation with appearance and social skills (Ong, Clarke, White, Johnson, Withey & Butler, 2007). Research by Baker (1992) and Malt & Ugland (1987) also found no evidence of the relationship between severity and distress, suggesting that it is as likely for someone with a milder visible difference to experience as high levels of distress as someone with one of greater severity. However, it is important to note that severity is a subjective concept and can be measured in many ways, including percentage of skin covered, clinician ratings or self-reports (Robinson, 1997). It is hypothesised by MacGregor (1990) that this distress may be in part related to the uncertainty experienced by people with a milder visible difference in social situations because unlike severe visible difference, where reactions are likely to be quite predictable, for those with milder visible difference it may be harder to predict reactions from others leading to a greater feeling of anxiety or tension. However, visibility has been found to be a more significant predictor of adjustment. Williams and Griffiths (1991) found visibility to be a predictor of adjustment in a population of patients who had sustained burn injuries and Porter, Buef, Lerner & Norlund, (1990) found that patients with psoriasis were less well-adjusted compared to patients with vitiligo and concluded this may be due to patients

with psoriasis being unable to camouflage their condition as easily as patients with vitiligo.

Furthermore, it is considered that visible differences that occur congenitally, or pre-memory may be experienced differently compared to those who acquire their visible difference through injury or illness later in life as someone who acquires an illness may experience feelings of loss (Harris, 1997) compared to someone with a congenital difference as they have not experienced life without the visible difference.

### *3.2.3 Gender differences*

Considering visibility through a different lens, gender differences are thought to play a role in adjustment to living with visible difference however little research has been conducted to explore this phenomenon (Robinson, 1997). Rumsey (1997) suggests that high value is placed on physical attractiveness within Western society particularly for women, suggesting that women are more likely to have greater difficulty adjusting to visible difference. On the other hand, Lanigan and Cotterill (1989) suggest that men are less likely to use camouflage make-up to disguise their disfigurement which could mean that men find it harder to adjust to their visible difference, or conversely, it may result in them developing alternative ways of coping, which may result in better adjustment. Therefore, gender could be conceptualised as either a risk factor or a resistance factor depending on the individual as visibility, personal temperament and social factors are all related to one's ability to cope and adjust to visible difference.

### *3.2.4 Family adjustment and coping*

A family unit can provide a sense of belonging, nurturance, emotional support and protection of vulnerable members in addition to practical and financial support. As such, family factors are considered to be a resistance factor in Wallander et al. (1989) model. Research indicates that with appropriate support, people with congenital or acquired visible difference can become

resilient, adapt and learn to cope and that social support, lower appearance salience and greater personal resilience have been found to be associated with better family adjustment and coping (Prior & O'Dell, 2009; Moss, Lawson & White, 2014; Roberts & Shute, 2012). Research addressing the impact of childhood illness suggests that the child's family system and its subsystems, such as sibling relationships and marital relationships, are important to consider in terms of identifying family strengths and coping strategies (Kazak, Rourke & Crump, 2003). For children with a visible difference, family acceptance is recognised to be crucial in supporting their psycho-social development (Thompson & Kent, 2001). While people with less severe visible difference may have a greater ability to camouflage the difference, greater social support and more developed coping strategies were found to be key factors in positive adjustment and resilience, regardless of the severity of the visible difference in the review by Thompson & Kent (2001). Within the birthmark specific literature, Kalick et al. (1981) found lower levels of psychological morbidity in an individual were related to higher levels of family support for 82 people undergoing laser treatment for their Port Wine Stain birthmarks. Lansdown, Rumsey, Bradbury, Carr & Partridge (1997) suggest that family support and feeling at ease within a familiar circle of people who can see the 'real' person behind the disfigurement is important to adjustment and coping.

It is acknowledged that while partners, carers and families are likely to be the greatest source of support for the individual, they may face their own challenges managing the reactions and their feelings and distress about the disfigurement (Rumsey & Harcourt, 2004). For example, parents need time at birth to adjust to the visible difference (Bradbury & Hewison, 1994). Leary & Kowalski, (1995) suggest that children with a facial difference may experience a sense of stigma unintentionally from their parents if they engender an environment that gives appearance a high value within the family. For further discussion, please refer to Rumsey & Harcourt (2012). Given the important role the family has in supporting an individual, this will now be discussed further.

### 3.3 Parent/infant relationship

When a baby is born with an unexpected visible difference, parents may be met with emotions that they were not anticipating. Research suggests that parental reaction to the disfigurement may influence the parental attachment with the child and the child's later adjustment to living with a visible difference (Bradbury & Hewison, 1994; Rumsey & Harcourt, 2004). This will now be explored in more detail.

Bowlby (1969) proposed an ethological theory of attachment that described the bond and the affection that develops between parents or the caregivers and infants. Ainsworth, Blehar, Waters & Wall (1978) examined individual differences in attachment style and concluded that there are different attachment styles that are related to maternal behaviour. One of the key propositions of attachment theory is the internal working model. This model proposes that infants start building internal cognitive models of their environment including experiences of other people. It is suggested that a child does this to be able to anticipate and to make sense of the world and of other people. New information can be assimilated and accommodated by the existing models in order to elaborate them. For young children attachment figures are particularly important as they allow the infant to begin to build mental representations of their own worth based on the caregiver's availability and willingness to provide care and protection (Ainsworth et al., 1978). As children begin to learn, the quality of these relationships may have a profound effect on the child's understanding of themselves, others and social interactions not only in childhood but also into adulthood. Therefore, it is important to consider the impact a visible difference may have on the early bonding process.

Much of the literature referred to in this section documents the findings from research into the field of CL/P as this is most widely researched within the wider field of facial visible difference. Walters (1997) suggested that disfigurement may have a negative impact on the initial bonding process which Bradbury and Hewison (1994) suggest to be, in part, the result of the parent's grieving for the 'perfect' baby they were anticipating. One study by

Barden et al. (1989) suggested that parents of children with a visible difference can be less nurturing and more distant than parents of children without. Another study by Despars et al. (2011) found that mothers of children with CL/P more frequently had insecure parental internal working models of the child. They also found that mothers of babies with CL/P displayed higher post-traumatic stress disorder symptoms when compared to mothers in a control group. Interestingly, the mothers who reported higher levels of post-traumatic stress symptoms were found to be more emotionally involved with their babies compared to the mothers who reported lower post-traumatic-stress symptoms and notably, the severity of the CL/P was not found to have an effect on the attachment or the severity of the post-traumatic stress symptoms experienced by the mothers.

Other researchers have also demonstrated how parental adjustment to a visible difference can have a negative impact on early attachment as mothers may find it harder to bond with her child (Bradbury & Hewison, 1994; Campis, DeMaso & White-Twente, 1995). However, this finding is not universal. Research by Collet and Speltz (2007) found no overall difference in secure attachment between mothers and their babies with CL/P in comparison with controls. Collet & Speltz (2007) go on to suggest that babies with a CL/P have higher rates of secure attachment at an earlier age, with the most severe clefts demonstrating the highest rates. The authors conclude that this finding may relate to the mothers perceiving their child as more vulnerable and this may intensify their maternal responsiveness, which, in turn, results in stronger attachments. Bradbury & Hewison (1994) report that fierce maternal protectiveness such as this may also have the potential to become problematic and lead to separation difficulties. Given this contrasting literature, it is suggested that there may be other contextual factors that could account for the differences in attachment found in these studies. Rumsey and Stock (2013) suggest that attachment must be considered alongside family factors such as socioeconomic status, family attachment history, parental coping styles, parental mental well-being, beliefs about the cause of the disfigurement and feelings of responsibility and other child related factors including temperament and co-morbidity with other disorders.

### 3.4 Summary

In summary, living with a visible difference is a multi-faceted issue that involves psychological, social, physical and cultural factors. In addition, the societal and cultural climate relating to appearance presents a challenging and arguably less accepting environment for people living with facial visible difference. It is apparent that people with facial visible difference have to cope with challenges such as social stigma and experience negative reactions from others and this can impact on an individual's well-being. Family adjustment and coping have been identified to be important factors in the early stages of a child's life to ensure the child has the greatest opportunity for developing adaptive schemas, self-esteem and resilience although this area is under researched. However, it can be concluded that in order for the child's optimal adjustment, parental adjustment and coping is paramount. Therefore, it is pertinent to explore the literature that focuses on parental experiences of adjusting to their child's congenital facial visible difference.

## **4. Reviewing the literature on parental adjustment**

### 4.1 How the review was conducted

A search was conducted to find the lived experience of parents with children who have facial birthmarks and few studies were found. As papers that relate specifically to parental experiences of having a child born with a facial birthmark are extremely scarce, the search was extended to the broader category of facial visible difference.

Parental adjustment to CL/P is most widely researched, followed by CF conditions, although it is noted that these are different conditions and have different associated challenges, such as functional issues relating to feeding and speaking. As a second step, the inclusion criteria was loosened to include papers that involved participation from both parents and their children and measures of quality of life and family satisfaction in order to fully explore the literature relating to birthmarks.

Using the search terms shown in Table 1 multiple searches were conducted using MEDLINE, SCOPUS, psycINFO, CINAHL and Google Scholar databases.

Table 1.

*Key search terms*

AND	NOT
facial OR face  disfigurement OR difference OR birthmark OR cleft lip & palate OR craniofacial  birthmark OR nevus/neavus/nevi OR Port Wine Stain OR haemangioma OR strawberry/facial neoplasm/vascular birthmark/pigmented birthmark  Parent* OR maternal OR paternal OR mother OR father OR family OR families  Experience, adjust* OR coping OR psychology, psych* psycho-social OR quality of life	acquired injury, burn, trauma, accident, physical disfigurement, disability, facial palsy, amputation, facial or head or neck or throat cancer  java birthmarks, computer software fruit, wine, hepatic haemangioma, internal haemangioma, Sturge Webber Syndrome,

Papers were excluded if they were medically or surgically focussed as opposed to psychologically focussed. Papers were also excluded if they were primarily concerned with functional difficulties such as feeding. The reference lists of the remaining papers were then examined to see if any relevant studies had been missed. The Centre for Appearance Research was contacted for further recommendations or unpublished papers in the field. All of the papers included in the review were papers written by psychologists,

allied medical professionals and academic researchers in the field. For a diagrammatic representation of this process, please refer to appendix Ai.

In total, eleven papers have been included in the review. These papers are from the UK, USA, Australia and Europe. Seven studies endorse qualitative methods while two endorse quantitative methods and two use mix methods. Four studies focussed on adjustment to CL/P, one focussed on the experiences of families with a child born with a facial haemangioma and three addressed the challenges faced by families with a child born with a CFA. One study surveyed the parents and children with a haemangioma using a quality of life measure and another study researched family satisfaction in families in which a child with a PWS was undergoing laser treatment. For a summary of the articles included, please the table below.

Table 2.

*Summary of the articles included in the literature review.*

<b>Authors</b>	<b>Methodology</b>	<b>Participants</b>	<b>Key Points</b>	<b>Critique</b>
<b>Bradbury &amp; Hewison (1994) UK</b>	Attributional analysis is used to cluster semi-structured interviews into 6 themes and then the interviews were scored on a 3-point scale against the identified themes in order to generate a measure of parental adjustment which was then analysed statistically.	25 families of children with CL/P, 34 families of children with hand deficit.	Overall, the adjustment between the two groups did not differ significantly. Themes related to having a child born with a disfigurement were recognised, but were not associated with severity or type of disfigurement. Perceived family support was the only significant variable found to relate to parental adjustment.	Good sample size but little information about the analysis, methodological rigor or validity of the analysis in the article. Successfully generated a measure that can be used for both clinical and research purposes.
<b>Hoorweg, Grootenhuis &amp; van der</b>	Survey of child and parental quality of life.	201 parents of children with haemangiomas.	Majority of parents and children with the haemangioma were not negatively affected by it. Parents	Endorsed a questionnaire specific to haemangiomas. No data for treatment on

<b>Horst (2009)</b> <i>The Netherlands</i>			reported strong feeling of panic and disbelief during growth phase. More negative effects reported by parents when haemangioma was in a visible place.	complex haemangiomas. Potential sample bias due to the setting the study took place in.
<b>Johansson &amp; Ringsberg (2004)</b> <i>Sweden</i>	A phenomenographic approach was used in which guided interviews were coded into categories.	20 mothers and 12 fathers of children with CL/P.	Two categories emerged. 1. The birth as an unexpected event – the related shock, worry and support and mixed opinions of medical professions. 2. Reaction – mainly positive reactions experienced some neutral reactions that were interpreted as lack of interest. Staring or comments occurred, concerns about scars especially for girls, some concerns around potential for speech difficulties.	Good sample size for qualitative research, however little information about the process of analysis documented in article. Interviews lasted 30-45 minutes. Phenomenological approaches require homogenous samples therefore raising the question about using males and females in the sample and both unilateral and bilateral clefts and with the children ranging in age from 1 month-5 years.
<b>Klein, Pope, Getahun &amp; Thompson (2006)</b> <i>USA</i>	Used interviews and narratives were coded into categories.	9 mothers of children (aged 9-14) with CFA.	Concern and protectiveness of child, worries about teasing and exclusion because of visible difference and concern about the child's emotional well-being. Parenting strategies aimed to promote autonomy, social & emotional adjustment and pointing out the child's strengths.	Mothers recruited through a support group for people with CFA therefore sample bias and sample size are limitations. Participants from similar socio-economic back grounds and ethnicity groups. Questions used in interview were not included in article and cannot be critiqued. However, steps taken to ensure reliability of coding and categories through using multiple people in data analysis.
<b>Klein, Pope &amp; Tan (2010)</b>	Structured telephone Interviews.	9 fathers of children with CFA.	Fathers were generally optimistic about their child's future, happiness and success.	Novel study of father's experiences of having a child with CFA. Small

Parental ad. to birthmark

<b>USA</b>			A smaller number expressed concerns about how others' reactions may affect their child's confidence or self-esteem.	sample size and little socio-economic, cultural or marital diversity in the sample. Interviews used a structured approach and only lasted 15-30 minutes therefore may not have fully captured father's full experiences. However, interview questions were developed based on Klein et al. 2006 research.
<b>Koot, De Waard-van der Spek, Peer, Mulder &amp; Oranje (2008) The Netherlands</b>	Children were physically assessed and interviewed and mothers and teachers completed questionnaires.	29 children with giant congenital melanocytic nevi and their parents and teachers.	Social problems were reported for 30% of the children and behavioural/emotional problems for 26.9%. There was no correlation between the visibility of the nevus, treatment or child age and psychological problems. Mother's reported a considerable psycho-social burden on themselves because of their child's birthmark.	Mixed methods approach allowing for a rounded approach to the research although questionnaires were general measure of child psycho-social wellbeing and not specific to adjustment issues related to having a birthmark or the associated treatment.
<b>Miller, Pit-ten Cate, Watson &amp; Geronemus. , (1999) USA</b>	Survey of stress and family satisfaction in families with a child with a PWS.	46 parents of 24 children with PWS receiving laser treatment.	Parents scored in the average range on the stress and family satisfaction measures when compared to a control group; five parents (11%) scored in the clinical range for stress. The findings indicate that although the majority of parents were in the average range for stress, some are more affected. Factors associated with lower levels of stress include having younger children at the time of treatment, more family cohesion, fewer parental concerns, and greater satisfaction in communication with medical professionals.	Small sample size, lacking in both socio-economic and marital diversity. No comparison group. Sample only includes parents whose child underwent treatment so potential sample bias. Questionnaires used are general stress and satisfaction measures that are not specific to PWS populations therefore provide an overall measure of wellbeing as opposed to specific PWS treatment impact.
<b>Nelson Kirk, Caress &amp; Glenny, (2011) UK</b>	In depth interviews analysed using a constructivist	Parents of 27 children of varying ages (0-15 years) with	Parents experience a range of, sometimes conflicting, emotions about their child's impairment. They worry about treatment and	Good sample size for grounded theory research and rigorous in methodology. Conducted

Parental ad. to birthmark

	grounded theory approach.	CL/P.	the potential for social stigma such as exclusion.	interviews with parents individually and some in dyads so data may be qualitatively different, although potentially more comprehensive. The voice of parents who did not want treatment for CL/P may be under represented in the sample.
<b>Stock &amp; Rumsey (2015) UK</b>	Individual qualitative telephone interviews.	15 fathers of children born with CL/P.	4 over-arching themes emerged: variations in care and support; appraisals of the cleft; perceptions of treatment; and looking back and moving forwards. Fathers reported experiences comparable to mothers with additional needs for greater information and further support.	Novel project exploring the father's perspective of CL/P. Appropriate sample size (until saturation was achieved) although fathers were self-selecting, introducing bias. Steps taken to ensure reliability of main themes.
<b>Tanner, Dechert &amp; Frieden, (1998) USA</b>	In depth interviews and qualitative analysis.	Parents of 25 children with facial haemangioma.	Parental reactions of grief and loss, particularly in the growing phase. Half of the participants expressed dissatisfaction with medical system. Social and family support were important factors in coping.	Good sample size, although different methods of collection (face to face and telephone interviews). Used ethnographic approach. Rigor or checks for validity are not considered in article.
<b>Williams 3<sup>rd</sup> Hochman, Rodgers, Brockbank, Shannon &amp; Lam, (2003) USA</b>	Telephone interviews (conducted retrospectively).	39 parents of children with haemangiomas.	The study found that haemangiomas have a negative effect on the child's family, with considerable worry caused, in part by negative public reactions or comments.	The authors used a structured interview approach using a questionnaire, limiting the data. Analysed using only descriptive statistics. Retrospective data. However, few studies have addressed the psycho-social impact of haemangiomas so despite its flaws it is a start.

The literature review has been structured according to common themes across the literature. These papers will now be discussed in relation to the themes before being critiqued holding in mind the guidelines for qualitative paper critique outlined by Elliot, Fisher & Rennie (1999) and the guidelines for critiquing quantitative research outlined by Barker, Pistrang & Elliot (2012).

#### 4.2 Parents' initial reaction

The initial emotions experienced by parents upon realising their baby has a visible difference are reported to be mixed; those of happiness for having a child, but also shock, loss, grief and worry (Tanner, Dechert & Frieden, 1998; Bradbury & Hewison, 1994; Johansson & Ringsberg, 2004; Hoornweg, Grootenhuis & van der Horst, 2009; Nelson Kirk, Caress & Glenny, 2011). These initial reactions appear to be consistent across different types of visible difference. Feelings of loss, in relation to the anticipated perfect baby was a common theme along with the associated concerns about the prognosis for the child's health and long term wellbeing. These feelings are noted to be present for parents of children with haemangiomas, despite the generally benign nature of haemangiomas and the prognosis for eventual involution (Tanner et al., 1998; Hoornweg et al., 2009).

Tanner et al. (1998) conducted interviews with 25 parents of children aged five months to eight years old recruited from a dermatology practice. Interviews took place either face to face or by telephone. The authors report that they analysed the data using 'open coding' but do not offer any further information about the qualitative approach they endorsed for analysis. They reported four main themes of parental emotion and adaption, including the initial experience of shock, experiences with public reactions, issues related to parent-child interaction and expressed satisfaction and dissatisfaction with medical care and concluded that medical professionals should be appropriately trained to offer support and guidance to parents that is specific to the challenges parents face looking after a child with a haemangioma. Hoornweg et al. (2009) endorsed a quantitative approach and used a reliable and validated health related quality of life measure and a haemangioma

specific measure designed for the study. They recruited 201 parents of children aged between one year and 15 years old and found that, according to the quality of life measure, the majority of parents and children were not negatively affected by the haemangioma. The results of the haemangioma specific questionnaire indicated that parents experienced high levels of shock and distress during the growth phase of the haemangioma, which is in keeping with the findings from Tanner et al. (1998) study. A second conclusion was that having a visible haemangioma or one with a complicated medical course may have an impact on the child's later psycho-social wellbeing.

Bradbury and Hewison (1994) compared the needs of families with a child with a CL/P with families who had a child with a hand deformity. 25 families with children with a cleft lip and 34 families with a child with a hand deformity took part. They found that the only significant difference between parental adjustment to both conditions was perceived family support and that the duration of the initial shock varied and this variation appeared to be related to parents' personal coping factors rather than the type or severity of the disfigurement. Parental coping was a theme in many papers and will be discussed later.

Nelson et al. (2011) conducted in depth interviews with 27 parents of children with CL/P of varying ages (0-15 years), and reported that they achieved theoretical saturation of their categories. They also found that the parents in their sample struggled with the initial shock and grief and sadness on finding out about the CL/P and facing medical challenges associated with surgery. Furthermore, they found that parents began to express concerns about their child's future and how living with a CL/P might affect their psychosocial development. The authors discuss the need for greater assessment of parental social and emotional well-being in CL/P services at this crucial stage.

A further issue that was described in a number of studies is the reactions and support from medical professionals. Research indicates that parents were frequently dissatisfied with the support and advice offered by medical professionals and felt that they lacked knowledge about how to handle the

situation when a baby is born with a visible difference (Johansson & Ringsberg, 2004; Nelson et al., 2012; Tanner et al., 1998; Miller, Pit-ten Cate, Watson & Geronemus, 1999). Johansson and Ringsberg (2004) recruited 20 families from a hospital setting and interviewed 20 mothers and 12 fathers of children with unilateral or bilateral CL/P. The interviews focussed on how parents perceived social and mental support from friends, family and professionals and the analysis endorsed a phenomenographic approach. Two main categories emerged, firstly, the unexpected event of having a child with CL/P and secondly the reactions from family, friends and the public. The authors conclude that some staff parents encountered had a low level of knowledge about CL/P and subsequently were unable to fully support parents.

#### 4.3 Parental coping

In addition to themes of grief, loss, the related tearfulness and sadness experienced by the parents in the initial stages, Bradbury & Hewison (1994) documented early parental coping strategies of avoidance, denial and anger. Furthermore, a need for meaning making was found to be important for parents to cope including understanding the cause of the visible difference. Combined in this meaning making process, some mothers expressed feelings of guilt, feeling unsure if it was caused by something they did during pregnancy (Bradbury & Hewison, 1994). Parental coping was also found to be a prevalent factor in adjustment in the study by Koot, De Waard-van der Spek, Peer, Mulder & Oranje (2008) who interviewed 29 children with giant congenital melanocytic nevi and their parents and teachers completed questionnaires about the behavioural and emotional problems and their own coping. They found that there was no correlation between the visibility of the nevus, treatment or the child's age and later psychological problems. However, they found that the mothers reported a considerable psycho-social burden on themselves because of their child's birthmark. In light of Bradbury and Hewison (1994) findings, one possible explanation for the burden reported by mothers in Koot et al. (1998) study is that it may be related to the

amount of support the mothers had access to, although this is not explored in the study.

There are conflicting views in the literature about severity and visibility impacting on parental adjustment. Bradbury & Hewison (1994) suggests that parental adjustment is not related to severity or visibility of the lesion, although Hoornweg et al. (2009) reported greater negative effects on parents when the haemangioma was in a visible location. Williams 3<sup>rd</sup>, Hochman, Rodgers, Brockbank, Shannon & Lam, (2003) conducted retrospective telephone interviews with 39 parents of children with haemangiomas. They endorsed a structured interview approach using a structured questionnaire and analysed the data using descriptive statistics. The study found that haemangiomas had a negative effect on the child's family as they caused considerable worry for the parents that was in part, caused by negative public reactions or comments.

It is apparent that parents have different needs that must be met to support them to cope and it is considered that the parent's personal view on the importance of appearance may be a factor, although this is not discussed or explored in any of the present studies being discussed. Miller et al. (1999) conducted a cross sectional survey to assess the parenting stress, family satisfaction and parental concerns to determine predictors of stress in parents of children with port wine stains. 46 parents participated in the study and the authors concluded that factors that contribute to lower levels of parental stress include having more family cohesion, fewer parental concerns, and greater satisfaction in communication with medical professionals. Social and family support have also been found to be significant factors in managing the adjustment process after having a child with a visible difference (Miller et al., 1999; Tanner et al., 1998).

Klein et al (2010) interviewed nine fathers of children with CFA using telephone interviews and an interview schedule developed from the interviews previously conducted with mothers. An earlier study by Klein et al. (2006) recruited nine mothers of children with CL/P to be interviewed using face-to-face in-depth semi-structured interviews. Following these interviews, mothers

were asked for permission to contact the fathers. The telephone interviews with the fathers included both open and closed ended questions and the closed ended answers corresponded to a rating scale and the open ended questions were recorded and sorted into categories. The interviews were 15-30 minutes in length and once they had analysed the data they identified a number of further support and information needs of fathers to promote coping including wanting clearer information about a range of issues. These included aetiology, such as the genetic and environmental factors involved; treatment options; strategies for coping with public reactions; how to talk to the child about the difference. Additionally, they reported requests to meet other families in the same situation as well as families who are further along the treatment journey. Stock and Rumsey (2015) reported similar findings including greater information needs for fathers in their study which endorsed telephone interviews with 15 white British fathers of children of any age with any type of CL/P from a wide geographical spread of the UK and analysed the data using thematic analysis. The need for greater support with how to manage public reactions was common across a number of studies in the literature (Williams 3<sup>rd</sup> et al., 2003; Klein et al., 2006; Klein et al., 2010; Stock & Rumsey, 2015).

Fathers in the Klein et al. (2010) study took an active approach to coping through supporting their children who had CFA by organizing activities to encourage their children to build social relationships. Comparing this with the study by Klein et al. (2006) in which nine mothers of children with CFA were interviewed, the parents in both studies reported the pleasure of parenting, using positive parenting approaches in addition to the challenges that faced their children and their worries and concern, particularly in terms of social situations and peer relationships. One difference between the parental approaches was that the mothers appeared to be more actively involved in giving advice to their children about how to manage social relationships, comments and public reaction, whereas the fathers seemed more focussed on matters outside of the family as they discussed issues including the likely occupational success of the child and acceptance within wider society. Considering the requests fathers made in the Klein et al. (2010) study and the

Stock & Rumsey, (2015) study for greater support with how to manage social difficulties, it may be that fathers, perhaps, felt unsure about how to advise their children on the issues of managing peer relationships and social comments.

#### 4.4 Managing the reactions of others

##### *4.4.1 Reactions of others at birth*

Nelson et al. (2011) described how parents had to contend with the reactions of family and friends who in the initial stages appeared unsure of how to act. Johansson & Ringsberg (2004) describe the majority of parents to have experienced support and positive comments however they describe the sadness parents experience when family and friends stayed away because of the child's CL/P. Furthermore, they describe the differences in medical professionals and discuss the interest shown by some as an invasion of privacy. For some fathers the idea of telling family and friends was a worrying prospect and although for many they responded kindly, some fathers reported distress in members of their extended family (Stock & Rumsey, 2015).

##### *4.4.2 Wider social reactions*

Some parents were reported to experience increased stress resulting from difficulties or anticipated difficulties with public reactions such as staring, insensitive remarks, whispering and questions about possible abuse (Tanner et al., 1998; Bradbury & Hewison, 1994; Williams 3<sup>rd</sup> et al., 2003; Nelson et al., 2011). These experiences are reported by the authors to have generated feelings of sadness, helplessness, anxiety and anger in parents. Bradbury & Hewison go on to discuss the measures parents felt compelled to take to protect their child, such as guarding the pram and anticipating responses and discuss this in terms of the parent trying to take control of the situation. Nelson et al. (2011) discuss mothers' experiences of social stigmatization and how these led them to make greater attempts to camouflage their baby's CL/P, while others were concerned about the child developing a complex about their visible difference and so would not make attempts to conceal it (Bradbury &

Hewison, 1994). Clearly there are differing experiences of parents' social reactions in the research. Johansson & Ringsberg (2004) reported mainly positive reactions from the public towards children with CL/P and some neutral reactions that were interpreted by parents as lack of interest. However, the parents in the study did report some occasional staring or comments occurring and expressed concerns about scars especially for girls, along with concerns around potential for speech difficulties.

Nelson et al. (2011) found that parents experienced negative remarks and stigmatizing reactions, particularly during pre-school years and that this led to the social exclusion of the family unit. Parents also expressed concerns for transitions that their baby will face such as starting nursery or school (Nelson et al., 2012; Hoornweg et al., 2009). Furthermore, parents expressed concern about their child's peer relationships and the likelihood they may experience teasing or bullying (Nelson et al., 2012; Johansson & Ringsberg, 2004; Klein et al., 2006; Bradbury & Hewison 1994; Hoornweg et al., 2009).

#### 4.5 Treatment and medical intervention

Treatment and medical intervention appears to be another stressor for parents. The need for clear advice about the treatment options and good communication with professionals is evident (Stock & Rumsey, 2015; Miller et al., 1999; Tanner et al., 1998; Bradbury & Hewison, 1994). Miller et al. (1999) discuss how information sharing with parents is not always satisfactory and can increase parental stress and highlight the importance of comprehensive information that is sensitive to the psychological needs of the parents and their children.

Moreover, the treatment itself can generate mixed emotions for parents. Bradbury & Hewison (1994) describe contrasting viewpoints about CL/P surgery. Nelson et al. (2011) report parents' conflicting feelings about 'normalizing' their child's appearance as this required putting the child through a number of surgeries. This is presented as a dilemma for parents, given their wish to have a child who is the 'same' as other children, but having a strong

urge to protect the child from harm. Additionally, the risks associated with anaesthesia were a concern for some parents (Johansson & Ringsberg, 2004; Nelson et al., 2011). However, other research indicates that for some parents, this decision is less difficult. One mother felt “she’s a normal toddler now whereas before she was a freak” (p.258, Bradbury & Hewison, 1994) and was reported to have withheld attachment until after the corrective surgery. In stark contrast to a different mother who felt sad for the loss of the visible difference “I miss it you know, because when she used to smile one half would curl over” (Bradbury & Hewison, 1994, p. 258).

#### 4.7 Critique of the literature

##### *4.7.1 Critique of the literature in this review*

The validity of these studies was assessed using the framework outlined by Elliot, Fisher & Rennie (1999). This framework has seven guidelines which are:

1. Owning one’s perspective
2. Situating the sample
3. Grounding in examples
4. Providing credibility checks
5. Coherence
6. Accomplishing general vs. specific research tasks
7. Resonating with readers

One of the main critiques of the studies reviewed, is possible implications of accessing participants through support groups or medical settings or in the case of Stock and Rumsey (2015) through the participants being self-selecting. This may result in the voice of those who have not accessed support being under represented in the literature. It may be that this underrepresented group are well-adjusted to the visible difference and do not feel the need to access support, however, on the other hand, it may also signify a group of people who are struggling to cope or are not aware of, or able to access support and advice. All of the studies reported the ethical

considerations they made before they conducted their studies and reported that they abided by guidelines of ethical practice as they conducted their research.

The qualitative research studies included in this review had appropriate sample sizes for qualitative research given that an appropriate sample size can be considered to be determined by the point of saturation, when no new information is being obtained (Stock & Rumsey, 2015). However, the majority of the studies provided little information about the process of analysis and did not publish an audit trail. Johansson & Ringsberg (2004) and Klein et al. (2010) reported the length of their interviews which appear to be shorter than other studies endorsing the same or similar methodologies, raising the question about whether the researchers had comprehensive data with enough breadth and depth to fully support and generate interpretation. However, Klein et al. (2010) report using both open and closed questions that generated both quantitative and qualitative data as a method to enable the interviews to remain shorter, while still managing to get breadth in their data. They also report making credibility checks by using two authors to categorise their qualitative data.

Thinking specifically about the requirements for qualitative analysis, transparency in relation to the researcher's position and demonstrating reflexivity is paramount. Looking at these studies, it is evident that Johansson & Ringsberg (2004) do not demonstrate reflexivity; they do not state their epistemological position or explain about researcher characteristics or potential researcher biases that may have affected the analysis. Johansson and Ringsberg (2004) endorsed an Interpretative Phenomenological Analysis (IPA) approach to their data analysis. In line with this method, they purposely selected participants, however their sample aimed to reflect diversity of experience, for example, they selected mothers and fathers of children aged one month – five years with unilateral and bilateral clefts therefore, it could be argued, that this is not a truly homogenous sample. Therefore, it is suggested that IPA may not have been the most appropriate choice of data analysis.

In contrast, Nelson et al. (2011) used a Constructivist Grounded Theory approach and they have clearly stated their epistemological stance and visibly demonstrate their analysis process and reflexivity.

Johansson and Ringsberg (2004) conducted interviews with parents individually and together without a methodological justification for these choices and Nelson et al. (2011) also conducted a combination of individual and dyad interviews and justified this in terms of meeting the needs of the participants so data may be qualitatively different. Neither of these studies show evidence of triangulation, for example using another data source or method of data collection which would have demonstrated rigor in the study. However, all of the studies that endorsed a qualitative method reported using more than one person in the analysis process such as employing multiple people to code the data, which illustrates rigor in the analysis.

Moving on to considering the quantitative research, the guidelines for quantitative research by Barker, Pistrang & Elliot (2012) were used to critique this literature.

Miller et al. (1999) used a modest sample size (46 parents), which appears to be lacking in both socio-economic and marital diversity. The sample only includes parents whose child underwent treatment and did not endorse a comparison group. They used a survey approach which utilised self-report questionnaires. Three of the questionnaires used (Parenting Stress Index, Family Satisfaction Scale, Child Behaviour Checklist) have been demonstrated to be valid and reliable measures, while the Parental Concerns Questionnaire that was used was specifically designed for their study and they do not offer any information about the validity and reliability of this measure. Williams 3<sup>rd</sup> et al. (2003) used a structured interview approach using a questionnaire designed for the study that limited the depth of the data collected and they analysed the data using only descriptive statistics. However, the authors recognise the limitations of their study and highlight that few studies have addressed the psycho-social impact of haemangiomas.

Unlike the majority of studies, Hoornweg et al. (2009) endorsed a questionnaire that was specific to haemangiomas as well as a health related quality of life measure that they state has been validated and is a reliable measure. The authors do not provide information about the reliability or validity of the haemangioma specific measure that they designed, although they state that they engaged service users and medical professionals in the development of the measure. For the most part research in this field utilises measures that are not specific to the identified population and therefore provide an overall measure of the child's psycho-social wellbeing, rather than a specific measure of the impact the visible difference or treatment has on the child. Furthermore, all of the quantitative studies included in this review have not demonstrated consideration of statistical power calculations when determining their sample sizes although Hoornweg et al (2009) achieved a large sample size of 201. Given the rarity of research in this field, it may be that many studies in this field are often hindered by time or money constraints or recruitment difficulties making larger sample sizes harder to achieve.

#### *4.7.2 Critique of the wider field of visible difference literature*

It is evident that the quality of the research in this field to date is variable. There is a clear need for larger scale, rigorous and longitudinal studies to measure the adjustment process not only in the initial stages of the child's life but throughout childhood and adolescence and the need to examine parents' experiences across the different stages of their children's lives and at times of natural transition. From the review of the literature conducted, the complex and multifaceted experience of having a child with a visible difference is evident. It is considered that quantitative methodological approaches are not sensitive to the complex emotional and psychological factors involved and so lose the personal meaning of living with a visible difference (Rumsey & Harcourt, 2005) highlighting the need for greater qualitative research in this field.

#### 4.8 Summary of the findings

Despite a growing literature, to date, little qualitative research has examined parents' perspectives in-depth, particularly in relation to birthmarks other than haemangiomas. The research discussed appears to indicate that parents go on a personal journey after the birth of their child that is helped or hindered by certain factors. Factors including inter-personal support, accurate and timely medical advice, personal resilience, problems solving skills and coping strategies appear to play a role in positive parental adjustment. In contrast, factors such as experiencing lower social acceptance, lower levels of personal support and greater social stigma and intrusive remarks appear to contribute to poorer overall adjustment. Furthermore, treatment options appear to be a difficult dilemma for parents and one which requires parents to have access to all of the information about the risks and benefits to be able to make an informed decision in their child's best interest. A key finding of this literature is the apparent scarcity of psychological input within these teams and the need for emotional and psycho-social support for parents.

It is considered that often surgery in CL/P and CFA presents a different set of challenges for parents than those of a child with a birthmark. For CL/P and CFA, the majority of cases surgery is required for functional reasons such as feeding and speaking. In this way, it could be argued that surgery is necessary firstly for the child's physical wellbeing and secondly for psycho-social reasons and this may affect the relationship the parent has with the treatment process. Whereas for the most part, surgery on birthmarks is elective and parents make the decision based on the best advice of the medical professionals and their personal opinions about their child's future. This distinction has not been made explicit in the articles discussed, although it is felt that this requires further research, beyond the scope of this project, to explore if parents do have differing needs related to the decision making process.

Only five studies were sourced which included some aspect of parental adjustment and coping following the birth of a child with a birthmark. Of these five studies, only two endorsed a qualitative approach and both of these

studies were specific to haemangiomas. The findings of these studies are quite contradictory, suggesting that further research is necessary to gain greater understanding in this area. Furthermore, the majority of studies have recruited parents who have children aged from birth through to their teenage years. While this offers insight for clinical professionals about the needs of the children and parents as the child grows up, research has yet to specifically explore the nuances of this crucial time after the birth of the child.

#### 4.9 Rationale for the project

Given how critical a child's early development and attachment relationships are acknowledged to be within wider clinical psychology, when coupled with the additional challenges specific to individuals born with a facial birthmark and their families, childhood is an even more critical time. It is important to understand the challenges faced by parents whose children have a facial birthmark in order to be able to understand how, as professionals, we can support parents appropriately so they can, in turn, support their children. Furthermore, given the appearance conscious society we live in, understanding the challenges facing families in adjusting to facial birthmarks is crucial for providing greater awareness and support for parents and for greater development of resources for families, psychologists and medical clinicians such as midwives, health visitors, dermatologists and plastic surgeons. Research has looked at particular factors affecting adjustment, but until now, no study has brought these together to think about the process of adjustment. This research will be aiming to understand the processes involved in the early stages of parental adjustment to having a child with a birthmark.

#### 4.10 Research objective

This study does not set out to test a current theory or to prove or disprove hypotheses. Instead, this project aims to explore the adjustment process of parents following the birth of their baby with a facial birthmark in order to better understand the factors that are involved in the process of adjustment

and coping. This will be achieved through the generation of data about the experiences of parents with first-hand knowledge of this subject.

#### 4.11 Research questions

Given this rationale, the following research questions will be explored.

1. How do parents adjust after the birth of a child with a facial birthmark?
2. What are the factors that are involved in this process of adjustment and coping?

### **5. Methodology**

#### 5.1 Design

An exploratory, non-experimental method was endorsed for this study as very little previous research has been conducted in this field meaning that there are no existing theories or hypotheses available to test. Using a qualitative method allowed for the subjective lived experiences of having a child born with a facial birthmark to be explored, enabling greater depth of understanding about the meanings the parents gave to their experiences. This was achieved using interviews and using constructivist Grounded Theory (GT) for the analysis.

##### *5.1.1 Brief overview of Constructivist Grounded Theory*

Constructivist GT uses the basic GT strategies such as coding, memoing and theoretical sampling and takes an iterative approach to data analysis. It differs from the earlier versions of GT as it moves away from the positivist assumptions in Glaser's and Strauss and Corbin's earlier versions (Charmaz, 2014). Charmaz (2014) argues that the constructivist approach answers many of the criticisms of the earlier approaches such as fragmenting the participant's story and relying on the researcher holding an authoritarian stance. Instead, she argues that it highlights the flexibility of the approach and

holds the stance that social reality is multiple, involves processes and is constructed. Furthermore, she acknowledges the researcher's position, perspectives and interactions as inherent in the research. In this way, the research can be viewed as constructed rather than discovered but acknowledges that it occurs under specific circumstances of which we may not be aware and may not be our choosing (Charmaz, 2014).

The data collection and data analysis occur and evolve simultaneously, allowing for concepts and categories to emerge from the data through constant comparative analysis (Charmaz, 2014). This iterative process is a corner stone of the GT method. Throughout each step of GT, the researcher is able return to the data and review the data allowing for direction in questioning and selective sampling in order to gain greater understanding of the emerging concepts. Through this flexible process emerging theories are derived directly from the data and can then be followed up (Charmaz, 2014).

### *5.1.2 Using interviews*

Hiller & DiLuzio (2004) believe that employing interviews as a method of data collection allows for an activist perspective and gives a voice to marginalised groups and unheard voices. It was considered that interviewing parents would give them the opportunity to talk in depth about their experiences. The interview process is considered by Charmaz (2014) to be a directed conversation. The use of this approach enables the researcher to build a rapport between themselves and the participant and opens up the opportunity to access the interviewee's true feelings and own interpretations of their experiences of the phenomena (Charmaz, 2014). The primary aim of conducting interviews using an interview guide as opposed to a semi-structured interview schedule was to allow for participants to share their understanding of their experience (McCann & Clark, 2003) in their own words, whilst at the same time being flexible enough to adapt to follow issues that emerged during the conversations (Charmaz, 2014). The interview guide for this study is included in appendix B.

### *5.1.3 Grounding the method in my epistemological position*

The research method was selected based on the suitability for answering the research question. However, to ensure a strong and appropriate research design, it was important to choose a methodological approach that was congruent with my beliefs about the nature of reality. Consciously considering my epistemological position illuminated the methodological option available. Taking a social constructionist approach determines the way that I understand and interpret the world, learn about it and make sense of it (Lincoln & Guba, 2005). I agree with the notion that there are different coexisting interpretations of any phenomenon and that each interpretation is equally as important. In line with this, a constructivist approach to GT was selected (Charmaz, 2014). It is important to note that Charmaz (2014) uses the terms social constructionism and constructivism interchangeably and that they are subsumed under the generic term 'constructivism'. Given that a constructivist GT approach that openly groups these terms together has been endorsed it is important to clarify the difference between these terms. Constructivism suggests that every person constructs an internal representation of the world based on their experiences and is interested in this cognitive process, while social constructionism has a social focus rather than an individual focus (Young & Colin, 2004).

I acknowledged myself as having a role within this process as a co-creator of knowledge and I recognised the need to remain reflexive throughout the process given my position as someone with first-hand experience. I accept that this will have shaped the GT model emerging from this research (Pigeon & Henwood, 2004). I also accept within the constructivist approach to GT that prior perspectives and existing literature could have a role in guiding the research giving rise to sensitising concepts that provided possible avenues which may be explored (Charmaz, 2014).

### *5.1.4 Consideration of possible methods*

Before a constructivist Grounded Theory (GT) approach was selected other qualitative approaches were considered including Interpretative

Phenomenological Analysis (IPA) and Narrative Analysis (NA). I will now briefly contrast these methods and explain why GT was selected.

A NA approach involves exploring people's use of storytelling, personal accounts of events and the process of telling those stories (Sarbin, 1986). It places emphasis on the discourses used and focuses on the language people use in their descriptions. These narratives are an individual's search for meaning in their lived experience (Bruner, 2002). It is considered that this search for meaning is also a cornerstone of phenomenological methods of enquiry such as IPA and both of these approaches are consistent with a social constructionist epistemology as they place significance on the search for meaning to make sense of our lived experiences. IPA was developed by Smith, Flowers & Osbourne (1997) to enable researchers to explore the subjective experiences of participants. IPA is based on the proposition that individuals ascribe meaning to events and it is this meaning that is of central concern. However, it was felt that a key to answering the research questions in this study required an approach which enabled the process of adjustment to be explored, rather than the meaning of having a child born with a facial birthmark. As such GT was selected for this research project.

## 5.2 Data collection

### *5.2.1 The Participants*

14 participants took part in this study. This was made up of six couples and one mother (the father was unable to participate). The participants were all parents of a child aged six months to two years old who had a facial birthmark. A Clinical Nurse Specialist was also recruited to participate in the study. The participants were all White British living within England and Wales and came from a range of socio-economic backgrounds. The age range of the participants was 23-45 years old. The child of interest was the first born child in three families, second born in two families and fifth born in one family.

Table 2.

*A summary of the parents' demographic information*

Participant*	Age	Parents job	Geographical location	Type of birthmark	Gender of the child	Position of the child in the family	Age of the child
<b>Sally</b>	42	Finance	South East	PWS	Male	3 <sup>rd</sup> born	18mths
<b>James</b>	45	Doctor	South East	PWS	Male	3 <sup>rd</sup> born	18mths
<b>Hannah</b>	36	Finance	South East	Haemangioma	Female	2 <sup>nd</sup> born	2yrs
<b>Harry</b>	36	Managerial	South East	Haemangioma	Female	2 <sup>nd</sup> born	2yrs
<b>Anna</b>	35	Teacher	Wales	PWS	Female	1 <sup>st</sup> born	2yrs
<b>Jack</b>	35	Stay at home dad	Wales	PWS	Female	1 <sup>st</sup> born	2yrs
<b>Sarah</b>	34	Stay at home mum	North	Sebaceous Neavus	Male	5 <sup>th</sup> born	8mths
<b>Penny</b>	23	Teaching Assistant	Midlands	PWS	Male	1 <sup>st</sup> born	7mths
<b>Tom</b>	33	Electrician	Midlands	PWS	Male	1 <sup>st</sup> born	7mths
<b>Kate</b>	36	Teacher	South West	Haemangioma	Female	1 <sup>st</sup> born	2yrs
<b>Charlie</b>	36	Teacher	South West	Haemangioma	Female	1 <sup>st</sup> born	2yrs
<b>Amy</b>	36	Business Analyst	South East	Haemangioma	Female	1 <sup>st</sup> born	6mths
<b>George</b>	36	Computer Programmer	South East	Haemangioma	Female	1 <sup>st</sup> born	6mths

\*Pseudonyms are being used to maintain participant's confidentiality

### 5.2.2 Recruitment

An initial advert was posted on the Birthmark Support Group requesting participants for the study. Initially purposive sampling was used to recruit the first participants and then, in line with taking a theoretical sampling approach, additional adverts were posted at each stage of the study defining the criteria for involvement at that stage in the study. Theoretical sampling was then used as it allowed for the further development of the emerging categories.

Participants were not randomly selected, they were selected once a tentative theoretical category had emerged in order to further define the properties of the category (Charmaz, 2014). As such, the criteria for selection were shaped

by the memos and the codes emerging in the data. It was important not to only select participants who had experiences that supported the emerging theory and so negative cases were also sought allowing for a better understanding of the emerging theory.

### *5.2.3 Materials*

A flexible interview guide was adapted over the course of the interviews and a voice recorder was employed to record the interviews. A notepad was used to keep field notes throughout the data collection period.

### *5.2.4 Developing an interview guide*

An interview guide was developed (appendix B). In line with researchers such as Charmaz (2014) who believe in offering an insider perspective, my personal experience, consultation with parents with lived experience and knowledge of the literature was incorporated when thinking about the interview questions in order to give rise to sensitising concepts (Charmaz, 2014). Within the questions, particular emphasis was placed on understanding the process of adjustment to the facial disfigurement.

It was important to confirm the appropriateness and suitability of the interview guide and ensure that the questions would not cause offense or upset the participants, while ensuring that the questions fully covered the topics of interest and were sufficiently wide so as to allow the interview to be driven by participants. Furthermore, it was important to make sure that the questions were clear and the participants would understand the questions they were being asked. As such, the interview guide was piloted with my parents as they have lived experience of raising a child with a facial difference.

It was also the intention to notice and explore new issues, brought up by the interviewees. The interview guide was then edited in light of topics raised in interviews and extended during the course of the interviews in order to gain a deeper understanding of concepts that were emerging in the data, aiming to saturate the categories (Charmaz, 2014). In order to maintain reflexivity

throughout the process note taking and memoing were used. In order to maintain rapport with the participants, any potentially difficult or upsetting issues that were raised in interviews were gently re-introduced in later interviews with the explanation that it was an issue that other parents had spoken about, before asking them to reflect on whether this had any personal meaning for them. The emerging concepts also guided the recruitment of the next participants as theoretical sampling was endorsed in this study.

#### *5.2.5 Considerations for the interview*

Interviews were considered to be the best approach to gather the data from parents and were in line with the GT approach. Although using focus groups or written accounts such as from online forums were also considered as sources for data collection, it was felt that focus groups might not allow for the individuals to tell their stories as openly as in a 1:1 interview situation. Furthermore, it would put greater burden on participants to travel to the location of the focus group which might have reduced participation. The use of online forum data was a second option for sourcing data, however it was felt that this would be less effective than interviews as it removes the ability to ask probing questions and to get beneath the surface of experiences to ensure rich data.

It was hoped that the parents participating in the study would view the interviews as a positive experience and a chance to be heard and that a good rapport would be developed (Hiller and DiLuzio, 2004). Charmaz's (2014) recommendations for high quality interviews were considered to ensure the quality of the data. For example, the researcher aimed to collect adequate background data and to probe into the parents' experiences and elicit a range of descriptions of their actions and the consequences of these actions and where possible to gain multiple views of them.

Prior to the research commencing, the advantages and disadvantages of interviewing parents separately or as parental dyad was considered.

Following discussion with the parents with lived experience, it was decided

that interviewing the parents separately may result in richer data as interviewing parents together may lead to less depth in the responses or avoidance of some topics if they were trying to protect one another's feelings. Another consideration before the interviews were conducted was the potential for causing distress to participants. Were this to have happened, participants would have been offered to pause or terminate the interview and would have been offered comfort and support.

In addition to recording the interviews it was felt important to keep my full attention on the participants during the interview in order to pick up on their non-verbal cues (Hiller and DiLuzio, 2004) and to be able to make notes while they were talking to aid the development of further questions. Remaining attentive to issues of difference with regards to gender, age, ethnicity and power within the interviews was another important consideration and was achieved through the use of a reflective journal.

#### *5.2.6 Interview procedure*

The interviews took place at a location of the participant's choice. In all cases, they chose for the interviews to take place at their home. As such, the department of Health and Life Sciences lone worker policy was adhered to.

Prior to the interview commencing participants had been given the opportunity to read the information sheet (appendix C) and had signed a consent form (appendix D). Participants were given the chance to ask any questions about the study and were reminded of their right to refuse to answer any questions, to terminate the interview at any point and to withdraw from the study at any time after the interview was completed. They were informed that their information would be destroyed upon their request to withdraw from the study. They were also informed about their confidentiality and anonymity being upheld throughout the research process and the use of pseudonyms in the write up to maintain this and about the dissemination of the research.

The interviews began with a brief introduction about myself as a trainee and as a person with a personal interest in the field. It was felt important to be

open with the participants in order to develop rapport. Charmaz (2014) considers researchers' openness in terms of their position with the research vital. It was considered that one way to address issues of power within the interview was to be open and engage in reflexive discussions with the parents. The interview guide began with an open question asking them to "tell me a bit about your family" before moving on to further questions using the interview guide as a tool while following the lead of the participant to explore topics in more depth.

Following the interviews, participants were given an opportunity to discuss the process of the interview and were again reminded about issues of confidentiality and consent and dissemination of the research was discussed again. Participants were thanked and offered a copy of the full report or draft of the paper for publication when the research was completed.

#### *5.2.7 Confidentiality and anonymity*

The interview recordings were downloaded on to my personal computer and the files named using a participant number and a corresponding pseudonym. The files were encrypted and password protected. The consent forms were kept in a locked cabinet at my home. From completion of each interview, every participant was referred to by their number or pseudonym.

#### *5.2.8 Transcription of the data*

The interviews were transcribed and word processed using my personal laptop and Microsoft Word 2013 and saved in password protected files. All of the names and identifiable information was removed from the transcripts and replaced with pseudonyms to ensure confidentiality.

#### *5.2.9 Ethical considerations*

Ethical approval for the study was gained from The University of Hertfordshire research ethics committee prior to participants being recruited (appendix E).

Two amendments were made during the research process and ethical extensions were sought and granted (appendix E). As participants were not recruited from an NHS service, NHS ethical approval was not required. The research was conducted according to the Code of Human Research Ethics laid out by the British Psychological Society (British Psychological Society, 2014).

### 5.3 Data analysis

The data analysis was carried out following the principles and guidelines for GT outlined by Charmaz (2014). The data was collected and analysed in stages allowing time for the transcription and preliminary coding of the initial data which then, in addition to memoing, guided the selection of further participants in order to explore emerging themes in more detail. This approach is consistent with theoretical sampling (Charmaz, 2014). Memoing was also useful throughout the analysis for developing ideas about possible relationships between codes and the possible categories. The memos were also used to document ideas and questions and at the later stages when thinking about the relationships between codes before grouping the codes into sub categories and categories between categories of data (appendix Fii).

The first stage of analysis was line-by-line coding focussing on action words and processes. This was a long and time consuming process. Four transcripts were line-by-line coded in order to fully explore the nuances of the data and to fully immerse the researcher in the data. Once this was achieved the initial codes were analysed and grouped into focused codes. These focused codes raised hypotheses about the process of adjustment the participants were discussing therefore further interviews were undertaken and participants were selected to further explore the hypotheses that were emerging from the analysis. These interviews were transcribed and analysed using the focussed codes while remaining reflexive and open to noticing new and alternative codes emerging and adding new codes to the analysis. The initial codes that were very prevalent in the data were carried forward as focussed codes and sub-categories.

Throughout the process previous transcripts were revisited and compared in order to explore the emerging categories in greater detail. Once all of the transcripts were coded and had been compared the codes were then compared and grouped into sub-categories and then categories using memoing and diagramming to aid the process. This was a time consuming process which evolved many times before the final categories were chosen.

At the final stage of analysis, the relationships between the categories were developed by looking at parents responses to the issues they had faced, their intentions and actions and the consequences of these. During the analysis, particularly the latter stages of the process, diagramming was used for the purposes of theoretical integration (Charmaz, 2014). Diagramming was helpful as it allowed potential relationships between categories to become clearer. Once the draft model depicting the overarching categories and their relationships was agreed upon a further interview was carried out with a Clinical Nurse Specialist who works with parents of children with birthmarks. Both the Clinical Nurse Specialist and parents with lived experience of having a child born with a facial birthmark were also consulted. The aim of these consultations was to fully understand the properties of each category and the relationships between them, and thus to approach theoretical saturation.

### *5.3.1 Methodological rigor*

The validity of this research was assessed using the framework outlined by Elliot, Fisher & Rennie (1999). This framework has seven guidelines which are:

1. Owning one's perspective
2. Situating the sample
3. Grounding in examples
4. Providing credibility checks
5. Coherence
6. Accomplishing general vs. specific research tasks
7. Resonating with readers

Elliot et al. (1999) begin by naming the importance of owning one's position. Sharing my values and assumptions can help the reader to construe my understanding of the data, but also to consider alternative perspectives. I have openly acknowledged my personal epistemology and personal interest and experience. I aimed to bring transparency to the enquiry and recognised how my interests, assumptions and values influence my understanding and the analysis.

Situating the sample appropriately ensures that the reader has sufficient relevant information about the participants. I have aimed to provide the reader with the relevant demographic information about the participants and stated the types of birthmark each child has as this has an impact on the parent's experiences.

The process of analysis outlined above followed the guidelines of Charmaz (2014). This facilitated rigor in the conduct of this study as it ensured that I endorsed purposive and theoretical sampling, achieved depth of analysis through an iterative coding process and memo writing and continuous questioning of the data to ensure theoretical sensitivity. Using this approach enabled me to remain open to new ideas emerging in the data.

To bring further transparency to the enquiry an analysis audit trail has been included in appendix F. This clearly demonstrates the process of data analysis and is supported with quotes taken from transcripts. Quotes are also used throughout the results section in the hope this will bring the participant's experiences to life for the reader. To further support the coherence of the research, two short accounts have been included in appendix G signifying the journey of two parents and demonstrating how the model has been constructed from the data. It is hoped that this audit trail will also show the coherence of the analysis. To further ensure coherence in the analysis a diagram of the process model has been included (See figure 1).

The credibility of the research has been checked throughout the process. I acknowledge that the data and analysis are co-constructed by the participants and myself as both parties bring their views, ideas, experiences and

knowledge to the research which shapes the process and the product (Charmaz, 2014). Taking a reflexive position allowed me to acknowledge how my lived experience and clinical knowledge may shape my assumptions and consider how these might have shaped the research. In order to do this, I met regularly with my research supervisors and jointly reviewed the transcripts, coding and memos and I kept a reflective journal (appendix I) throughout. The supervisors of this study were asked to independently analyse sections of the transcripts allowing for a comparison of codes ensuring that I was coding effectively and capturing the essence of the participant's experience appropriately.

Additionally, my parents were recruited to be consultants on the study as they have lived experience. The consultants were used multiple times during the study, offering advice on the initial interview guide, reviewing the data codes and the categories and then again to review the final model. Another step taken to ensure the credibility of the research was recruiting a Clinical Nurse Specialist who had 17 years of experience in the field as she was able to offer an alternative perspective and provided her professional perspective and comments on the developed model (appendix H).

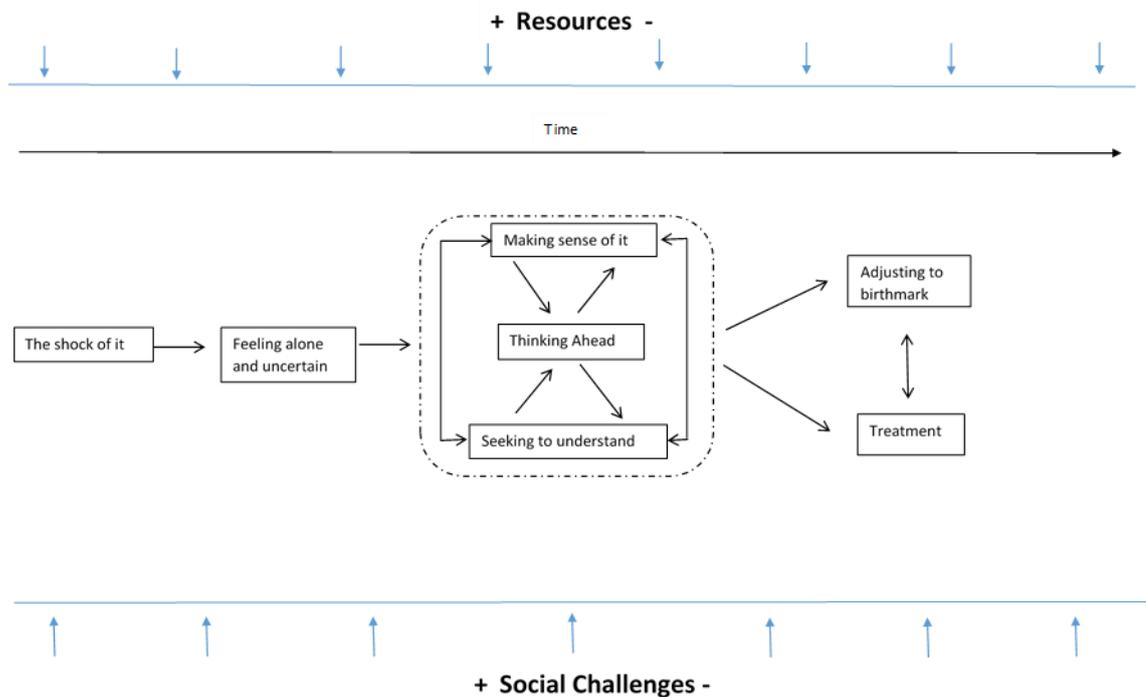
A further consideration outlined by Elliot et al. (1999) is the need to provide clarity to the reader about the general and specific research tasks and to clearly state the limitations. This research offers a deeper understanding about the experiences of parental adjustment following a child being born with a facial birthmark. However, these findings might have relevance and suggest avenues for further researcher in the broader field of parental adjustment to facial differences. This is explained in detail in the discussion section.

## 6. Results

### 6.1 Introducing the model

Figure 1.

*Detailed model of parental adjustment following the birth of a child with a facial birthmark.*



This model illustrates the journey taken by parents as they were adjusting to their child's birthmark.

It considers factors such as the available resources and the social challenges parents faced and how these influenced their experiences. The model demonstrates that 'resources' and 'social challenges' were involved at each stage of the parents' journey and that the amount and availability of the resources were different for each person, as were the extremes of the social challenges they faced. The central flow diagram illustrates the process that the parents appeared to move through as they adjusted to their child's birthmark. They appeared to move through the model at different speeds. Some, seemed to get stuck for a time in parts of the process and had to work

to resolve this in order to progress with their journey. In order to illustrate this process, the journey of two participants has been documented in appendix G.

## 6.2 Exploring the model

I will begin by explaining the 'resources' and 'social challenges' categories and then move on to explore each of the stages involved in the central process of the model. However, for transparency in the data analysis, a table of the categories and models of each category have been included in appendix F. It was important to consider whether the factors that were constructed in this model were shared by other parents on a similar journey. In order to gain greater insight into the experiences parents have adjusting to their child's facial birthmark, Ta Clinical Nurse Specialist with 17 years of experience working with parents of children who have birthmarks and other vascular conditions was interviewed. She was also asked to critique this model and offer suggestions and comments. She suggested that the model was a good representation of the experiences that parents she has worked with have spoken about. A summary of this interview has been included in appendix H for reference.

## 6.3 Resources

The participants talked widely about a number of different resources offered to them for support and whether these aided or hindered their process of coming to terms with their child's birthmark. Potential resources included access to professional input, support from family and friends, help gained through support groups and on the internet, in addition to the personal resources of each parent. The availability and quality of these resources appeared to impact on the parents' ability to manage the challenges they faced, as well as impacting on the overall time it took them to adjust to the birthmark. However, it was apparent that being under resourced in one area on its own, did not necessarily hinder the adjustment process if the parent was well resourced in other areas. The sub-categories included in this category can each be

considered on a continuum; being a resource to a greater or lesser extent for each parent. The resources will now be discussed in more detail.

### *6.3.1 Experience of medical professionals*

Medical professionals were an important and necessary part of the journey for all participants, as in all cases, they were present immediately after the child with a visible difference was born. For some parents, the support, advice and comfort offered by the medical professionals helped them to cope but other parents reported feeling as though they were not being taken seriously or that they were being passed around the medical system without a clear direction. This was understandably a source of frustration and confusion and added to the stress that these parents experienced.

*Amy - "it was all very frustrating when it felt like the midwife, the GP and A&E all agreed there was something wrong and someone needed to look into it and someone needed to take ownership but no one seemed to agree who owned the problem."*

Many parents felt that the medical professionals they encountered, had very poor knowledge and experience of firstly recognising birthmarks, secondly understanding the potential complications of these birthmarks and thirdly knowing where to refer the babies on to. Given the wide geographic variation of the participants, this appears to be a broad ranging experience as opposed to only occurring for participants in one region. Furthermore, this was not limited to a specific profession as the parents reported experiencing this with midwives, health visitors, nurses, hospital doctors and GP's. The Clinical Nurse Specialist who was interviewed also confirmed that these experiences are commonly reported by many of the parents she has encountered in her clinical practice.

*Sally – "She [health visitor] is there to pick up on things so I'd expect her to say your baby has extensive birthmarks, have you been to see someone as it might be an indicator for this or for that and she didn't say anything, appalling really."*

Hannah - *"it's really frustrating that people don't listen, why don't GP's know about this, why don't health visitors know about this? They are the first line coming into contact with these babies."*

However not all participants shared these experiences. For one family it was more positive and they were signposted appropriately and referred in a timely manner which reduced the distress they experienced. Although it is noted that this family had the additional resource of the father being in the medical profession and having friends who were paediatricians who he was able to contact. This particularly strong resource meant that this family were able to move through the adjustment process quicker than other participants.

Sally - *"so the paediatrician was good in terms of going straight off to get more information, get referrals straight away."*

Another issue raised was the bedside manner of medical professionals. This had caused distress for some participants when, perhaps, the medical professional had not considered the stage of adjustment the parents were in or their emotional state, and, therefore, modified their manner accordingly to meet the emotional and information needs of the parents.

Anna – *"there was a consultant stood at the end of the bed peering into her cot, um...he wasn't a people person and he said all the wrong things."*

However, this was not a problem with all medical professionals. Many parents reported feeling safe, comforted and able to trust in the professionals they encountered along their journey and this resource aided their adjustment and coping. Generally, parents reported greater support from professionals who were specialists in the field of birthmarks. It may be that the knowledge these professionals have about birthmarks allowed for the needs of the parents to be met more effectively which, in turn, reduced their distress.

Hannah – *"X [a clinical nurse specialist] was great, like X would reply at all times of day and night which was amazing, she's always on her phone just answering silly worries and what do I do about this or that, or*

*what does it look like or oh God I missed a dose or whatever, she was great at providing comfort.”*

### 6.3.2 Inter-personal support

Some of the participants reported feeling that they were able to seek support and comfort from their partner and spoke of good communication within the couple's relationship being an asset for coping with the emotional impact the birthmark had on them.

Hannah – *“He is a lot calmer than me and he is good at calming me down and giving me a hug or whatever and telling me I was doing the right thing.”*

However, for others, discussing their concerns with their partner did not feel appropriate at times. There appeared to be two main reasons for this. The first was that these parents spoke of wanting to protect their partner from their concerns and the second was they felt that talking at length about their concerns may become unhelpful or distress their partners.

James – *“I guess it's not all the worries it's all the details of the different syndromes particularly. Why didn't I tell her? Because I know she would just worry and we didn't know and there was no purpose behind it.”*

Penny – *“You don't want to talk about it because then I'd upset him, and for what? Then we are both upset.”*

Some couples relied on each other for practical support, especially those who had been, or were currently in the treatment phase with their child. Some reported that working together in this way had helped them to cope with the challenges they were facing. It may be that this team-work provided not only practical support but emotional support and reassurance for both parents.

Harry – *“I think we, we did talk about it a lot, we tried to sort of .....Just be as .....be as, open about it as we could in terms of our own feelings about it and yes we were upset, but no we both were very much, this is what we've got to do isn't it ...and I, I don't think either of us ever*

*questioned it ...but...you know...talking about it also just reinforced it that we sort of ...can't...can't not, you know."*

Extended family members were also discussed in many of the interviews. For some, extended family were seen as an asset; supporting the parents and showing interest and concern about the child's well-being; being emotionally available and supportive, including in terms of practical arrangements and advice; helping them to cope with the emotional and practical aspects of the situation.

*Sally - "she [sister] was really supportive, really helped me when I was feeling weepy she would pander to me."*

*Charlie - "Mum being 20 mins down the road was quite comforting."*

For one parent, her mother's exclamation about not knowing how she was coping, offered her a sense of achievement that she had been able to cope through the experience.

*Hannah - "My mum, my mum, I suppose was a bit more of a panicker, so she'd say things like oh God, I don't know how you are coping with all of this! Which in a weird way was helpful, like it made me realise, yeah I am coping with this and like you wouldn't have been able to."*

However, for others the extended family member's reactions were a source of concern or were found to be unhelpful or unsupportive which may have contributed to the parents' distress and feelings of isolation. Additionally, the reactions of extended family members may be some of the first reactions parents witness from people outside of the immediate family and may be the parents' first experience of how society might react to their child's appearance.

*Amy – "she made a comment about, oh can she [mother in law] take a photo of the haemangioma to show her sister but don't worry she won't show anyone else and obviously it wouldn't be one for the family album. Whereas as far as I'm concerned you take photos of my baby whichever way she happens to face."*

Social support from friends and National Childbirth Trust (NCT) groups were also mentioned as sources of support. More frequently it was the mothers who referred to having social support. This may be related to the mother's being on maternity leave and having greater opportunity to socialise with other mothers compared to the fathers who had all returned to work.

*Penny - "They were really good like that was....like your real friends talk openly to you about it."... "They just loved him instantly and it didn't matter and that was lovely."*

### 6.3.3 Personal resources

Seven parents spoke of factors that they felt influenced their ability to cope with the birthmark, such as age, resourcefulness and personality types. These personal resources appeared to influence the adjustment process. For example, Sally described herself as having strong personal resources, specifically talking about her practical approach to problems which allowed her to seek information and plan ahead. Hannah talked about being a control freak, which she felt was an asset to her when she was managing the treatment regime, but it had also led to her feeling overwhelmed by the situation at times as she felt quite helpless.

*Hannah – "I'm quite a control freak and I wasn't able to control it or stop it doing anything."*

Another factor that appeared to be relevant was the parenting experience they had previously. Four families were first time parents, and three families had previous parenting experience from having at least one other child. It may be that first time parents had different information needs if they were required to understand everything involved with caring for a new born baby as well as comprehending their child's birthmark. An additional challenge for the families that had to treat their child's haemangioma at an early age, may have been differentiating between the treatment side effects and other baby behaviours.

*Sarah – “as an experienced mum like, I know if he is developing well, I don't need to see a doctor for that.”*

*Anna – “We'd never seen a freshly born baby anyway so it was all a bit new to us.”*

Career paths were also resources for some parents. One father was a doctor who was able to contact colleagues for advice and this helped him and his wife to gain an understanding of the birthmark and its related challenges more quickly than for other parents. For another family the grandmother was a nurse and this was reported to be an asset to the family. Another factor that is related to career choice was the financial resources the family had. For one family, they found this to be advantageous as it allowed them to access medical support more quickly as they were able to afford private treatment.

*Harry – “So it wasn't until it really, really got very nasty and infected that suddenly we....sought, through a different angle, we went the specialist angle and we went private and saw a specific person in a posh hospital in London.”*

The parents appeared to cope with the emotions and the uncertainty that they faced in different ways. Their personality types and access to resources may have influenced their ability to cope. Worrying about the birthmark and seeking support and problem solving were common strategies that some parents in this study utilised, however, for others, they managed their concerns through denial or avoidance of the subject.

*Tom - “I've taken a step back, I've sort of like, I don't know if I try not to think about it as much”... “I've just shut off to it as I feel he is what he is.”*

It is likely that the parents who were more naturally inclined to problem solve, were more actively engaged in the process of adjustment and consequently were more able to seek out new resources and draw on their personal resources to cope. Whereas, the parents who were more inclined to try to avoid thinking about the birthmark or the associated challenges may have found it more difficult to progress on their journey. Furthermore, it

is considered that the partners of the parents who were avoidant may have felt a greater isolation and found it more difficult to seek support and share their concerns which, in turn, may have increased their distress.

#### *6.3.4 Support groups*

The Birthmark Support Group and other support groups were recognised to be very helpful for many of the participants. Generally, it was the mothers who were actively involved on the site and found it very helpful to gain advice and share experiences with others. This provided a safe platform for them to discuss their worries and share difficult experiences and gain support from others in the same or similar situations, normalising their experiences. In this way, this resource assisted the process of normalisation discussed in the 'making sense of it' stage'.

*Penny - "that's why joining the sites, the parents have given me like more information than anyone else because they are in the same boat as you."*

A Clinical Nurse Specialist who regularly contributed on the group was also recognised widely as a source of support and her assistance with practical advice and guidance was clearly invaluable to many.

*Kate - "I put a photo on the birthmark support group and said somebody help us find what it is and someone from X came through and said this is what it is I want to see her"*

*Hannah - "because everyone's in the same boat and it was sort of comforting and quite a help that one of the nurses we see at X was one of the founders."*

The support group also offered an opportunity for parents to compare their child with other children and for many this left them feeling grateful that their child's birthmark was having less of an impact on their lives than for many on the group. For others, the photographs that parents post on the group offered

a chance to show their child photographs of other children's birthmarks and normalise the birthmark.

*Anna - "she knows that her facial appearance is a bit different to some other people but being on the Facebook group is great because I can show her pictures of other people who have similar special marks."*

However, one father and the Clinical Nurse Specialist raised some concerns about the use of support groups and expressed that at times greater moderation of the information could be useful.

*James - I think it's sometimes...sometimes its...well X is great when she steps in but there are times when you can see the lack of ...sometimes the slight lack of moderation of the content means that I think people build people's worries more than is necessary.*

### *6.3.5 Birthmark related resources*

A number of birthmark related resources were mentioned including a book called *Sam's Birthmark* and a teddy bear which was made by the organisation Changing Faces. It was evident that resources such as these provided parents with some tools to support them to explain the birthmark to their child, siblings and other children. Furthermore, this was found to help the child to have the words to explain what their birthmark is to others.

*Anna – "We done um... we got a book called Sam's birthmark.... It's OK now she's two but I think there'll come a time when she'll get more questions but at the moment it's been a really helpful tool actually as an introduction into what it is."*

### 6.4 Social difficulties

Social difficulties can also be conceptualised on a continuum as they influenced parents' adjustment to a greater or lesser extent. Those who encountered more social challenges and had less personal resource appeared to move through the process more slowly. The social difficulties that

parents encountered were stressors for many parents and could cause them to feel embarrassed, anxious and self-conscious. It also fed into many parents' concerns about how well society would receive their child as they grow up and influenced thoughts and fears about their child being bullied in the future. This is related to their need to think ahead and plan and prepare for the future which will be discussed in the 'Thinking Ahead' category. Parents encountered a range of social difficulties and reactions from members of the public that influenced their experiences. Some had encountered situations that they had found highly distressing, while others encountered fewer social difficulties. Issues such as the visibility and permanency of the birthmark seemed to be a mediating factor in the strength of reaction from members of the public.

#### *6.4.1 Experiencing reactions from members of the public*

All of the parents had experienced members of the public, extended family members, friends, work colleagues or children reacting to seeing their child's birthmark. This appeared to be irrespective of the size, permanency or visibility of the birthmark. Some parents had experienced milder reactions from the public while others had experienced very strong reactions and found these to be distressing and anxiety provoking.

Parents appeared to perceive the reactions from children and adults differently. The majority of parents appeared to make sense of children's reactions as an innocent and natural curiosity and consequently responded kindly to them and tried to explain to the children about birthmarks.

*Penny - "just let them ask, they're kids, if they say why is he red you just explain what you can and if they say he's red, I say oh yes he is, they don't mean it nastily."*

*Kate - "These were special needs boys with behavioural problems so they could have been quite cruel....as soon as you were just, they could tell you were being honest and you put it in child friendly terms that they could understand and that curiosity was over and done with."*

The parents in the study spoke of trying to remain calm and even tempered when they experienced reactions from members of the public. One explanation parents gave for this was the hope that answering the questions and imparting knowledge about birthmarks might lessen the stigma that their child would face in the future. Although the majority of the parents accepted that it is natural to be curious and to look when you see something unfamiliar, they reported feeling that they felt more annoyed or upset by adults reacting strongly as they felt that they should know better. For some parents, experiencing members of the public react unkindly was very upsetting.

*Amy - " one guy who peered in and at the time it was a very rare time when she was haemangioma side up and he looked in and went ugh and just walked off."*

*Anna – "But then she said she looks like the Phantom of the Opera and I was just really upset... I thought so what, you think my child is some sort of monster that hides in the shadows, you know, and is feared um and I said goodbye Merry Christmas because it was just before Christmas time and I smiled as I walked away, but then cried all the way home and cried for several hours when I got home."*

#### *6.4.2 Feeling judged*

Many parents reported feeling that they may be judged because of their child's facial difference and there appears to be a number of dimensions to this. Participants spoke of witnessing people judging their child on his or her appearance, the parents being judged or blamed for the birthmark or that they were being accused of harming their child.

Thinking about parents feeling judged, their personality, confidence and past experience appear to be factors that played a role in how they imagine others might respond to the birthmark.

*Amy – "Ummm I felt very self-conscious about it, I remember being really nervous about going to meet people at work to show the baby because I felt everyone would look at my ugly baby and judge me on it."*

Another challenge parents faced was that people naturally tried to make sense of what they were seeing and some parents had come across members of the public who made assumptions about the birthmark and thought it was an injury, possibly caused by the parent.

*Jack - "when a kid asked me why I'd done it to her and practically accused me of I don't know what, I didn't really stop to ask. I just blurted back that I hadn't done anything and that it was a birthmark and that she was born with it."*

*George – "you see people seeing and thinking oh my God what have the parents done to this poor child. So we do get a lot of people wondering if this is an injury, wondering if it's because of parents."*

#### 6.4.3 Social Influences

There were a number of factors that were mentioned by the parents that appear to relate to the social and cultural norms of Western society. One such factor was the gender of the child. This appeared to influence the perception of some parents' concern about their child having a birthmark and influence how well they anticipate their child will be able to cope with it as well as how well they will be received by society.

*Sally - "I think it helps that he's a boy to be honest and not a girl bearing in mind appearances."*

*Hannah - "I know it's only aesthetic, I know that but, she's a girl and aesthetically it's important, like she's a girl."*

Another expectation that a few parents spoke of facing, was the assumption that they should want to remove their child's birthmark. It may be that this is a reflection of Western appearance norms such as flawless skin. Also, this may be a reflection of an implicit medical narrative in society about being able to 'fix problems' that are not accepted as the norm.

Penny - *"They almost think that we should want it gone, we don't want it gone, well, at the beginning....and they almost talk to you as if you don't want it there."*

The parents appeared to make sense of people's reactions in the context of both the Western view of appearance ideals and more broadly, the effect of generational differences of knowledge and their personal experiences of social norms and tolerance of difference in society.

Anna - *"Every time someone said it's nothing, its fine, it actually made me feel okay, society is receiving her more positively than I anticipated."*

Amy - *"Well for a start it's harder to cover up on your face, I think something about society that you look at facial defects as being ugly."*

For some parents, considering how society may respond to their child's birthmark and the challenges their child may face factored into their decisions about whether to treat their child's birthmark.

Tom - *"I just think what difference does it mean being half red?...and we've said that we wouldn't do anything....wouldn't try the laser if he could go from 6 to I dunno 20 and get past the teenage years."*

Amy - *"I would have a strong motivation to do it [treatment] for my own personal reasons so I don't have that going into work and everyone judging me and saying what did you do wrong during pregnancy...um...how have you ended up with a freak child."*

Looking at this sub-category, it is evident that the participants' beliefs, experiences and expectations about how society would receive their child influenced the meaning they made about the birthmark, their motivation to prepare their child to cope with living with a facial difference and it was a factor that they considered in their decisions about treatment.

### 6.5 The stages parents move through as they adjust to their child's birthmark

The factors that have been discussed so far, appeared to feed in to all of the categories which have been constructed as the process of the model and

influenced the ease and speed with which parents moved through this process of adjusting to their child's birthmark. The journey that the parents appear to move through will now be discussed.

## 6.6 The shock of it

### *6.6.1 Expecting perfection*

*Anna - "I think everyone expects to have a perfect baby don't they?"*

A number of parents, generally the mothers, spoke about how they had thought about their baby before it was born and wondered about what it would look like and felt shocked when they saw their baby's appearance. Social narratives about having a baby, previous experience of having children and hearing about other people's experiences were spoken about and contributed to the participants' expectation of having their 'perfect' baby.

*Penny - "We knew he'd be big so you've got a vision in your head and he came out and he wasn't what I considered at the time my perfect baby".... "Everyone says when they're born you've never felt anything like, like their perfect. I looked at him and to me he wasn't and I think that was really hard."*

*Amy – "and I was just going silently well objectively no she's not perfect because of ...you know...there's not a lot to judge a baby on, you can only really judge them on their physical appearance, they don't have a personality, not at, you know, six weeks before their due date, they're quite slug like."*

An interesting point raised by the Clinical Nurse Specialist, was that it may be that the shock experienced by parents of children with haemangiomas might be different to parents of children with a PWS, as initially they have their "perfect" baby but within a few days the birthmark is beginning to grow rapidly. Consequently, it may be that they experienced a greater sense of loss than those who are confronted with a PWS at birth.

### 6.6.2 *Feeling shocked*

The shock of seeing the birthmark left many of the parents feeling quite overwhelmed with a mixture of many emotions. This almost had a numbing effect for many of the parents in the study, as they reported feeling stunned and unsure and this feeling of shock was increased for many when they were informed about the related syndromes that can occur with birthmarks. For those parents who had a child with a haemangioma, they reported feeling surprised and this was particularly related to the speed at which the haemangioma grew. This feeling of shock may have been difficult for parents to articulate, as it seemed to involve feelings of joy and excitement for having had a baby that are then combined with feelings of guilt, unanticipated uncertainty and worry.

Penny - *"There wasn't an obvious emotion. It was sort of everything."*

Anna – *"He started mentioning Sturge Webber syndrome and Klippel Trenauney and lifelong disfigurement and we just freaked out big time. And we just sort of looked at each other and said oh my God she's got a Port Wine Stain."*

During this time some mothers said it was difficult to know how to express the emotions they felt but the majority reported feeling very tearful. It must be noted that the emotion the mothers reported is likely to have also been affected by their hormones following child birth.

### 6.7 Feeling alone and uncertain

Following the initial shock, parents spoke of feeling alone with their thoughts, confused about why it had happened and what it was. For some parents, particularly those who strongly thought that their baby wasn't perfect and found it difficult to share those feelings or seek support, reported feeling more isolated. This category captures this period of uncertainty and loneliness the parents were faced with after their child was born.

Penny – *"I felt horrible guilt that I even felt like that, that felt horrible and you feel really lonely"... "So....so you can't talk, it's really lonely and you*

*don't say it.".... "It feels wrong how you feel, that isn't how you imagine having a baby to feel, it's like you can't go and admit to someone that's how you feel."*

All of the parents spoke of uncertainty in relation to what the birthmark was and whether it had been accurately diagnosed. This 'not knowing' seemed to contribute to the doubt and isolation the parents experienced.

*Sally - "she didn't quite know, said that it could be part of a traumatic birth or something but blatantly wasn't as it took barely an hour and half to have him."*

*Jack - "they said oh there's bruising on her face and um, we'd had quite a prolonged labour and we thought maybe it was a bruise."*

Once the birthmark had been diagnosed correctly the parents were then informed of the syndromes and complications that can occur when a child has a birthmark. This left many parents with a greater uncertainty and more unanswered questions.

*Sarah – "it could be birthmark related and it's just not knowing, it's the waiting game to know if he's affected."*

*Penny – "that it [Sturge Webber] came with learning difficulties and glaucoma and seizures and I think that's about it and ...and then I panicked because I didn't know anything about this."*

At this stage many parents began to make early attempts to understand why the birthmark was there. Three mothers quickly blamed themselves for the birthmark, leading to feelings of guilt and responsibility, while others moved more swiftly on to the next stage of seeking to understand.

*Penny - "I went through this guilt stage um and sort of thinking oh God what if it's something I did? .... I'd smashed up the car and I thought what if the car crash, like what if I'd bumped it or something."*

*Anna - "and then when it isn't a perfect baby you think ohh, what have I done?".... "But before it happened I'd been clubbing twice in the time that I think I was pregnant and for ages I was like arrgh, what if I've*

*made this happen in some way. Um ...but the dermatologist was like, you're crazy. That's not how it works, so I feel happy now that I'm not responsible for it in anyway. But it's funny because there's no one else in the family with it, and I look around for other birthmarks to try and explain it, but there's no logic."*

## 6.8 Seeking to understand

Seeking to understand seemed to be the logical next step for all participants from the doubts and questions they faced in the feeling alone stage. Parents appeared to reach out and utilise the resources available to them to help them to understand as quickly as possible. It is at this stage that long waiting lists and inappropriate referrals hindered the journey of some parents, increasing the time it took them to fully understand what they were dealing with. Many parents turned to the internet to look for advice with mixed results. This stage appeared to go hand in hand with the 'making sense of the birthmark' category. Parents seemed to move fluidly between these categories as they gradually gained information about the birthmark and the associated risks, compared their situation with others and assimilated all of the information, gradually allowing them to see the birthmark within the wider context. Seeking to understand represents the active steps parents took, drawing on their resources to be able to understand about the birthmark and their options moving forwards.

### *6.8.1 Information searching*

In order to cope with the uncertainty of the situation many parents quickly began to try to understand what they were dealing with. Generally, parents felt that medical professionals were poorly educated about birthmarks and so began their own research using the internet. For many, this is how they found websites which contained useful information, e.g. the Birthmark Support Group and similar support organisations that enabled them to gain knowledge from professionals and other parents.

Penny - *"I've left hospital with a piece of paper and just googled everything."*

Hannah – *"I thought oh my God, I didn't know what to do and at that point I started googling it crazily.".... "I was on the Facebook group and googling all the time and constantly looking on the GOSH website."*

Another way parents were able to get information was through appointments with specialists. However, for many parents, with the exception of the family who accessed medical professionals privately, this meant being on the waiting lists to be seen, thus prolonging the time until they could have their questions about responsibility for the birthmark and the threats of the syndromes answered.

Anna – *"and we didn't see anybody then till she was eight weeks old when we saw like a dermatologist and a paediatrician."*

Some of the parents felt the need to information search more than others. It appears that this may be related to the resources available to each parent and their personality and coping styles as some parents spoke of preferring to feel in control and felt empowered by understanding the birthmark and the possible complications, while others preferred not to know. For some, information searching was initially helpful, but after a while it became counter-productive and exacerbated their stress.

Hannah - *"I found myself spending hours on, I, people were, I was beginning to get a bit affected by the scare stories, winding myself up a bit."*

Charlie - *"I mean the internet is full of things to scare you."*

### 6.8.2 Having answers

The concluding step within information searching was achieved once they felt that they understood the birthmark and the implications. This appears to be an important resolution that parents needed in order for them to progress on to thinking about treatment options and adjusting to the birthmark.

Penny - *"the glaucoma was fine, I was alright considering he'd given me an answer. Then he told me yeah, he probably will.....so then I could deal with that.".... "When saw the genetic lady she put my mind to rest with the Sturge Webber."*

## 6.9 Making sense of it

This category captures the process that parents go through, trying to make sense of their child having a birthmark and what that meant for them. This process appeared to occur concurrently with the parents' efforts to understand the birthmark. This category also captures the way the different parents in the study made sense of the birthmark in terms of the language they use to describe it and the meaning this gave to the birthmark. In this way, this category captures the reflective aspects of the parents' processes and the meaning they ascribed to having a child with a birthmark.

### *6.9.1 Putting it in perspective*

For many parents, the initial shock and then the uncertainty, doubt and threat of possible associated syndromes was quite overwhelming. For many parents, this process led them to consider 'the worst case scenario' and to compare their situation with others that they have encountered through their jobs, social circles or other parents on the Birthmark Support Group. For many, this process helped them to put the birthmark into perspective. This worst case scenario thinking seemed to have been helpful for the participants, perhaps giving them a different bench mark from the 'perfect baby' benchmark they were previously working from. In this way it may have offered a medium that allowed them to manage their expectations. This idea is quite a common Western concept that is often summed up in the classic expression 'I suppose it could be worse'.

Tom - *"So obviously when we are looking at pictures on the internet of other kids. On X we can't see it as much as he's ours, but we look at other kids and go bloody hell, theirs is bad."*

*Amy – “I mean it felt odd because I was getting quite worried about what was wrong and I’ve had friends who have had far worse with their children. I’ve had two friends who’ve had, whose children have had cancer ...ummmm...errr...a friend from work who had twins a month before I did, one was born with previously um....previously undiagnosed downs syndrome and required intestinal surgery within hours of being born and then scheduled open heart surgery two months later...”*

### 6.9.2 Making meaning

For many parents, particularly the parents who had children with a PWS, finding meaning and purpose in the birthmark appears to be an important step towards accepting it. One way parents spoke about it was in terms of the birthmark being one part of the child’s character. This suggests that these parents had moved from an initial stance that was shaped by social influences and the western appearance ideals which align birthmarks with ‘problems’ or ‘blemishes’ to a new position of finding the birthmark to be a feature that is accepted and spoken about positively and endearingly.

*Penny - “If he gets angry, it’s like a little hulk, but red! Or if he’s hungry.”*

This comment by Penny was made in reference to how the birthmark changes colour depending on the mood of her son.

*Anna - “It’s a great barometer of her moods and health actually, when she’s hot or angry its bright red and if she’s cold it’s pale.”*

For many parents, finding language that felt appropriate to them to describe and talk about the birthmark was important. Some parents viewed the birthmark through a medical lens while others used language that engendered greater acceptance of the birthmark. Factors such as the permanency of the birthmark appeared to be related to the choice of language and terminology used.

*Amy – “Yeah ...her large one is definitely medical, I describe that as a benign tumour when I say haemangioma...I never say strawberry*

*birthmark”....”I say haemangioma and if people don’t understand what that means I say a type of benign tumour, it’s a vascular abnormality.”*

For some parents the birthmarks were described in medical language and referred to as a ‘problem’ or in the case of one family, words such as tumour were used. The purpose of using these terms may have allowed other people to empathise more with the parents and may have offered terminology that is more widely recognised than PWS or haemangioma.

Another phrase that was debated by families was the use of the word special. For some parents using this word aimed to promote the child’s confidence and acceptance of their difference, while for other parents this was seen to be a less helpful language.

*Kate - "and they used to say that it means you've been kissed by an angel.”*

*Sally - "We don’t say special, your special mark because when you have more than one child, what’s special about them?"*

*Anna - "well lots of um 'oh look at your beautiful mark' and talking positively about her mark."*

A difference in the language used also appeared to differ based on the type of birthmark. ‘Haemangiomas’ were more commonly spoken about in medical terms, particularly when they ulcerated and subsequently required treatment. Furthermore, it may be that haemangiomas can be conceptualised more easily as a medical ‘problem’ as they can be effectively treated and will eventually disappear where as a PWS is life-long.

### *6.9.3 Normalising experiences*

For many parents, particularly the mothers, having medical professionals and other parents, for instance on the Birthmark Support Group, normalise their worries and concerns was described to be helpful. This may have helped to reduce the isolation that they reported feeling in the ‘feeling alone’ category.

The Clinical Nurse Specialist who was interviewed also confirmed that in her experience, normalising distress and acknowledging concerns is very reassuring for parents.

*Sally - "people just realise that they are not the only ones, they are not isolated, they can put anything down really and people won't be judgemental."*

*Kate - " I mean you start talking to people and they say my so and so has one of those on this part of their body and they've left it and it was absolutely fine, kind of thing...you think oh it's one of those things that if only people talked about more."*

#### 6.10 Thinking ahead

Making sense of the birthmark and information searching, appeared to allow participants to understand what their baby having a birthmark meant medically, socially and personally. It seems that for most participants, after gaining this firmer understanding of what the birthmark is and ascribing meaning to the experience of their child having a birthmark, they were then able to begin thinking about the future, and what might lie in store for their child growing up with a visible difference. At this stage parents appeared to pre-empt potential challenges that they and their child may face, such as stares, comments and bullying. For some, thinking ahead began early in the process, while for others, it appeared that their focus was more on understanding and making sense of the birthmark initially, and considering future implications and challenges followed on from this.

The Clinical Nurse Specialist commented that in her experience, some parents think ahead and worry about the future, rather than thinking of practical steps they could take to manage the situation. It is considered that parents who try to problem solve more naturally may adjust better while other parents may need the support and advice from others to introduce new ways of coping.

### 6.10.1 How to talk about it

Many parents spoke of spending time thinking about the practical task of how they were going to explain the birthmark to their child and how they were going to help them to cope with the reactions they may experience from other children or adults. The birthmark related resources played a large role in the category. Some parents had thought about using more practical strategies such as giving the child a way to explain it and, or, using role play to practice coping with social remarks.

*Sally – “we’ve done a bit of role play with his sister, so, I’ve said to her, you know, what do you, this is what to say to people say what’s on his face and you say it’s just a birthmark, he was born with it, not to make a big deal of it but to be prepared, and, probably, I’ll say the same to him when he gets older”..... “So we try like more of the role play and we might get that book ‘Sam’s Birthmark’ he’ll definitely think that’s about him, and we have a teddy from that Changing Faces charity.”*

*Anna – “So we’ve learnt it off by heart and whenever anybody asks us at playgroup or anyone asks her... Yeah and she knows what to say and she says it’s my birthmark it’s been there since day one, it’s in the right place, big smile and off she goes.”*

Other parents spoke of instilling a greater awareness and tolerance of difference in their child, hoping that this would prepare their child for the experiences they may face. More frequently with the parents in this study, it was the mothers who spoke about using more practical strategies, while the fathers spoke about normalising differences in appearance.

*Tom – “I think that... we’ve talked about it, and thought about what the best thing to do is and like to let him know that people at times may say stuff about it and the other thing we’ve considered is drilling into him that no one is perfect.”*

*James – “I think we’ve talked about just not making a big deal of it, making clear it’s part of him, it’s just who he is and we’ve all got birthmarks and its immaterial really....ummmmm...at the end of the*

*day...ummm...if people have got a problem with it it's more their problem than it is his. Ummm...he is who he is and he needs to just enjoy who he is, at the end of the day is...I think our view on it."*

As well as thinking about how to talk to their child, the majority of parents had considered how to talk to others about their child's birthmark. It appeared that this was related to a number of factors, including if they felt self-conscious or judged by others or when they were trying to avoid an awkward situation such as someone saying the wrong thing. Many parents spent time pre-empting the types of questions they may be asked and the reactions others may have in order to feel prepared to face such situations. It was noted that the parents who appeared to have not planned in this way were more anxious and avoidant of social encounters in comparison to those who had.

*Anna - "I rang my Mum and the first thing I said was I'd had a little girl and the second thing was she's got a facial birthmark because I thought I don't want Mum to come in and go urgh what's that?!"*

*Hannah - "I didn't want to go out or take her into shops in case of what people might say."*

Another dimension of knowing what to say in public situations was when members of the public mistook the birthmark for something else. Participants spoke of experiencing this and generating word patterns to manage these situations.

*Sally - "there are some times those people say oh my cousin had that and it faded and blah blah blah and I have to say no it's not true in X's case."*

*Jack - "they will always ask the question, is that a strawberry haemangioma or oh that's going to fade isn't it, like my brother had one of those and it faded and no it's like no, it's not going to fade. That's an awkward conversation to have."*

It was apparent that interactions with members of the public were a general concern for many of the parents. Many felt that it was important to have

thought about their responses ahead of time and to answer questions thoughtfully in an effort to engender greater acceptance, while others found this to be intrusive and reflected on times that they have struggled to maintain their composure. Many reported that they worked hard to maintain their composure within public settings, but at times relied on their resources, including their partners, family and friends, for support to cope when they were out of the public eye.

#### *6.10.2 Worrying about the future*

Many parents expressed concerns about what the future might hold for their child, in terms of treatment and the social challenges they may face such as when they start school or nursery. Some parents were thinking even further into the future and considering how their child might cope as a teenager and into adulthood.

*Sally – “On the one hand I think he’ll be fine and will still be the centre of attention at school and have lots of friends, um, but on the other hand sometimes I do think, what is going to happen? Is his confidence going to be dented? Um, how will it change his personality?”*

*Hannah – “she’s basically scarred for life and she may find that when she wants to put her hair up she can’t do that, but when she’s 14 and all her friends are doing that as it’s the fashion and she can’t.”*

The majority of parents had either experienced unkind reactions from people or read about other people’s experiences. For many parents, this generated a feeling of protectiveness which was expressed in their worries and planning for the future.

*Penny - “I just thought that’s it, maybe I should home school him and then you think hold on I can’t shelter the poor kid from everything.”*

*Sally - “I don’t know how bad or good it will be, I sometimes get waves that kind of.....protectionism.”.... “But um, yeah you do think what if it gets worse, what do you do? So you do get very protective.”*

It is accepted to be normal for parents to feel protective of their children, although some parents such as Penny had some initially strong reactions, wanting to shelter her son from any potential unkind remarks, while other mothers were more engaged in trying to boost their child's confidence and provide their child with the skills to manage the social challenges that they will inevitably face. In an effort to reduce the social impact their child's birthmark another option parents had was to explore treatment options.

## 6.11 Treatment

### *6.11.1 To treat or not to treat?*

Deciding whether to treat their child's birthmark was a dilemma that all of the parents spoke about. Thoughts about treatment often began in the 'Seeking to Understand' stage, when parents were researching about the birthmark and were considered again as part of the process of 'Thinking Ahead'. These stages occurred prior to any parents moving into treatment, but for clarity, this sub-category has been included with the 'Treatment' category. For those with a PWS or sebaceous naevus the decisions about treatment were not immediate, but for those parents who had a child with a haemangioma the decision about treatment was required sooner. Factors including medical advice, the risks of the treatment, how the child may feel about it in the future and their acceptance of the birthmark appeared to be involved in the parents' thoughts about treatment.

*Amy – “My husband and I did make the decision when we went to Great Ormond Street that if they thought it was just cosmetic, since haemangiomas are temporary cosmetic issues we would not risk medication. But the opinion of the doctors was that it was a functional issue that was going to affect her hearing, affecting her feeding, it would affect her posture it was better for medical reasons rather than cosmetic reasons to take the medication”.....”but if it's a cosmetic thing that's going to clear up then let's not do something for my own vanity of wanting to have a cute child for the first seven years, when actually it would go away naturally in seven years.”*

*Jack – “We nearly always end up saying we don't really like the idea of her face without it now. Because that's the face that we know um... but in an ideal world, we'd wait and ask her whether she wanted to treat it. But we have been told in no uncertain terms that if we don't get it done early it won't have much effect at all.”*

The treatment for haemangiomas is a beta-blocker which, depending on the size, severity and location of the haemangioma, can be taken either orally or as a topical ointment. The risks for the topical ointment Timolol were described to be minor, while the oral medication Propanolol was a greater cause of concern for parents as it has potential side effects such as affecting heart rate and blood sugar levels.

*Amy – “I had concerns that the Propanolol, the treatment they use, has only been going for six years so there's no long term studies into the impact .....ummm....You hear risks of developmental delays .....Ummm....you .....there's kind of ...all kinds of rumours on the internet about maybe it causes problems with teeth development but that may be the haemangioma itself, or maybe it's just coincidence.*

*Charlie -"cos when they sort of said it's a beta blocker...and it, the treatment that they gave was a beta blocker you instantly then, if you start researching what a beta blocker is well that's to do with heart attacks or something because it slows down the blood vessels. You think oh what, what should we really be putting this on our baby."*

For the parents of children with a PWS or sebaceous naevus the options of laser treatment for the PWS, or surgery or laser treatment for the sebaceous naevus would result in the child having to have a general anaesthetic. This was a concern to all of the parents considering laser treatment.

*James – “I think the only ummm thing is the anaesthetic risk for a child that young, that's all .....ummm....he's not going to remember much else to be honest.”*

*Jack – “I don't want to traumatise her too much by putting her through it because it's definitely a traumatic experience to go into the anaesthetic.*

*Umm... but also I don't want to make a decision not to have it and then her to find out that you know she could have had her PWS treated and that may have had some effect at a later stage have her discover that she could have had less pronounced birthmark when she's older."*

However, for the parents who took part in this study, they had all decided it was in their child's best interest to treat their birthmark after weighing up the risks. A factor in this decision appeared to be the parents' thoughts about the reactions of others and how having a visible difference may result in additional social challenges for their child as well as possible future treatment requirements.

*James – "Yeah I think it would be good because it would, may minimize what people's initial reaction would be to it."*

*Amy - "given the likelihood that hers had got to be that big, that when she was older she would want treatment it would probably on a balance of risks have been better to have given her the less invasive treatment when she was younger to save her having to go through something a lot more traumatic when she was older."*

The decision about whether to treat appeared to be a difficult decision for all of the parents. Making decisions for their child without being able to ask them for their opinion was a source of apprehension for many parents and they expressed concerns that their child may regret their choices. However, having appropriate knowledge and medical support during the decision making and the treatment process appeared to be a great support to the parents.

### *6.11.2 Treatment management*

The treatment routine for the parents who had a child with a haemangioma, involved an intensive routine that required the parents to plan and organise themselves appropriately to manage this. For some parents, this initially was quite an adjustment, while others were able to incorporate it into their routine

with relative ease. However, the severity of the haemangioma and whether it had ulcerated factored in to the treatment management.

Hannah – *“so, trying to get her to have the propranolol was really hard and we had to build up the dose as well so it wasn’t very much and we had to give it 3 times a day, and there were all sorts of rules about....if she’s had propranolol she must eat within, have milk within 6 hours of it otherwise she might have a hypo, which terrified the living day lights out of me”..... “they said it needed to be dressed and that she should have a bandage all the way around her head like a headband that covered half of her ear. And there were several layers to the dressing.”.....”My husband likes to go in early, to work, but he wasn’t able to do that anymore as it took both of us, he had to help me, we did it every morning from mid-September to Christmas so it was 3 ½ months.”*

For those who had begun laser treatment, the routine of treatment spanned over a time as the treatment process involves having patch tests first before starting the most appropriate treatment which occurs every few months.

Sally – *“so he had a patch test that didn’t work and we went back for another one which has worked, but not massively...they whacked up the laser on it um, so we go back in January.”*

Anna – *“when she was one we started laser treatment ...ahhhh... she started laser treatment.” ....”She has them every three or four months under general anaesthetic.”*

The daily routine of treatment for the parents of children with haemangiomas initially appeared to be difficult and anxiety provoking, both in adjusting to the timing of the medication and managing the baby’s blood sugar levels as hypoglycaemia was one of the risks of the treatment. However, the parents appeared to settle into a routine allowing them to manage the treatment effectively. Hannah reported that having the support of their GP’s and local pharmacy was an asset to her coping with this routine. For the two families that had begun the laser treatment aside from

the anaesthetic risk, managing the routine of treatment and aftercare appeared to have gone smoothly for both families.

## 6.12 Accepting the birthmark

### 6.12.1 *Accepting the birthmark*

Not all of the parents in this study had reached the point of accepting their child's birthmark. It appears that they needed to move through the stages of this process in order to be able to move towards acceptance of the birthmark. However, a number of factors appear to influence the parents' acceptance of their child's birthmark. Firstly, the permanency of the birthmark appears to influence whether or not the parents felt that they needed to accept the birthmark.

*Anna - "It's never going to disappear and I think it you've got to like it or lump it, you know what are you going to do, you've got this gorgeous little baby that you've just made, that you've waited a long, long time for and is wonderful, you're going to like it, aren't you, eventually."*

For those parents whose child had a haemangioma they were not necessarily facing a life-long impact on their child's appearance, so it could be suggested that this may have made it easier to accept the birthmark.

*Kate – "Yeah...I don't...It's kind of a chapter in her life and when that chapter's closed we'll just shelve the book so to speak and then just get it out when we need to."....."Yeah, like I say I think we were just very lucky that it was so small and....it doesn't...hasn't had any impact on her that's stopped her doing anything."*

A second feature of this acceptance seems to be when parents start to notice the birthmark less and start to focus on other aspects of their child's personality, physical appearance and development.

*Sally - "I do still see it but not all of the time, you don't notice it.".... "I think he is a very attractive baby."...."he is such an engaging toddler,*

*very smiley and people, you know, he does attract people, he's got that sort of.... charisma."*

A third factor that appears to be related to the acceptance of the birthmark is the location and size of it. For those who had larger and more visible birthmarks, adjusting to the birthmark may have been made more difficult in light of the greater social challenges they faced, although it was noted that the parents were more able to accept public curiosity once they had accepted the birthmark compared to when they are at earlier stages of adjustment.

*Penny - " Now, like it is X it is him, do you know what I mean and what we have found is that everybody knows him even though they don't recognise me, we go out and they...they'll know him, and say we've met before and we'll not remember them.....but they must remember like meeting him, I mean, he's half red, like how often do you meet a baby that's half red."*

It may be that parents find it more difficult to manage the social challenges at the earlier stages of adjustment as they had not yet gone through the process of understanding and making sense of the birthmark and accessed their resources to help them to cope.

#### *6.12.2 Leaving a mark*

Reflecting back on their experiences appeared to be an important aspect of accepting the birthmark in their lives. For some, this experience had been very upsetting and stressful and they were left feeling as though they had missed out on some aspects of their child's infancy.

*Hannah - "like in a way, it spoilt my enjoyment of her, like I was so excited to hear I was having a girl as I have a boy already and I really wanted a girl so I was really delighted but then, I dunno, I couldn't really enjoy her for those first few months."*

While for those whose child had a smaller birthmark, there was a sense of relief that it wasn't worse and that it hadn't impacted on the child or the family.

*Kate - "we were lucky that it's so small and wasn't raised and it didn't affect anything."*

### 6.12.3 Promoting birthmarks

Many parents expressed a wish to promote awareness of birthmarks in both the public and professional domain. Many reported how grateful they were to the people had supported them to cope including other parents who they had spoken to in the support groups. Many wanted to give back in some way and to support others on a similar journey.

*Kate - "now we know what we are looking out for a bit and should it happen to any others we have umm.... also anybody else we know we can give advice and support."*

*Amy - "I've suddenly become quite an expert at diagnosing everyone else's birthmarks."*

Parents also spoke of the lack of knowledge about birthmarks within medical professions and the public arena and wished to promote better training for staff and greater awareness for the general public.

*George – "Well....I mean....errr ...the biggest concern, and it's not just the birthmarks unfortunately but it is primarily, is the lack of medical knowledge."*

*Harry – "we're not talking about cancer cures here but we're talking about something which is, is not difficult, ....it's simple, you know...it can be, you know, just a bit of education and we can avoid a load of problems."*

Being able to reflect back on their experiences may suggest that parents have adjusted sufficiently to having their child's birthmark in their lives and that they have sufficient resources to be able to cope and to offer support to others.

## **7. Discussion**

The findings of this study will now be discussed in relation to the research questions posed and existing literature. The potential methodological issues, clinical implications and areas for future research are all considered and the research experience is reflected upon.

### 7.1 Revisiting the research questions

The following section discusses the main findings of this research and relevant literature in relation to the two research questions which were:

1. How do parents adjust after the birth of a child with a facial birthmark?
2. What are the factors that are involved in this process of adjustment and coping?

### 7.2 How do parents adjust after the birth of a child with a facial birthmark?

The participants in this research project appeared to go on a journey after their child was born with a facial birthmark. All of the parents who participated in the study reflected on the change in how they felt about their child's birthmark initially, to how they felt currently and although not all participants appeared to have fully accepted the birthmark, they had all evolved strategies to cope. The ability for parents to move along this journey was facilitated by their access to resources and could be hindered by limited resource or the social challenges they faced. This journey will now be elaborated through the discussion of the specific factors that were found to be involved in this process.

### 7.3 The factors involved in parents adjusting to having a child born with a facial birthmark

#### *7.3.1 Access to resources*

The parents in this study described a number of facilitating factors that enabled their journey from the shock of it, to the acceptance of the birthmark. Some parents had greater access to resources than others who were less

well-resourced and it appears that this was a factor in the time it took them to move towards accepting their baby's birthmark. Resources include interpersonal support, intrapersonal qualities, medical support and material resources such as financial wealth. Access to birthmark specific resources and support groups appeared to support better adjustment. These factors are similar to those involved in building resilience. Resilience can be built when a person is able to interact effectively with their environment and develop processes that promote well-being and/or reduce risk (Bradford, 1997). The findings of this study are consistent with much of the existing literature in the field of visible difference, that has found that resilience is associated with better adjustment and coping (Prior & O'Dell, 2009; Moss, Lawson & White, 2014; Roberts & Shute, 2012; Thompson & Kent, 2001; Tanner et al., 1998). Many of the factors in the resources category influenced how well parents in this study adjusted and these elements have been previously documented as factors which, if available, can promote resilience. The resources category maps on the resistance factors described in the model of child and maternal adjustment to chronic health conditions proposed by Wallander et al. (1989) and Wallander & Varni (1992) which incorporates Lazarus & Folkman's Transactional model. In its own right, Lazarus & Folkman's (1984) Transactional model has been used to focus on the experiences of people with visible difference; identifying adaptive (e.g. seeking support) and maladaptive (e.g. avoidance) coping strategies. The model developed in this study does not explicitly acknowledge the difference between adaptive and maladaptive strategies endorsed by the parents. Instead, it acknowledges more generally the importance of the accessibility to resources and support which, in turn, may promote greater adaptive coping. Adopting this perspective allows for each resource to be considered on a continuum as opposed to the more binary categories of adaptive and maladaptive strategies.

Medical professionals are a key resource and the parents in this study reported that at times they were dissatisfied with the advice they were given. This is not a unique experience as research indicates that parents in other studies discussed in the literature review also reported being disappointed

with the support and advice offered by medical professionals as they felt that the medical professionals lacked knowledge about how to handle the situation when a baby is born with a visible difference (Johansson & Ringsberg, 2004; Tanner et al., 1998; Miller et al., 1999; Stock & Rumsey, 2015). The results of this study are in keeping with existing literature and highlight that inadequate professional support can invoke significant distress for parents and has the potential to impact longer term adjustment and coping, whilst positive experiences can be containing for parents and facilitate greater coping and overall adjustment.

### *7.3.2 Societal and cultural influences*

The results of this study support the findings of previous research which suggests that individuals with a facial difference and their carers, are subjected to social stigma based on their appearance and experience negative reactions from others including stares and in some cases rude or intrusive comments and other forms of unwanted response (Thompson & Kent, 2001). Research indicates that people's reactions may be the response of the complex interplay between a number of factors including evolutionary influences (Park, Faulkner & Shaller, 2003; Ryan, Oaten, Stevenson & Case, 2012), perceiver disgust (Shanmugarajah, Gaiind, Clarke & Butler, 2012) and social and cultural stereotypes (Wheeler & Kim, 1997). Some parents in this study reported experiencing increased stress resulting from these difficulties or anticipated difficulties with public reactions such as staring, insensitive remarks and questions about possible abuse. These issues have also been reported in other studies (Tanner et al., 1998; Bradbury & Hewison, 1994; Williams 3rd et al., 2003; Roberts & Shute, 2011; Nelson et al., 2011; Nelson et al., 2012). These experiences are reported to have generated feelings of sadness, helplessness, anxiety and anger in parents. Zweegers & Van der Vleuten (2012) report that some parents change their behaviours in response to social stigma such as avoiding public places and making attempts to hide the birthmark and this was certainly the case for some participants in this study, particularly in the early stages of their adjustment. This research

suggests that the resourcefulness and resilience of a parent may be a factor in learning to manage unwanted reactions and social stigma. Participants who endorsed active coping strategies appeared to adjust better and appeared to have developed strategies and word patterns which helped them to navigate through social interactions. On the other hand, participants who reported finding public reactions more anxiety provoking, perhaps due to the uncertainty of not knowing if people would comment or stare, appeared to endorse more avoidant coping strategies and at times, reported feeling overwhelmed by the situation.

The gender of the child is thought to play a role in adjusting to living with visible difference although little research has been conducted to explore this phenomenon (Robinson, 1997). Rumsey (1997) suggests that high value is placed on physical attractiveness within Western society particularly for women, suggesting that women are more likely to have greater difficulty adjusting to visible difference. Although previous literature refers to first hand experiences of living with a visible difference, the findings of this study indicate that parents also consider the gender of their child to be a factor that influences adjustment. The parents in this study who had daughters were concerned about appearance perhaps a little more than the parents who had sons. This appeared to relate to the fear of social stigma and the parents' own perceptions and understanding of gender and stereotypes. Another consideration for the parents was whether their child would be robust enough to cope with any social stigma they may encounter and how, as parents they could support their child to manage this. The findings of this study support those of Klein et al. (2006) and Klein et al. (2010) who reported differences in the approaches mothers and fathers took to supporting their children. It is apparent that mothers in this research and in Klein et al. (2006) study took a more active approach such as supporting their children to develop strategies to manage social stigma, whereas the fathers considered the wider social discourses about appearance and social norms. However, the exception to this was one male participant in the study who was a stay at home father and so encountered more social difficulties than other fathers reported and took a similar approach to his wife, talking positively about the birthmark and

encouraging their daughter to learn phrases to explain her birthmark to others. This suggests that it may be the frequency with which social difficulties are encountered that plays a role in the approach that parents take to coping with social stigma rather than a gender difference. Further research is necessary to investigate this hypothesis further.

### *7.3.3 Experiences of finding out about the birthmark*

The parents who took part in this study described the shock they felt when they realised their child had a facial birthmark. These experiences are similar to those described in existing research which suggests that parents of children with any type of visible difference, experience a mixture of emotions such as shock, guilt and grief (Vanz & Ribeiro, 2011; Bradbury & Hewison, 1994; Tanner et al., 1998). Parents in this study also described feelings of loss in relation to the anticipated 'perfect' baby. This is a theme that has previously been found along with the associated concerns about the prognosis for the child's health and long term wellbeing (Bradbury & Hewison, 1994). These feelings were discussed by parents who had children with PWS birthmarks and haemangiomas. This is consistent with existing literature about parents of children with haemangiomas, despite the generally benign nature of haemangiomas and the prognosis for eventual involution (Tanner et al., 1998; Hoornweg et al., 2009). It is also consistent with Bradbury & Hewison (1994) and Koot et al. (2008), whose studies indicates that the duration of the initial shock varied and this variation appeared to be related to parents' personal coping factors rather than the type or severity of the birthmark.

In this study some parents reported that initially it was a lonely time and three participants also felt extreme guilt as they wondered if it was something they had done that had caused the birthmark. This led to them having many questions about how and why it had happened. Williams 3<sup>rd</sup> et al. (2009) found that parents of children with CL/P had similar reactions to their experiences, worrying about their child's future and asking 'why us?'. There is evidence in this study that parents with greater resources, for example, the

doctor who had knowledge and contacts and one family who had private healthcare were able to move forwards more easily.

#### *7.3.4 The process of understanding and giving meaning to the birthmark*

The central section of the model outlined in this research illustrates a dynamic process where parents are iteratively working to obtain and assimilate information before moving on to the possibly more difficult process of aggregating the information and consequences of the birthmark. While other literature supports the earlier stages described in this model, it is considered that this is the first attempt to conceptualise the process parents go through as they come to terms with their baby's birthmark. For the parents in this study, this process appeared to be fluid and involved factors including their ability to seek out the information they needed to understand the birthmark and any associated risks, to have their thoughts about their child's appearance and any health concerns or other experiences normalised and to find a language that felt appropriate for them to talk about the birthmark. In addition to these processes, parents in this study were also considering the future and the possible challenges they may face and they were beginning to consider how they would overcome or manage these challenges.

Looking more closely at the concept of thinking ahead, parents reported wanting to prepare their child to manage the comments and reactions they are likely to encounter from others. Parents also expressed concern about their child's future peer relationships and the likelihood that they may experience teasing or bullying which is a common concern for parents of children with a visible difference (Nelson et al., 2011; Nelson et al., 2012; Johansson & Ringsberg, 2004; Klein et al., 2006; Bradbury & Hewison 1994; Hoornweg et al., 2009). As they grow up children will, for the first time, be expected to integrate with peers at nursery, playgroup or school. While the family environment may have accepted a facial difference, in their expanding social world a child may face new challenges managing other children's curiosity and responses, including unkind remarks about the birthmark (Rumsey, & Harcourt, 2005, 2012). Understandably, parents in this study expressed

feelings of protectiveness towards their children because of their visible difference and this concept has been found previously in the literature (Bradbury & Hewison, 1994; Klein et al., 2006; Nelson et al., 2011; Nelson et al., 2012). The way in which a child is able to manage these experiences and is supported to develop coping strategies may impact their self-esteem and self-concept and subsequently their success in developing interpersonal relationships (Berger & Dalton, 2009). The parents in this study promoted parenting strategies aimed at normalising individual differences, including appearance related differences, providing the child with the language to explain about their birthmark and recognising the positive attributes of the child. These findings are similar to those reported by Klein et al. (2006) who discussed parenting strategies aimed at promoting autonomy, social and emotional adjustment and pointing out the child's strengths.

#### *7.3.4.1 Use of language and terminology*

In referring to their child's birthmark there were a number of differences in the language and terminology parents used and it is considered that this variation may be, in part, related to the way parents made sense of the birthmark. Viewing this through a social constructionist lens, it is evident that the social meaning of a condition or illness is distinct from the biological condition or disease (Eisenberg 1977). The meanings the parents gave to the birthmark and their subsequent experience of it may be shaped by their cultural and social systems. For example, one participant who was particularly concerned about being judged by others chose to use more medical terminology to describe her baby's haemangioma and this may have been related to her wishing to help others to understand about the birthmark or alternatively because this promotes the notion that it can be treated and made better. Birthmarks, as is the case with illness more generally, can have a medical component that is distinct from the lived experience. This difference may occur if it has a particular social or cultural meaning attributed to it. However, Conrad & Barker (2010) argue that this distinction often goes unnoticed or is taken for granted. Many of the parents in this study spoke of coming across

non-specialist professionals who, the parents felt, appeared to be less receptive to their concerns and that the birthmark was treated as a less significant issue than if it had greater medical implications. An example of this was spoken about by one mother who said that she had felt the need to inform her GP about the MRI scans and tests her son had in the hope that this would be of greater significance to her doctor and that then her concerns would be taken more seriously.

The meanings that parents give to the birthmark may have an impact on their acceptance of their child's appearance and later on, on the child's construction of what it means to have a birthmark. Cultural analysts note that medical conditions can have metaphorical connotations. For example, cancer may be associated with being evil while obesity may be associated with terms such as gluttony (Conrad & Barker, 2010). Although many of the parents in this study chose not to use metaphorical language, one participant spoke of hearing people refer to birthmarks as a sign of being kissed by an angel and another family had encountered people who said that they are a sign of good luck. One participant had also encountered a member of the public who likened her daughter's birthmark to the mask worn by the Phantom of the Opera, which was distressing to this mother, although, it suggests that in some ways metaphor may be useful for helping people to find the words to talk about issues that they otherwise do not have the language for. Therefore, it is considered that there is nothing inherent about a birthmark that makes it stigmatizing. Instead, it is the society's response to the visible difference as a result of cultural and social norms (Conrad & Barker, 2010) that engenders less acceptance of visible difference, while medical terminology promotes an implicit understanding of the birthmark being a 'problem' that can be 'fixed'.

### *7.3.5 The treatment dilemma*

The parents report having conflicting feelings about making choices about their child's appearance, particularly when this required putting the child through a number of surgeries. For some parents having to put their child through a general anaesthetic was a significant factor in this dilemma.

Parents discussed the dilemma of wanting to reduce the visible difference in the hope that this would reduce the social challenges the child would encounter as they grow up but having a strong urge to protect the child from harm and from the possible distress and discomfort treatment may cause. Furthermore, two couples spoke of having mixed feelings about changing their child's appearance through treatment as they had accepted the birthmark as being part of their child's face. An additional factor that parents spoke of was one of making decisions on their child's behalf and the concerns they had about making a decision now that in the future their child may regret. Nelson et al. (2011) reports similar findings from a group of parents of children with CL/P who spoke of wishing to 'normalise' their child's appearance while weighing that up against the risks and their urge to protect their child from harm. The way in which this information was delivered to parents and the time, clarity and manner in which the information was shared impacted on the distress experienced by parents and their ability to cope with this dilemma. Similar findings were reported by Miller et al. (1999) who suggested that professionals need to be more aware of the stage of psychological adjustment parents are at and need to share the information sensitively, giving them time to comprehend the information and ask any questions they may have. This is the first study that elucidates this adjustment process and these findings suggest that treatment related decisions were only really processed by parents after they had developed an understanding of the birthmark, given meaning to the birthmark and begun to think about the implications for the future. Signposting parents towards support groups appeared to aid the adjustment process as it allowed them to access support from other parents in similar situations, or from parents who are further along their journeys who could share their experiences. This was a notable resource for parents in this study.

It is noted that medical professionals do not routinely assess the impact a visible difference has on parental psycho-social well-being which may be an important factor especially when parents are required to make significant decisions regarding their child's treatment. This is important as it would ensure the decisions made are in the best interests of the child rather than

perhaps driven by the parents own appearance consciousness, fear of being judged or wish for a 'perfect' baby as discussed by one parent in this study.

### *7.3.6 The importance of parental adjustment*

It is evident that, following the birth of a child with a facial birthmark there was a range of reactions experienced by parents. Some parents reported being able to adjust more easily, while others experienced greater difficulty coming to terms with their child's birthmark. One mother reported that she found it difficult to bond with her baby initially and another spoke of being shocked anew each time she saw her son. Two mothers reported feeling like they missed out on something in the early stages of their baby's life as they were focussed on the birthmark. Considering these experiences, it is evident that for some parents, having a child born with a facial birthmark may have impacted on their early bonding process. However, this was not the case for all of the parents and it was noted that generally it was the mothers in the study who found it more distressing. One hypothesis is that this may be related to the sense of loss that was reported. Also the guilt that some mother's experienced during the early stages as they felt responsible for the birthmark may be another factor. Another suggestion is that this distress may have been more intense for mothers as they were at home on maternity leave while the fathers were at work and thus reminded of it less frequently.

Attachment figures are particularly important to infants as they allow them to begin to build mental representations of their own worth based on the parents or care givers availability and willingness to provide protection and care (Ainsworth et al., 1978). As a child grows, the quality of these relationships may have an impact on the child's understanding of themselves, others and social interactions not only in childhood but into adulthood. Given the variation of experience in this study, it is suggested that other contextual factors and access to resources could account for the differences in early bonding experiences discussed by these families. This finding lends support to Rumsey and Stock (2013) who suggest that attachment must be considered alongside family factors such as socioeconomic status, family attachment

history, parental coping styles, parental mental well-being, beliefs about the cause of the disfigurement and feelings of responsibility and other child related factors including temperament and co-morbidity with other disorders.

#### 7.4 Theoretical approaches to parental adjustment

Searching the literature, it appears that this is the first model that addresses solely parental adjustment and proposes a staged process model. Contrasting the model proposed in this study with Wallander et al.'s (1989) model it is apparent that the social challenges faced by these families could be a risk to adjustment and this was not acknowledged as a risk in their model, while issues such as functional independence were less relevant for the parents in this study. However, it is acknowledged that his model was based on research into chronic health conditions and so this difference may reflect the different challenges faced by the parents in the current study.

The central component of the model described in this study has similarities to the Crisis model proposed by Moos & Schaefer (1984). Although the Crisis model is conceptualised for people who are newly diagnosed with a health condition it will be discussed as it is consistent with central aspects of the current study's model. The Crisis model is a conceptual model that describes three factors that influence the outcome of the crisis and are mediated by the coping processes of the person. These factors are firstly, illness related factors; the risk that the illness poses including if it is disfiguring, disabling, painful or life-threatening. Secondly, personal factors and background are considered; whether the person has the experience, skills and resilience to draw upon. Personality factors such as optimism and pessimism and quality of life play a role in this factor. Thirdly, physical, social and environmental factors; the physical problems that the illness may cause the person such as mobility or functional problems and the person's social support network and it is acknowledged that a person's support network can undermine coping if they are experienced to be unhelpful or if they give inaccurate advice. The Crisis model conceptualises that these factors influence a process of coping that involves the cognitive appraisal (perceived meaning of the illness),

adaptive tasks and coping skills as the factors that produce the outcome of the crisis. This process is consistent with the central component of the model in this study; parents seeking out the information they need to understand, and to consider the personal meaning of their child having a birthmark and drawing on their resources to seek support and plan ahead for the challenges they may face.

Moving to look at appearance specific literature, it is evident that social challenges and the impact that social and cultural appearance norms have are widely recognised as a challenge for people with visible difference and receive greater acknowledgement than in chronic health literature. Rumsey & Harcourt (2005, 2012) recognise that there has been a tendency to rebuff the use of specific models of adjustment to visible difference because of the multitude of factors that are involved and the challenge in being able to capture some of these dimensions satisfactorily. However, certain models have been used to underpin research. Leventhal's self-regulatory model (Leventhal, Meyer & Nerenz, 1980) has been used within appearance research as it is reasonable to consider that a person's beliefs about the consequences, cause, duration, seriousness of a visible difference might influence the distress or behaviour of the individual. Although this model considered the experiences of the person with the visible difference, comparing these factors with the experiences of the parents in this study it is evident that these factors are equally as present in parental distress and behaviour and therefore it is likely that they affect the overall adjustment of parents as well.

### 7.5 Clinical implications arising from this study

This was the first qualitative study that has explored parental adjustment following the birth of a child with a facial birthmark and drew on the experiences of parents whose children had different types of birthmarks. It is also the first study in this specific field that utilised a GT approach for data analysis. The findings highlight the need for professionals to be more aware of the parents' stage of adjustment and how they can support parents in

accessing appropriate resources. As a minimum, professionals need to be made more aware of charitable organisations such as the Birthmark Support Group and Changing Faces as this would enable them to signpost parents and empower them to interact with other parents who are facing the same issues, thus normalising the uncertainty, worries and challenges that they may face. More preferably, while I acknowledge the scale of intervention this would require, the implementation of standardized training for all healthcare professionals who come into contact with a child with a facial birthmark, or even more broadly children with a facial difference would be important to raises the awareness of these conditions. This could provide healthcare professionals with the knowledge and confidence to recognise these conditions and sensitively support the families. Building on this further, developing an appropriate pathway to ensure parents are able to access the correct medical advice for their children could be highly advantageous, reducing the uncertainty for the parents and supporting them to gain understanding of the condition in a timely manner. Promoting consultation with professionals who work in specialist centres could be an alternative way to access accurate information for professionals. This would be particularly important for parents who have certain beliefs about the aetiology of the birthmark such as the mothers in this study who quickly blamed themselves and felt responsible as it is thought that these feelings increased the distress they experienced.

Furthermore, the professionals who make up the multi-disciplinary teams who support these families should include Clinical Psychologists as it is clear from the literature that living with a facial difference has psycho-social consequences. This research highlights that parents begin to worry about these consequences very early in the child's life and that this concern is a factor when considering treatment options and so providing psychological support and advice early in the process could contribute to parental adjustment and support them in making decisions about treatment and potentially support that attachment relationship between parent and child. Many parents were concerned about the future possibility of their child being bullied because of their appearance and the parents had different ideas about

how to manage this. Unfortunately, research indicates that bullying is often a concern for those who have a visible difference (Rumsey & Harcourt, 2005, 2012). While some parents had their own ideas about how to support their children, none of the parents spoke of being offered any professional support or advice around how to manage this. This is of significant importance because the parents' perception of the child's appearance and own resourcefulness may influence the child's adjustment and longer term self-esteem and psycho-social functioning.

A further cause of concern for parents was how to talk about the birthmark with their child. Some had accessed story-books and teddy bears from charities while others were unaware of such resources. As such, it would be useful for organisations and specialists working in this field to promote such resources for parents. While some of the parents encounter milder reactions from members of the public, others had experienced unpleasant interactions that had left them feeling distressed. Some parents had planned ahead and had naturally begun to formulate some responses for themselves, while others had not. Following on from this, a recommendation is that parents may benefit from information, such as a booklet that offers support and advice about the range of reactions they may experience from the public and advice about how to respond. This may aid them in understanding why this can happen and help them to develop some useful coping strategies that, in turn, may reduce the impact of negative social interactions.

The Birthmark Support Group and other support groups were recognised by all of the mothers and the majority of fathers to be a significant asset to them. Speaking to others in the same or similar situations helped allay their concerns and offered a platform to gain advice and learn about helpful coping strategies. Additionally, the support group offered parents a place to talk about their experiences and they described their participation in the group and in this research project as an opportunity to give back to others. However, one consideration for these support groups is about the moderation of the comments and advice on the website. This was raised by one of the fathers and the Clinical Nurse Specialist who were both concerned about advice

being given by parents who have lived experience but are not professionals accidentally offering inaccurate advice to other parents. It may be that greater moderation of the comments is required, or that a disclaimer should be used which would highlight this fact on these groups.

#### 7.6 Methodological considerations: strengths

The participants were recruited using a UK based international support group and although all of the participants were based in the UK, the participants were recruited from a wide geographical area. GT seeks to gain a wide variety of experiences within sample, so geographical variation was particularly advantageous. As such, the conclusions drawn from this study are not limited to a particular region or to one NHS Trust. However, it is acknowledged that the participants who took part were all White British and so the model's ability to provide insight to other cultures is limited. Given the experiences discussed, it may be that variation in cultural norms and appearance norms is an additional factor to consider and requires further research.

Peel, Parry, Douglas & Lawton (2006) reflected that within research it may be people who reflect the extreme ends of the scale who wish to talk about their experiences and so it may be that this is reflected in the model than has been constructed in this research project. However, this is a novel population who may not have had the opportunity to discuss or to reflect on their experiences and this may have contributed to their motivation to participate. This would explain the high response rates to the adverts for the project. Both mothers and fathers were recruited to take part in this study and so, in contrast to much of the data in the field of visible difference, this study included the father's perspectives as well as the mothers. Although, for the most part parents expressed similar views, it felt important to consider both perspectives.

A further strength of the methodology, given the novelty of this research was in the use of an approach that allowed in-depth exploration of participants' experiences. Each interview was carefully analysed to ensure depth of

analysis in the coding process, as this was crucial for maintaining the rigour of the study. Although I was new to GT and to qualitative analysis more generally, I endeavoured to ensure the quality of the research by reading about GT and seeking regular supervision with my supervisor who is an experienced GT researcher in addition to the teaching we received as part of the research module. As previously described, the quality and transparency of the analysis was supported by peer review in the form of two audits of analysis undertaken by each of my supervisors, who are both clinical psychologists, one with expertise in GT research and health psychology and the other who has expertise in the field of child health psychology and has prior experience of using qualitative methods in research.

#### 7.7 Methodological considerations: limitations

One critique of the methodology is that all of the participants were recruited from the Birthmark Support Group. Considering that support, including help from support groups were important features in supporting parents to adjust to their child's birthmark, it may be that there are parents who have not found out about, or do not have access to this source of support who may be having different experiences. Additionally, it may be that the parents who responded to the adverts reflect a particular subset of parents while other parents may not have come forward. For example, parents who were finding it very difficult to cope, or the opposite, not feeling the need to seek support, may not have wanted to take part. A second critique is that despite making multiple attempts to recruit a negative case; someone who had actively chosen not to treat their child's birthmark, this was not possible. Again, this may be a reflection on the use of a support group as the means of recruitment.

A second limitation of this study was time. Although it is felt that a sample of 13 participants and a further interview with a professional working in the field for the purpose of working towards theoretical saturation is sufficient for the requirements of this project, this scale did not allow for the full saturation of the categories which would have increased the robustness of this study.

Consequently, the findings of this study should be viewed more in terms of a

pilot study and further larger scale research with a greater number of participants recruited through multiple routes such as through the NHS as well as the voluntary sector would be required to corroborate, develop and refine this model.

Although the interview guide was viewed and critiqued by parents with lived experience, another improvement to the methodology may have been to conduct a pilot interview to develop it further. The question “tell me a bit about yourself and your family” was the first question and was used to open the conversation and provide some context about the participant however, it elicited some confusion in a few participants, who seemed unsure how to answer the question. It may be that the question could have been worded differently, and that the need to rephrase that question might have been noted in a pilot interview. It is acknowledged that the interview guide will have had some effect on the answers elicited from participants and therefore will have impacted on the construction of the categories and in turn, the model as it was guiding the interview. However, the interview guide was used flexibly with additional questions being formulated in response to the answers given ensuring a genuine attempt to explore the underlying issues involved.

### 7.8 Suggestions for further research

Conducting a larger scale research project could continue to develop the categories identified and this would help to develop a greater understanding about the issues this population face. Furthermore, incorporating greater cultural diversity in to the sample would help to evolve this model by considering additional factors that may have been under represented in this group of participants. Alternatively, it could be interesting to conduct a study in its own right that focuses on how people from non-white British cultures make sense of visible difference.

Conducting a research project that compares parental adjustment to facial birthmarks with birthmarks located elsewhere on the body could help to ascertain whether the visibility of the birthmark plays a role in the parents’

response to the birthmark and to their decision making process about whether or not to treat the birthmark.

Additionally, other research has suggested that parental appearance consciousness can play a role in adjustment. Although this concept did not emerge in the participant interviews, it was not explicitly asked about. Therefore, further research could explore whether parental appearance consciousness is a mediating factor in the adjustment process.

Repeating this study with parents of older children, perhaps at times of transition such as when they start school, could help promote understanding of how this adjustment process impacts on the child. It would allow for a greater understanding about the factors that influence the child's acceptance of their birthmark as they become old enough to recognise their visible difference. It could explore whether parental adjustment to the birthmark impacts on how the child makes sense of their birthmark and how they cope with the challenges they may face. Another study could explore whether the results of this study could have applicability in similar populations such as CL/P or CFA.

#### 7.10 Study reflections

Reflexivity is essential in qualitative research in order to enable the researcher to consider their own values, interest and assumptions and how these influence the analysis. I therefore kept a reflective journal (appendix I) and undertook regular research supervision and used memos in line with GT research guidelines. Keeping a reflective journal and considering my position as a psychologist and as someone with lived experience was crucial given the influence I had as the primary data analyst and a co-constructor of the model. Given my position as someone with lived experience, I have my own experiences and perceptions about living with a visible difference and so I tried to remain aware of my position for example if it was making me more or less curious about certain issues that were raised or if similarities in the

language we used may have meant that the meaning of words the participants used were taken for granted.

I was quite overwhelmed by the positive response to the adverts to participate in the project and I was struck by the warm welcome I received by all of the parents, inviting me into their homes. Through conducting interviews, I have developed my interview skills and by listening to the recordings and transcribing the data I was able to learn more about my interview style. I noticed that at times I would change the phrasing of my question part way through asking the question and this could confuse the participant. On other occasions I would ask two questions at the same time which could result in the second part of the question not being answered if the first part of the question led in a new direction. During the interviews parents were sharing their experiences, thoughts and beliefs and I was always aware of my own facial scars and how the presence of these, if they noticed them may have been affecting their responses to my questions, for example, if they were trying not to say something that I might find offensive. I tried to manage this by naming it and putting each participant at ease, letting them know that I valued their experiences and opinions and promoted that they could talk however they wanted to. Also, I was aware that the flexibility of using an interview guide as opposed to a more structured interview schedule gave me greater scope to be flexible about the questions. It was a privilege to hear their experiences and I felt a huge responsibility to do justice to their experiences when I was analysing the data and I constantly questioned whether I had sufficiently captured the essence of their experience. I was grateful for the iterative process of GT in that respect as I was able to code and re-code where necessary and use in vivo codes to ensure I had captured the essence.

I also considered the emotional impact that studying this topic may have for me and found myself to be touched by their experiences, but no more than other's experiences, such as those that I encounter in clinical practice. I felt quite surprised by this and spent a great deal of time reflecting on this. Was it that I was exploring the parental adjustment rather than lived experience that created an emotional distance or was I closing myself off emotionally from the

subject matter? I was aware of the impact this would have on my analysis and so felt it was crucial to ensure this was not the case. I concluded that it was related to no longer perceiving myself to have a birthmark, instead I see myself as scarred which, although a facial difference, feels different to having a birthmark. I also noticed that I shared the parents' frustration at the medical system as the parents I spoke to were facing many of the same challenges such as the poor awareness of birthmarks that my parents faced nearly 30 years ago despite the developments in technology and increased access to resources such as through the internet.

#### *7.10.1 Parents as consultants on the project*

I have included the account below of my parents input to the project as a means of thinking through some of the ethical issues that this brought up, and to consider the decision making process which led to this. My decision to use my own parents as consultants on the project was not one decision, but a series of decisions which occurred as the project unfolded. I knew early on that I wanted to consult with people with lived experience of parenting a child with a birthmark, in order to ensure that the design of my study, and in particular, my interview schedule was relevant and applicable to my research. At the outset, I intended to recruit a family to take this position, however, time constraints made this difficult and I was not able to recruit anyone to this position. I therefore turned to my parents to take this role. Looking back at this decision, I think I should have taken more time to think and reflect on the implications of involving them in my project in this way. It was in many ways, a very natural thing for me to do, as they have always been such a huge part of my own journey of being born with a birthmark, so conversations around this topic were frequent, open and honest throughout my childhood, adolescence and into adulthood.

I did not however act hastily, and explicitly considered issues around confidentiality and anonymity of participants, as well as more personal factors before asking them to have any part of the research. These factors continued

to be implicit in my thinking, at all points in the process. When they were invited to be consultants on the study I considered whether there was potential for it to be upsetting for them or for myself if I found it difficult to hear their open opinions and so we spoke openly and explored this and felt that, given the openness we have always had in regards to my birthmark, this would not be a problem. I also took steps to ensure that I defined their role each time I approached them in order to differentiate between their role as my parents, trying to support and care for me, and the advisory role that I was asking them to step into as consultants on my project. I'm clear that involving them had no unconscious motivation, for example, to attempt to talk to them about aspects of my own experience that had been 'unspoken' or to resolve difficulties in our relationship. It really was more of a pragmatic decision, and I was very aware that they would have useful contributions to make, and in light of time constraints, did not see the complexities of involving them.

However, on reflection, were time to have allowed, making the time to build rapport with another family with lived experience as I had initially planned to, and then using them as consultants on the project may have offered a similar richness to the research process while maintaining a greater personal distance. In turn, this could have reduced any potential complications or bias that the personal relationship I have with my parents could have introduced.

Initially I had only intended to consult with them at the planning stages, during the development of my interview guide. At this point they were asked to review the draft of the questions as it was considered that they may be able to suggest topics or areas to consider that I may not have thought about. They felt that I had already considered the majority of issues that they thought about although one area to explore that I had not thought about was the reactions of the medical professionals and how medical professionals and medical input affected the parents' adjustment. This was incorporated into the interview guide and I later reflected on how useful this had been when it emerged during the literature review in other studies about parental adjustment. This consultation also led me to consider whether to interview parents as dyads or separately. It was suggested that I may achieve richer

data by interviewing parents separately as it was possible that interviewing them together could perhaps lead to less depth in the responses or avoidance of some topics if they were trying to protect one another's feelings.

I had not planned to use my parents again in this role, however during the analysis I thought that a 'lived experience perspective' could help to enrich my understanding of a few concepts in the data. So I approached my parents again after consideration of the possible issues of doing this. During the consultations my parents saw some anonymised quotes (from my analysis table) to help me to explain the essence of an emerging hypothesis and later they saw a draft of the model. I was conscious of ensuring the confidentiality and anonymity of the participants throughout the consultations. On reflection, the structured way in which the consultations were approached created a boundary, for example, I led with an idea or hypothesis emerging from the data and asked for their reflections. While this was implemented as an approach to allow us to stay on task, this also limited the scope or depth of the consultations.

My parents reflected on the use of technology and how much the internet as well as advances in medical technology has influenced the ability for parents to access support. They considered how this has enabled parents today to find out about the birthmarks themselves, access personal support, professional support and to connect with other families in similar situations, something that was difficult for them. We also thought about the medical advances that have led to new treatment options. I wrote memos on technology and medical advances following this consultation. This allowed me to reflect on and consider wider issues that may have influenced the parental adjustment process. They were then consulted a final time and asked to review the final draft of the model. At this stage they reflected that the model was reasonably consistent with their experience and commented on, in their experience, how much the ability to access resources was a key component for coping.

I used my reflective diary and supervision to think about the personal meaning this topic had for myself but, on reflection, I should have given greater

consideration to having my parents consult on the project given the personal resonance the topic has. Thinking about why this did not happen with my supervisor, we reflected that the supervisions were generally task focussed and we did not give much space to reflection. Again, it was felt that this was an oversight which resulted from a combination of factors including time constraints, this being my first time using the GT method so our supervisions were often task and method focussed and we felt that we were trying to maintain a momentum that we had generated which allowed me to complete this project in eight months. In hindsight, it was very important for us to have made time for reflection.

Following the completion of the research I have spent time reflecting personally and with my supervisor about the choice to use my parents in a consultative role on the project. Reflecting on the impact their involvement may have had on the research itself, one potential difficulty of involving my parents is that it may have influenced my perception or the way I construed the data. This could result from the interplay between my own lived experience and our joint experience and the narratives of my parent's experiences. Through this further consideration, I accept that this may have introduced a personal bias into the research. I acknowledge that this should have been thought about in more detail, including further consideration with my research supervisor. Taking this time to reflect on this project has reminded me of the importance to take the time to reflect more deeply and particularly at times of high pressure, stress or when under time constraints to ensure that oversights or mistakes are not made. This is an important lesson to learn and I will take this forward from this project and be more aware of personally and professionally.

### 7.11 Conclusion

The current research project has made a novel contribution to the currently scarce research and literature on parental adjustment following the birth of a child with a facial birthmark. The findings of this study are consistent with previous research on parental adjustment following the birth of a child with a

facial visible difference. However, the findings were able to further elaborate the process of adjustment, describing the processes involved in adjustment for this population. Recommendations include greater training and awareness for health professionals who come into contact with the families and greater psychological input at an early stage to support parental adjustment and to offer advice about the psycho-social impact of having a birthmark.

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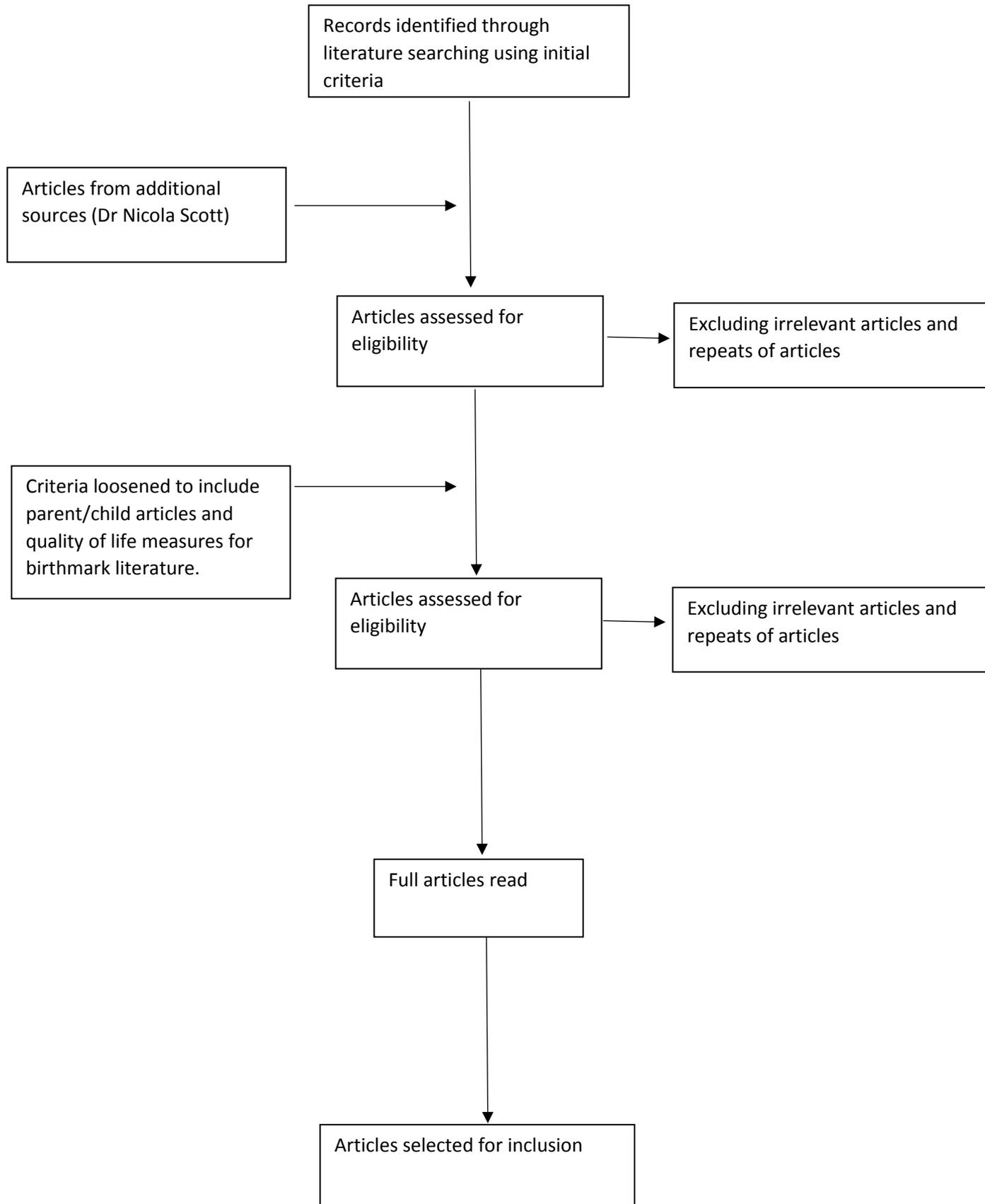
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## **Appendices**

Contents of the appendices

- A. Literature Review
  - i. Flow chart of papers selected
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- F. Analysis audit trail
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Appendix Ai: A flow chart demonstrating how papers were selected for the literature review



Appendix B: Original interview guide

Guiding questions

1. Tell me a bit about yourself/your child/your family (any other children etc.?)
2. How did you feel when you first found out about the birthmark? (Do you have a clear diagnosis; do you understand what the long term prognosis is?)

Prompts

Areas to consider:

Reactions: family/ friends/ public/ hospital staff (immediate and longer term)

Challenges/lessons/coping/support (types of support/usefulness)

Relationships – with child, inter-sibling, marital, social/peers

Things for the future - What have you learned from these experiences? – What would you pass on to others?

Appendix C: Participant information sheet

## **PARTICIPANT INFORMATION SHEET**

### **Parental adjustment after the birth of their child with a facial birthmark**

#### **Introduction**

You are being invited to take part in a research study. Before you decide whether to do so, it is important that you understand the research that is being done and what your involvement will include. Please take the time to read the following information carefully and discuss it with others if you wish. Do not hesitate to ask us anything that is not clear or for any further information you would like to help you make your decision. Please do take your time to decide whether or not you wish to take part. Thank you for reading this.

#### **What is the purpose of this study?**

My name is Lucy Gait, I am a third year Trainee Clinical Psychologist from the University of Hertfordshire and I am conducting this research for my 3rd year Doctoral research project.

The purpose of this study is to explore the experiences of parents who have a child who has a congenital facial birthmark. The hope is that this research will be able to inform professionals who work in this area and improve services and support.

#### **Do I have to take part?**

It is completely up to you whether or not you decide to take part in this study. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. Agreeing to join the study does not mean that you have to complete it. You are free to withdraw at any stage without giving a reason. A decision to withdraw at any time, or a decision not to take part at all, will not affect any part of your care or the support you receive.

#### **What will happen to me if I take part?**

If you decide to take part in this study, you will be asked to take part in a semi-structured interview with myself that will last for approximately one hour. The interview can take place at a location of your choice. The questions that you will be asked will relate to your experiences since the birth of your child and your thoughts about the future.

If there were any significant changes to the method, you would be informed and asked to re-consent.

#### **What are the possible advantages and disadvantages of taking part?**

By choosing whether or not to take part will not affect your opportunities or future support. Agreeing to take part will provide you with an opportunity to share your experiences and for psychologists, clinicians, researchers and organisations to

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strengthen and deepen their understanding of the impact facial birthmarks has on parents and families.

### **How will my taking part in this study be kept confidential?**

The interviews will take place at a time and place of your choosing. Your interview will be recorded and kept in encrypted and password protected files to ensure confidentiality. Once the data is transcribed, it will be kept in locked files and your transcript will be assigned a unique code. In the research write up, you will be referred to by another name and any information contained in quotes will be anonymized to ensure your confidentiality is maintained.

### **What will happen to the results of the research study?**

The results from the study will be written up in a doctoral thesis for the purposes of gaining the Doctorate in Clinical Psychology qualification. It is hoped that the findings will be shared in academic publications and/or presentations however, any information that is made public following this research will be fully anonymised.

The data collected in the study will be kept securely for 5 years at the University of Hertfordshire years in accordance with the policy for awarding the Doctorate of Clinical Psychology to the researcher and for the purposes of publication. If the researcher wishes to use the data for any purpose, other than the study outlined in this information sheet, you will be informed and additional consent will be sought from you.

### **Who has reviewed this study?**

The study has been reviewed by The University of Hertfordshire Health and Human Sciences Ethics Committee with Delegated Authority [Protocol Number: aLMS/PGR/UH/02105(2)] to protect your safety, rights and well-being. This study has been reviewed and can be conducted as it has been granted favourable opinion with no raised objections on ethical grounds.

As this research project is also part of the academic commitment, it has been formally and peer reviewed by the University of Hertfordshire's Doctoral Programme in Clinical Psychology.

### **Who is supervising the project?**

The project is being supervised by Dr Helen Ellis- Caird (Clinical Psychologist and Research Lead at University of Hertfordshire) and Dr Helen Davies (Clinical Psychologist at Luton Paediatric Liaison Service).

### **Who can I contact if I have any questions?**

If you have any additional questions or any concerns or complaints during the study, please contact the principal investigator (details below) or research supervisor (details below).

**Lucy Gait –** [REDACTED]

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[Redacted]

Dr Helen Ellis-Caird

[Redacted]

Dr Helen Davies -

[Redacted]

**Thank you very much for reading this information and giving consideration to taking part in this study.**

Appendix D: Consent form

**University of Hertfordshire  
CONSENT FORM**

**Parental adjustment after the birth of their child with a facial  
birthmark**

I, the undersigned [*please give your name here, in BLOCK CAPITALS*]

.....  
.....  
of [*please give contact details here, sufficient to enable the investigator to get in touch with you, such as a postal or email address*]

.....  
.....  
hereby freely agree to take part in the study entitled [*insert name of study here*]

.....  
.....

**1** I confirm that I have been given a Participant Information Sheet (a copy of which is attached to this form) giving particulars of the study, including its aim(s), methods and design, the names and contact details of key people and, as appropriate, the risks and potential benefits, and any plans for follow-up studies that might involve further approaches to participants. I have been given details of my involvement in the study. I have been told that in the event of any significant change to the aim(s) or design of the study I will be informed, and asked to renew my consent to participate in it.

**2** I have been assured that I may withdraw from the study at any time without disadvantage or having to give a reason.

**3** I have been given information about the advantages and possible disadvantages of taking part in the study.

**4** I have been told how information relating to me (data obtained in the course of the study, and data provided by me about myself) will be handled: how it will be kept secure, who will have access to it, and how it will or may be used.

Signature of participant.....

Date.....

Signature of (principal) investigator.....

Date.....

Name of (principal) investigator [*in BLOCK CAPITALS please*]

.....  
.....

Appendix Ei:

UNIVERSITY OF HERTFORDSHIRE

## FORM EC1: APPLICATION FOR ETHICAL APPROVAL OF A STUDY INVOLVING HUMAN PARTICIPANTS

(See Guidance Notes)

<p><b><u>Relevant ECDA:</u></b></p> <p>Science &amp; Technology <input type="checkbox"/></p> <p style="text-align: center;">x</p> <p>Health &amp; Human Sciences <input type="checkbox"/></p>	<p style="text-align: center;"><b>OFFICE USE ONLY</b></p> <p><b><u>Protocol Number:</u></b></p>
---	---

Office Use only	Date Received by Clerk:
<b>Expedited Review</b>	
Approved by Reviewer 1 ( <i>sign &amp; date</i> )	Approved by Reviewer 2 ( <i>sign &amp; date</i> )
<b>Further Action:</b> ( <i>tick appropriate box and provide details</i> )	
Request Further Information <input type="checkbox"/>	<u>Details:</u>
Refer for Substantive Review <input type="checkbox"/>	
Refer for Full review <input type="checkbox"/>	
Reject <input type="checkbox"/>	
<b>Substantive Review</b>	
Approved by Reviewer 1 ( <i>sign &amp; date</i> )	Approved by Reviewer 2 ( <i>sign &amp; date</i> )
Approved by Reviewer 3 ( <i>sign &amp; date</i> )	Approved by Reviewer 4 ( <i>sign &amp; date</i> )
<b>Further Action:</b> ( <i>tick appropriate box and provide details</i> )	
Request Further Information <input type="checkbox"/>	<u>Details:</u>
Refer for Full review <input type="checkbox"/>	
Reject <input type="checkbox"/>	
<b>Full Review</b>	
Request Further Information <input type="checkbox"/>	<u>Details:</u>
Reject <input type="checkbox"/>	
<b>CONFIRMATION OF APPROVAL</b>	
[To be completed by the Chairman or Vice-Chairman of the relevant ECDA, or by the Chairman of the University Ethics Committee – (see GN 2.1.4)]	
I confirm that this application has been approved by or on behalf of the committee named below.	

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Name/Sign.....Date.....

Name of committee .....

## DECLARATIONS

### DECLARATION BY APPLICANT (See GN 2.1.3)

- (i) I undertake, to the best of my ability, to abide by accepted ethical principles in carrying out the study.
- (ii) I undertake to explain the nature of the study and all possible risks to potential participants, to the extent required to comply with both the letter and the spirit of my replies to the foregoing questions (including information contained in Appendices 1 & 2).
- (iii) Data relating to participants will be handled with great care. No data relating to named or identifiable participants will be passed on to others without the written consent of the participants concerned, unless they have already consented to such sharing of data when they agreed to take part in the study.
- (iv) All participants will be informed **(a)** that they are not obliged to take part in the study, and **(b)** that they may withdraw at any time without disadvantage or having to give a reason.

Where the participant is a minor or is otherwise unable, for any reason, to give full consent on their own, references here to participants being given an explanation or information, or being asked to give their consent, are to be understood as referring to the person giving consent on their behalf. (See Q 19 above; also GN Pt. 3, and especially 3.6 & 3.7)

Enter your name here.....Lucy Gait.....Date  
.....10/10/2015.....

**If you are a member of staff, please obtain the signature of your line manager to indicate their agreement to this application:**

.....

(Signature)

(Name in BLOCK  
CAPITALS and  
position within the School)

### DECLARATION BY SUPERVISOR (see GN 2.1.3)

I confirm that the proposed study has been appropriately vetted within the School in respect of its aims and methods as a piece of research; that I have discussed this application for Ethics Committee approval with the applicant and approve its submission; and that I accept responsibility for guiding the applicant so as to ensure compliance with the terms of the protocol and with any

applicable ethical code(s).
Enter your name here.....Clare Norris.....Date.....

## Instructions for Applicants

**Applicants are advised to read the Guidance Notes before completing this form.** Use of this form is mandatory [see UPR RE01, SS 7.1 to 7.3]. Your School may choose to add additional School specific questions in a separate appendix to the Form EC1. Please clarify with your Supervisor whether your School has chosen to append any extra questions.

Approval must be sought **and granted** before any investigation involving human participants begins [UPR RE01, S 4.4(iii)].

### Abbreviations

GN=Guidance Notes  
UPR=University Policies & Regulations  
Q=Question  
S=Section  
SS=Sections  
Pt =Part

**PLEASE NOTE:** Where alternative answers are offered, put a cross in the appropriate box.

For example:  YES

Where a "write in" answer is requested, begin in the space provided below the question and continue as necessary. **All questions must be answered. Please answer in BLACK.**

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## 1. THE STUDY

**Q1.** Please give the title (or provisional title) of the proposed study. (NB – you will be asked for further details later)

**How do parents adjust after the birth of their child with a facial birthmark.**

## 2. THE APPLICANT

**Q2.** Please answer either **Q2.1** or **Q2.2** by providing the information requested.  
**Q2.1** should be answered by individual applicants, both staff and students, who require protocol approval for work which they themselves intend to carry out.  
**Q2.2** should be answered by academic staff requiring approval for standard protocols governing classroom practical work (or equivalent work) to be carried out by a specified group of students. (See GN 2.2.1 & 2.2.19)

**Q2.1.** Name of applicant/(principal) researcher

Status:

(a) undergraduate

(b) postgraduate (taught/research)

(c) academic staff

(d) other - please give details here

School/Department: Doctorate in Clinical Psychology

Programme of study or award (e.g.. BA/MSc/PhD/Staff research):  
Doctorate in Clinical Psychology

E-mail address: [l.gait@herts.ac.uk](mailto:l.gait@herts.ac.uk)

Name of supervisor : Clare Norris

Supervisor's contact details (email, extension number):  
[c.norris@herts.ac.uk](mailto:c.norris@herts.ac.uk)

**Q2.2. Class Protocol Applications Only.**

Name of applicant/(principal) researcher (member of staff):

School/Department

Programme of study or award (e.g.. BSc/MA)

Module Title

Year/group to be governed by the protocol

Number of students conducting the study

Programme Tutor (if different from the applicant)

E-mail address

**Please note: Risk Assessment Form EC5 is mandatory for all Class Protocol Applications and must accompany this application.**

**3. DETAILS OF THE PROPOSED STUDY**

**Q3. (a)** Is it likely that your application will require NHS approval? (See GN 2.2.2)

YES

NO

(If YES, please answer **(b)** & **(c)**  
on to **Q4**)

(If NO, please continue

**(b)** Please confirm whether your research involves any of the following:

<input type="checkbox"/>	NHS Patients	<input type="checkbox"/>
<input type="checkbox"/>	Clinical trial of an investigational product	<input type="checkbox"/>
<input type="checkbox"/>	Clinical trial of a medical device	<input type="checkbox"/>
<input type="checkbox"/>	Exposure to any ionising radiation	<input type="checkbox"/>
<input type="checkbox"/>	Adults who lack the capacity to consent	<input type="checkbox"/>
<input type="checkbox"/>	Human Tissue	<input type="checkbox"/>
<p><b>(c)</b> Please confirm whether this study is considered to be a Clinical Trial of Investigational Medical Products (CTIMP) or Clinical Trial of Investigational Medical Devices. (See GN 2.2.2)</p>		
<input type="checkbox"/>	YES	<input checked="" type="checkbox"/> NO
<p>If YES, please indicate if the study involves any of the following categories:</p>		
<input type="checkbox"/>	Children under 5	<input type="checkbox"/>
<input type="checkbox"/>	Pregnant women	<input type="checkbox"/>
<input type="checkbox"/>	A group of more than 5,000 people	<input type="checkbox"/>
<input type="checkbox"/>	Study would be undertaken overseas	<input type="checkbox"/>
<p><b>If your study is likely to require NHS approval or is a Clinical Trial of Investigational Medical Products or Devices, DO NOT complete this form any further and submit it to your relevant ECDA at this stage. All NHS applications must be made on an IRAS form. If your study is a Clinical Trial of Investigational Medical Products or Devices involving one of the above specified categories, you will be contacted by the relevant ECDA with information on the next steps. Please note, you will be issued with a UH Protocol Number but this will not be valid until you have sent your relevant ECDA a copy of your NHS approval.</b></p>		

**Q4.** Please give a short synopsis of your proposed study; stating its aims and highlighting, if appropriate, where these aims relate to the use of human participants. (See GN 2.2.3)  
Please enter details here.

#### Psycho-social impact of disfigurement

The psycho-social impact of 'disfigurement' also referred to as 'difference' has been an area of research interest for the past 40 years. Much of this research has explored the experience of those who have congenital or acquired disfigurement and focused on adjustment, coping and associated emotional wellbeing.

The research conducted within this field and more broadly in appearance literature falls in to one of two interconnected categories as described by Cash, (1990). The first perspective is that of the outsider. Studies have looked at the social and cultural impact of disfigurement and how this is perceived within society. The second perspective is the internal experience of the individual and the impact that living with a disfigurement has on self-concept, emotional wellbeing and overall quality of life. Within the literature, individuals with a visible difference are frequently reported to experience difficulties in forming relationships both socially and romantically and to frequently experience negative reactions from others, including stares and in some cases rude or intrusive comments and discrimination (Porter et al., 1986; Porter et al., 1987; Porter et al., 1990; Jowett & Ryan, 1985; Lanigan & Cotterill, 1989 and Van der Donk et al., 1994). Consequently, it is not surprising that studies into the psycho-social impact of disfigurement conclude that many individuals experience higher than average levels of psychological distress, including anxiety, depression, and low self-esteem (Lansdown et al., 1997).

However, more positively, research also indicates that with appropriate support people with congenital or acquired disfigurement can adapt and learn to cope. Social support, lower appearance consciousness and greater personal resilience have been found to be associated with better adjustment and coping (Prior & O'Dell, 2009, Roberts & Shute, 2012).

#### Impact for children & families

For children, family acceptance is recognised to be crucial for their psychosocial development and higher levels of social support have been found to relate to better adjustment following injury or trauma (Wills, 1997; Blakeney et al., 1988). Kalick et al.

(1981) found low levels of psychological morbidity in their sample of patients who had port wine stain birthmarks was related to higher levels of family support. However, the literature suggests that parents of disfigured children can be less nurturing and more distant than parents of non-disfigured children (Barden et al. 1989) and other researchers have demonstrated how the presence of a disfigurement has a negative impact on attachment (Barden et al., 1989; Bradbury & Hewison, 1994, Campis et al., 1995). Researchers suggest that appearance consciousness may develop in early childhood where parental emphasis has been placed on appearance (Leary & Kowalski, 1995). Campis et al. (1995), supports the suggestion that shame, which may be learned in childhood, forms an important and fundamental aspect of the problems that disfigured people can experience. This research suggests that interventions that aim to facilitate family support systems and assist with the parent's process of adjustment would be helpful both for the parent themselves and for the child's long term psychological wellbeing.

In line with this, a searched was conducted to find literature on the lived experience of parents with children who have disfigurement. Surprisingly, few studies were found. Of those found, three studies focussed on adjustment to cleft lip/palate, two focussed on the experiences of families with a child born with a haemangioma and two addressed the challenges faced by families with a child born with a craniofacial abnormality. Of these papers, five endorsed a qualitative methodology, two endorsed quantitative methods.

One qualitative study conducted by Tanner et al. (1998) looked at the experiences of adjustment for families and children born with a facial haemangioma. They discuss themes of parental emotion and adaptation; experiences with public reactions; issues related to parent-child interactions; and expressed satisfaction/dissatisfaction with medical care and concluded that facial haemangiomas in young children are frequently associated with parental reactions of loss and grief, despite the generally benign nature of the lesion and the prognosis for eventual involution. The other study into adjustment following the diagnosis of a haemangioma was conducted quantitatively and compared the location and severity of the haemangioma with quality of life and adjustment in children. They concluded that having a haemangioma, especially one with a complicated course or a visible location could result in psychosocial problems later in life, mostly related to physical appearance (Hoornweg et al. 2009).

The studies that addressed the impact of a cleft lip & palate concluded that throughout the childhood and adolescence, parents experienced conflicting emotions

about their child's impairment, uncertainty about cleft treatment, and stigmatizing attitudes from others. Some parents were found to attempt to manage emotional tensions by pursuing cleft treatments; however the interventions themselves were source of conflict and worry for them. Other themes of concern, functional difficulties related to eating and speech and social stigmatization were present (Nelson et al., 2012; Johansson & Ringsberg, 2004; Kuttenger et al., 2010).

Of the studies addressing craniofacial abnormality, one looked specifically at the father's perspective of having a child with a craniofacial abnormality (Klein et al., 2010). The result of these interviews indicated that most fathers felt highly optimistic about their children's potential for happiness and life success. However worry and concerns were expressed. All fathers described positive attributes in their children and they tended to support their children's peer relationships through encouraging and organizing activities with friends. The second study by Roberts & Shute (2012) endorsed thematic analysis of interviews with mothers and fathers of children with craniofacial abnormalities and revealed four major stress-related themes (self-acceptance, responses of others, disabilities and impairments, and treatment) and two positive themes included personal qualities and support. The findings of this study are in line with much of the literature written from the individual's perspective of living with a disfigurement.

Given how critical a child's early development and attachment relationships are acknowledged to be within clinical psychology, it is important to understand the challenges faced by parents whose children have disfigurements. More specifically, in the appearance conscious society of today, understanding the challenges facing families in adjusting to facial disfigurement could allow for the development of resources for families, psychologists and medical clinicians such as midwives, dermatologists and plastic surgeons.

This leads to the current research question:

How do new parents adjust to their child being born with a facial disfigurement such as a birthmark?

**Q5.** Please give a brief explanation of the design of the study and the methods and procedures used, highlighting in particular where these involve the use of human participants. You should clearly state the nature of the involvement the human participants will have in your proposed study and the extent of their commitment. Thus you must complete and attach the Form EC6 (Participant Information Sheet) (see Appendix 2). Be sure to provide sufficient detail for the Committee to be clear what is involved in the proposed study, particularly in relation to the

human participants. (See GN 2.2.4)

## Design

A Semi-structured interview schedule will be designed with consultation input from parents who have experience of this difficulty. Semi-structured interviews will then be conducted with 8-10 participants who will be parents of children aged 3-6 months with a facial birthmark.

Additionally in line with the process of Grounded Theory, once the interview data has been collected and analysed up to 12 months of data from the Birthmark Support Group forums and their Facebook page will be analysed in order to triangulate the themes from the interviews. Before analysing the data names will be changed to pseudonyms and details of location will be altered. The websites will be analysed using university computers in the university library and at home using a personal computer. For this triangulation process website content only will be analysed, participants will not be directly contacted.

## Procedure

Participants will be recruited through charity organisations such as the birthmark support group. Information sheets and consent forms will be given to participants. The interviews will last roughly one hour and will take place at a location of the participants' choice. The interviews will be recorded allowing the transcription of the data for analysis. The data will be transcribed and grounded theory will be used to analysis the data.

**Q6.** Please give the starting date and finishing date. (For meaning of "starting date" and "finishing date", see GN 2.2.5)

As soon as ethical approval is granted to 1<sup>st</sup> June, 2016.

**Q7.** Where will the study take place? (If this is on UH Campus, who will permission be obtained from e.g.. your Module Leader, Programme Tutor, Pro-Vice Chancellor (Student Experience) or the Dean of Students. If this is NOT on UH Campus, please attach a copy of the written permission, given by the proprietor, manager or other person with such authority over the premises, to use the premises for the purposes of carrying out this research (see Appendix 2)) (See GN 2.2.6)

Please enter details here.

The interviews with will take place at a location of the participants choice and the website data will be accessed and analysed in University computers and the researchers personal computer.

**Q8.** If the location is off campus, have you considered whether a risk assessment is necessary for the proposed location? (in respect of hazards/risks affecting both the participants and researchers) Please see **Form EC5** (see Appendix 2, which is an example of a risk assessment form.) Please use this example if a risk

assessment is necessary, and you have not been provided with a subject specific risk assessment form by your School or Supervisor. (See GN 2.2.7)

The interviews will take place in a location of the participants choice and website content only will be analysed, participants will not be directly contacted.

For full details please see attached Form EC5 and completed LMS risk assessment form.

**Q9. (a)** Will anyone other than yourself and the participants be present with you when conducting this study? (See GN 2.2.8)

YES

NO

If YES, please state the relationship between anyone else who is present other than the applicant and/or participants? (e.g.. health professional, parent/guardian)

**(b)** Will the proposed study be conducted in confidence? If NOT, what steps will be taken to ensure confidentiality of the participants' information. (See GN 2.2.8)

The study will be conducted in confidence and any electronically recorded data will be encrypted and password protected and will be kept in locked cabinets.

For the data triangulation, only the birthmark support group website will be used with the permission of the charity. Any identifying information will be changed or removed before the data is analysed to ensure confidentiality.

#### 4. HARMS, HAZARDS & RISKS

**Q10.** Will this study involve invasive procedures on the human participants? (See GN 2.2.9)

Yes

No

**Q13**

(If YES, please fill out **Appendix 1 –**

(If NO, answer **Q11, Q12,**

**Increased Hazards and Risks.**

**& Q14)**

Once this is complete, move on to **Q15)**

**Q11, Q12, Q13 & Q14 - NON INVASIVE STUDIES ONLY**

Note: You are advised to read GN 2.2.10, 2.2.11, 2.2.12 & 2.2.13 carefully before you answer the following questions.

**Q11.** Are there potential hazards to participant(s) and/or researcher(s) from the proposed study? (See 2.2.10)

YES

If YES,

Indicate their nature here.

Indicate here what precautions will be taken to avoid or minimise any adverse effects.

**Q12.** Will or could the study cause discomfort or distress of a mental or emotional character to participants and/or researcher(s)? (See NG 2.2.11)

YES

NO

If YES,

Indicate its nature here.

The risk is minimal, however, the information elicited by the participants may be sensitive in nature and this may be distressing for them to talk about. It may also be distressing for the researcher to hear distressing stories.

Indicate here what precautions will be taken to avoid or minimise such adverse effects.

The participant will be encouraged to keep themselves safe throughout the interview and that they only need to talk about things that they are comfortable to speak about. They will be informed that they can pause or end the interview at any point and they will be provided with a list of organisations such as changing faces and the birthmark support group they can contact if they feel distressed.

The researcher will be able to use her personal or professional support systems and supervision to manage any distress.

**Q13.** Will or could medical or other aftercare and/or support be needed by participants and/or researcher(s) as a result of the study? (See GN 2.2.12)

YES

NO

**Q14. (a)** If you have answered 'YES' to Q11, Q12 & Q13, please state here the previous experience (and/or any relevant training) of the supervisor (or academic member of staff applying for a standard protocol) of investigations involving the hazards, risks, discomfort or distress detailed in those answers. (See GN 2.2.13)

The researcher has personal experience of facial disfigurement and the supervising psychologists Dr Clare Norris and Dr Helen Ellis-Caird have experience of supporting research students and clinical experience of working with children, parents and families and Helen has experience in analysing data using Grounded Theory.

**(b)** Please describe in appropriate detail what you would do should the adverse effects or events which you believe could arise from your study, and which you have mentioned in your replies to the previous questions, occur. (See UPR RE01, S 2.3 (ii) and GN 2.2.13)

If the participant became distressed the interview would be paused and the researcher would offer support to the participant. They would be given the opportunity to take a break or end the interview.

## 5. ABOUT YOUR PARTICIPANTS

**Q15.** Please give a brief description of the kind of people you hope/intend to have as participants, for instance, a sample of the general population, University students, people affected by a particular medical condition, children aged 5 to 7, employees of a particular firm, people who support a particular political party.

Participants

Participants will be parents of children aged 3-6 months who were born with a facial birthmark.

**Q16.** Please state here approximately how many participants you hope will participate in your study.

Approximately 10 participants will be recruited to the study.

**Q17.** By completing this form, you are indicating that you are reasonably sure that you will be successful in obtaining the number of participants which you hope/intend to recruit. Please outline here how you intend to recruit them. (See GN 2.2.14)

The birthmark support group have agreed to support the process of recruitment and to advertise on their website and Facebook page. Participants will be invited to take part in the study and they will be able to opt in.

## 6. CONFIDENTIALITY AND CONSENT

[For guidance on issues relating to consent, see GN 2.2.15 & Pt. 3.]

**Q18.** Is it intended to seek informed consent from the participants?

YES

NO

(See UPR RE01, S 2.3 & 2.4 and GN 3.1)

If NO, please explain why it is considered unnecessary or impossible or otherwise inappropriate to seek informed consent.

If YES, please attach a copy of the Consent Form to be used (See Form EC3 & EC4 for reference and GN 3.2), or describe here how consent is to be obtained and recorded. The information you give must be sufficient to enable the Committee to understand exactly what it is that prospective participants are being asked to agree to.

**Q19.** If the participant is a minor (under 18 years of age), or is otherwise unable for any reason to give full consent on their own, state here whose consent will be obtained and how? (See especially GN 3.6 & 3.7)

**Q20.** Are personal data of any sort (such as name, age, gender, occupation, contact details or images) to be obtained from or in respect of any participant? (See GN 2.2.16)

YES

NO

If YES,

**(a)** Give details here of personal data to be gathered, and indicate how it will be stored.

Participants' confidentiality will be maintained throughout. All participant data and transcripts that might identify them, will be accessed only by the research team. Any such data/files will be anonymised by assigning each person a unique participant code and stored in a locked filing cabinet in the researcher's house or in a locked filing cabinet at the University of Hertfordshire. Consent forms will be stored in the same way. Any audio data will be password protected and the file will be encrypted. At the end of the study, all data will be stored at the University of Hertfordshire for up to 5 years for the purpose of publication. The participants will have consented to the above.

For the data triangulation, only the birthmark support group website will be used with the permission of the charity. Any identifying information will be changed or removed before the data is analysed to ensure confidentiality.

**(b)** State here what steps will be taken to prevent or regulate access to personal data beyond the immediate investigative team?

Personal data and consent forms will be stored in a lockable filing cabinet either at the principle researcher's home or the university. Any audio data will be password protected and the file will be encrypted. The identifiable information will be kept separate from the client's coded information and only the principle researcher will have access to the codes assigned to participants.

**(c)** Indicate here what assurances will be given to participants about the security of, and access to, personal data.

Participants will be assured that the transcripts will not be available to anyone outside of the research team, that the transcript data will be stored against a unique participant code, and that the only person who has access to both the unique participant code and identifying information will be the principle researcher.

**(d)** State here, as far as you are able to do so, how long personal data collected during the study will be retained, and what arrangements have been made for its secure storage.

At the end of the study, all data will be stored at the University of Hertfordshire, in a lockable cabinet, for up to 5 years for the purpose of publication.

**Q21.** Is it intended (or possible) that data might be used beyond the present study? (See GN 2.2.16)

<input type="checkbox"/>	YES	<input checked="" type="checkbox"/>	NO
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If YES, please give here an indication of the kind of further use that is intended (or which may be possible).

If NO, will the data be kept for a set period and then destroyed under secure conditions?

<input checked="" type="checkbox"/>	YES	<input type="checkbox"/>	NO
-------------------------------------	-----	--------------------------	----

If NO, please explain here why not.

**Q22.** If your study involves work with children and/or vulnerable adults you will require a satisfactory Enhanced Criminal Records Bureau Disclosure. (See GN 2.2.17) Please indicate as appropriate:

(a) CRB Disclosure not required	<input type="checkbox"/>
(b) CRB Disclosure required and obtained	<input checked="" type="checkbox"/>

If a satisfactory CRB Disclosure is required, a copy of this must be attached to Appendix 2 in order for reviewers to be able to consider your application.

## 7. REWARDS

**Q23. (a)** Are you receiving any financial or other reward connected with this study? (See UPR RE01, 2.3)

<input type="checkbox"/>	YES	<input checked="" type="checkbox"/>	NO
--------------------------	-----	-------------------------------------	----

If YES, give details here.

**(b)** Are participants going to receive any financial or other reward connected with the study?

YES  NO

If YES, give details here

**(c)** Will anybody else (including any other members of the investigative team) receive any financial or other reward connected with this study?

YES  NO

If YES, give details here.

## 8. OTHER RELEVANT MATTERS

**Q24.** Enter here anything else you want to say in support of your application, or which you believe may assist the Committee in reaching its decision.

## **APPENDIX 1 – INCREASED HAZARDS AND RISKS**

This section is to be completed if your answer to Q10 affirms the **USE OF INVASIVE PROCEDURES** in your study.

Note: You are advised to read GN 2.2.10, 2.2.11, 2.2.12, 2.2.13 & 2.2.18 carefully before you answer the following questions.

**QA1.** Please give details of the procedures to be used (e.g.. injection of a substance, insertion of a catheter, taking of a blood or saliva sample), and any harm, discomfort or distress that their use may cause to participants and/or researcher(s). (See GN 2.2.10)

**(a)** Indicate here what precautions will be taken to avoid or minimise any adverse effects.

**QA2.** Will the study involve the administration of any substance(s)? (See GN 2.2.10)

YES

NO

If YES,

**(a)** Give details here of the substance(s), the dose or amount to be given, likely effects (including duration) and any potential hazards to participant(s) and/or researcher(s).

**(b)** Indicate here what precautions will be taken to avoid or minimise any adverse

effects.

**QA3.** Are there any potential hazards to participant(s) and/or researcher(s) arising from the use of the proposed invasive procedures? (See GN 2.2.10)

YES

NO

If YES,

**(a)** Indicate their nature here.

**(b)** Indicate here what precautions will be taken to avoid or minimise any adverse effects.

**QA4.** Will or could the study cause discomfort or distress of a mental or emotional character to participants and/or researcher(s)? (See GN 2.2.11)

YES

NO

If YES,

**(a)** Indicate its nature here

**(b)** Indicate here what precautions will be taken to avoid or minimise such adverse effects.

**QA5.** Medical or other aftercare and/or support must be made available for participants and/or researcher(s) who require it where invasive procedures have been used in the study. Please detail what aftercare and/or support will be available and in what circumstances it is intended to be used. (See UPR RE01, S 2.3 (ii) and GN 2.2.12)

**QA6. (a)** Please state here previous experience (and/or any relevant training) of the supervisor (or academic member of staff applying for a standard protocol) of investigations involving hazards, risks, discomfort or distress as specified. (See GN 2.2.13)

**(b)** Please describe in appropriate detail what you would do should the adverse effects or events which you believe could arise from your study, and which you have mentioned in your replies to the previous questions, occur.

**QA7.** In the event that the study reveals that a participant has a pre-existing medical condition (of which they may or may not be aware), and which could affect their present or future health or that of others, they should be informed of this in an appropriate manner and advised of follow-up action that they should take. (See GN 2.2.18) Advice should be sought as to whether information should be passed to their GP and a decision taken whether they should be allowed to continue to take part in the study. If a potential participant is not willing to agree to such action being taken in these circumstances, they should not be allowed to take part in the study. Please indicate here what arrangements have been made for complying with these requirements.

**Please revert to Q15.**

## **APPENDIX 2 – DOCUMENTS TO BE ATTACHED**

Please attach the following documents if you have affirmed possession of them in the relevant questions:

- |   |  |
|---|--|
| (a) Permission from the location to be used to carry out this study (Q7)<br>(This includes permission to use a location on UH Campus and any location off of UH Campus which requires permission to use.)   | <input type="text" value="N/A"/>           |
| (b) Risk assessment for off campus location (See Form EC5) (Q8)   | <input checked="checked" type="checkbox"/> |
| (c) Copy of Consent Form (See Form EC3 & Form EC4) (Q18)  | <input checked="checked" type="checkbox"/> |
| (d) Copy of Form EC6 (Participant Information Sheet) (Q5)   | <input checked="checked" type="checkbox"/> |
| (e) CRB Disclosure (Q22)  | <input type="checkbox"/>                   |
| (f) A copy of the proposed questionnaire and/or interview schedule (if appropriate for this study). For unstructured methods, please provide details of the subject areas that will be covered and any boundaries that have been agreed with your Supervisor. | <input checked="checked" type="checkbox"/> |

**University of Hertfordshire  
CONSENT FORM**

**Parental adjustment after the birth of their child with a facial  
birthmark**

I, the undersigned [*please give your name here, in BLOCK CAPITALS*]

.....  
.....  
of [*please give contact details here, sufficient to enable the investigator to get in touch with you, such as a postal or email address*]

.....  
.....  
hereby freely agree to take part in the study entitled [*insert name of study here*]

.....  
.....

**1** I confirm that I have been given a Participant Information Sheet (a copy of which is attached to this form) giving particulars of the study, including its aim(s), methods and design, the names and contact details of key people and, as appropriate, the risks and potential benefits, and any plans for follow-up studies that might involve further approaches to participants. I have been given details of my involvement in the study. I have been told that in the event of any significant change to the aim(s) or design of the study I will be informed, and asked to renew my consent to participate in it.

**2** I have been assured that I may withdraw from the study at any time without disadvantage or having to give a reason.

**3** I have been given information about the advantages and possible disadvantages of taking part in the study.

**4** I have been told how information relating to me (data obtained in the course of the study, and data provided by me about myself) will be handled: how it will be kept secure, who will have access to it, and how it will or may be used.

Signature of participant.....

Date.....

Signature of (principal) investigator.....

Date.....

Name of (principal) investigator [*in BLOCK CAPITALS please*]

**FORM EC5 – STANDARD RISK ASSESSMENT FORM**

**NAME: Lucy Gait**

**DATE: 6/10/ 2015**

Activity Description	Interviews with participants for MRP research				
1. IDENTIFY HAZARDS	2. WHO COULD BE HARMED & HOW?		3. EVALUATE THE RISKS	4. ACTION NEEDED	
<u>Activities/tasks and associated hazards</u> Describe the activities involved in the study and any associated hazards. Check sector specific/UH specific Guidance. (NB. Describe any equipment to be used and read manufacturer's instructions and note any hazards that arise, particularly from incorrect use.)	<u>Who is at risk?</u> e.g., participants, researchers, other people at the location, the owner / manager / workers at the location etc.	<u>How could they be harmed?</u> What sort of accident could occur, e.g., trips, slips, falls, lifting equipment etc., handling chemical substances, use of invasive procedures and correct disposal of equipment etc. What type of injury is likely?	<u>Are there any precautions currently in place to prevent the hazard?</u> Are there standard operating procedures or rules for the premises? Have there been agreed levels of supervision of the study? Will trained medical staff be present? Etc.	<u>Are there any risks that are not controlled or not adequately controlled?</u>	<u>List the action that needs to be taken to reduce/manage the risks arising from your study?</u>
Interviews with participants	Participant & clinician,  It is possible that slips, trips or falls could occur at the locations of the interviews.		Care will be taken by the researcher to be alert to potential hazards and avoid them and to alert participants to them to ensure their safety.	No additional actions required.	
Lone working	Researcher		The researcher is aware of lone working policies and would abide by these. The researcher would not enter somewhere that obviously appears unsafe or dangerous. The researcher would	No additional action required.	

Parental ad. to birthmark

		<p>phone a contact such as their supervisor and inform them of where they are, when they are entering a premises and what time they expect to leave. The researcher would then call to confirm they have left. The researcher is Breakaway trained and although highly unlikely to be required, she could use this if necessary.</p>	

<p>Signed by applicant:</p> <p>L. Gait</p>	<p>Signed by Off Campus Location Representative:</p> <p>(NB. This needs to be the person who has given permission to use the location)</p>
--	--

## **PARTICIPANT INFORMATION SHEET**

### **Parental adjustment after the birth of their child with a facial birthmark**

#### **Introduction**

You are being invited to take part in a research study. Before you decide whether to do so, it is important that you understand the research that is being done and what your involvement will include. Please take the time to read the following information carefully and discuss it with others if you wish. Do not hesitate to ask us anything that is not clear or for any further information you would like to help you make your decision. Please do take your time to decide whether or not you wish to take part. Thank you for reading this.

#### **What is the purpose of this study?**

My name is Lucy Gait, I am a third year Trainee Clinical Psychologist from the University of Hertfordshire and I am conducting this research for my 3rd year Doctoral research project.

The purpose of this study is to explore the experiences of parents who have a child who has a congenital birthmark. The hope is that this research will be able to inform professionals who work in this area and improve services and support.

#### **Do I have to take part?**

It is completely up to you whether or not you decide to take part in this study. If you do decide to take part you will be given this information sheet to keep and be asked to sign a consent form. Agreeing to join the study does not mean that you have to complete it. You are free to withdraw at any stage without giving a reason. A decision to withdraw at any time, or a decision not to take part at all, will not affect any part of your care or the support you receive.

#### **What will happen to me if I take part?**

If you decide to take part in this study, you will be asked to take part in a semi-structured interview with myself that will last for approximately one hour. The interview can take place at a location of your choice. The questions that you will be asked will relate to your experiences since the birth of your child and your thoughts about the future.

If there were any significant changes to the method you would be informed and asked to re-consent.

#### **What are the possible advantages and disadvantages of taking part?**

By choosing whether or not to take part will not affect your opportunities or future support. Agreeing to take part will provide you with an opportunity to share your experiences and for psychologists, clinicians, researchers and organisations to strengthen and deepen their understanding of the impact facial birthmarks has on parents and families.

### **How will my taking part in this study be kept confidential?**

The interviews will take place at a time and place of your choosing. Your interview will be recorded and kept in encrypted and password protected files to ensure confidentiality. Once the data is transcribed, it will be kept in locked files and your transcript will be assigned a unique code. In the research write up, you will be referred to by another name and any information contained in quotes will be anonymized to ensure your confidentiality is maintained.

### **What will happen to the results of the research study?**

The results from the study will be written up in a doctoral thesis for the purposes of gaining the Doctorate in Clinical Psychology qualification. It is hoped that the findings will be shared in academic publications and/or presentations however, any information that is made public following this research will be fully anonymised.

The data collected in the study will be kept securely for 5 years at the University of Hertfordshire years in accordance with the policy for awarding the Doctorate of Clinical Psychology to the researcher and for the purposes of publication. If the researcher wishes to use the data for any purpose, other than the study outlined in this information sheet, you will be informed and additional consent will be sought from you.

### **Who has reviewed this study?**

The study has been reviewed by The University of Hertfordshire Health and Human Sciences Ethics Committee with Delegated Authority [Protocol Number LMS/PGR/UH/02105] to protect your safety, rights and well-being. This study has been reviewed and can be conducted as it has been granted favourable opinion with no raised objections on ethical grounds.

As this research project is also part of the academic commitment, it has been formally and peer reviewed by the University of Hertfordshire's Doctoral Programme in Clinical Psychology.

### **Who is supervising the project?**

The project is being supervised by Dr Clare Norris (Clinical Psychologist and Lecturer at University of Hertfordshire) and Dr Helen Ellis- Caird (Clinical Psychologist and Research Lead at University of Hertfordshire).

### **Who can I contact if I have any questions?**

If you have any additional questions or any concerns or complaints during the study, please contact the principal investigator (details below) or research supervisor (details below).

**Lucy Gait** – Tel: 07980 019785  
c/o Clinical Psychology Doctorate Admin Office  
University of Hertfordshire  
Health Research Building  
College Lane, Hatfield  
Herts  
AL10 9AB

Email: [l.gait@netmail.herts.ac.uk](mailto:l.gait@netmail.herts.ac.uk)

Parental ad. to birthmark

**Dr Clare Norris** – Tel:01707 286322 (office)

University of Hertfordshire  
Health Research Building  
College Lane, Hatfield  
Herts  
AL10 9AB

Email: [c.norris@herts.ac.uk](mailto:c.norris@herts.ac.uk)

**Dr Helen Ellis- Caird** - Tel:01707 286322 (office)

University of Hertfordshire  
Health Research Building  
College Lane, Hatfield  
Herts  
AL10 9AB

Email: [h.ellis-caird@herts.ac.uk](mailto:h.ellis-caird@herts.ac.uk)

**Thank you very much for reading this information and giving consideration to taking part in this study.**

### **Proposed semi-structured interview schedule**

1. Tell me a bit about yourself/your child/your family (any other children etc.?)
2. How did you feel when you first found out about the birthmark? (Do you have a clear diagnosis, do you understand what the long term prognosis is?)
3. What if anything did you know about birthmarks?
4. How have you learned to handle/cope with it?
5. How has this affected your relationship with your child?
6. Has it affected your relationship with your partner?
7. What were the reactions of the staff at the hospital?
8. How did your family/friends react?
9. Tell me about how you go about handling reactions of other people e.g. members of the public?
10. What challenges have you faced since X was born? (personally, socially, medically)
11. Could you tell me about the lessons you have learned through these experiences?
12. What are your concerns/worries for the future?
13. What are your hopes for the future?
14. What would you say are the most important things to consider when your child has a birthmark?.....Have these views changed at all since X was born?
15. After having these experiences, what advice would you give to other parents in the same situation/ professionals?

Parental ad. to birthmark

Eii – 1<sup>st</sup> EC2 form

UNIVERSITY OF HERTFORDSHIRE

ETHICS COMMITTEE FOR STUDIES INVOLVING THE USE OF HUMAN PARTICIPANTS  
(‘ETHICS COMMITTEE’)

**FORM EC2: APPLICATION FOR MODIFICATION AND/OR EXTENSION TO  
AN EXISTING PROTOCOL APPROVAL**

- 1 **Protocol Number and Title of original application:**  
(Please provide the original application and attachments. This application will not be accepted without them)

How do parents adjust after the birth of their child with a facial birthmark?

Protocol number: Protocol Number LMS/PGR/UH/02105

Applicant name: Lucy Gait

Student registration number: 12240945

Applicant e-mail address: lucy.gait@herts.ac.uk

Work address (if appropriate)

Supervisor’s name: Dr Clare Norris

Supervisor’s School & Department: Doctorate of Psychology (Health & Human Sciences)

Supervisor’s e-mail address: c.norris@herts.ac.uk

- 2 **Specify the nature of the modification/extension**

i Revised title of study (if applicable)

ii Time (specify months and year): From: To:

iii Additional worker(s). (Name workers or supervisor to be added to the protocol approval)

Name:

Work address:

E-mail address

iv Change of supervisor from Dr Clare Norris[name] to Dr Helen Davies [name]

Please complete declaration and give reason in Section 3

Declaration by new supervisor:

Parental ad. to birthmark

I have reviewed the ethics protocol paperwork for this study and am aware of any conditions which must be adhered to.

Signed .....

v Location of study:

vi Details of modification: (Please list all changes being made from the original procedure on a separate sheet)

**Requested Modifications:**

**Increasing the age requirement of the children from 3-6 months to 0-2 years to increase opportunity for more parents to participate**

**3 Reason for extension/modification request**

(please explain)

1. The change of supervisor has been discussed within the team and it is felt the Helen, as a paediatric psychologist has greater expertise in the field.
2. Increasing the age range of the children with birthmarks increases the opportunity for more parents to participate in the study which will allow for a greater number of interviews to be completed which will aid the quality of the research and aid the recruitment process.

**4 Hazards**

Does the modification or extension present additional hazards to the participant/investigator?

YES

NO

If YES, a new risk assessment may be necessary. Subject specific forms may also be necessary; you should therefore contact your Supervisor or School to see whether this is the case.

Signature of Applicant .....L.Gait.....Date 22/11/15

Support by Supervisor.....Date.....

UNIVERSITY OF HERTFORDSHIRE

ETHICS COMMITTEE FOR STUDIES INVOLVING THE USE OF HUMAN PARTICIPANTS  
(‘ETHICS COMMITTEE’)

Eiii: Second EC2 form

## FORM EC2: APPLICATION FOR MODIFICATION AND/OR EXTENSION TO AN EXISTING PROTOCOL APPROVAL

**1 Protocol Number and Title of original application:**

(Please provide the original application and attachments. This application will not be accepted without them)

How do parents adjust after the birth of their child with a facial birthmark?

Protocol number: Protocol Number LMS/PGR/UH/02105 (1)

Applicant name: Lucy Gait

Student registration number: 12240945

Applicant e-mail address: lucy.gait@herts.ac.uk

Work address (if appropriate)

Supervisor’s name: Dr Helen Davies

Supervisor’s School & Department: Doctorate of Psychology (Health & Human Sciences)

Supervisor’s e-mail address: h.ellis-caird@herts.ac.uk

**2 Specify the nature of the modification/extension**

i Revised title of study (if applicable)

ii Time (specify months and year): From: To:

iii Additional worker(s). (Name workers or supervisor to be added to the protocol approval)

Name:

Work address:

E-mail address

iv Please complete declaration and give reason in Section 3

Declaration by new supervisor:

I have reviewed the ethics protocol paperwork for this study and am aware of any conditions which must be adhered to.

Parental ad. to birthmark

Signed .....

v Location of study:

vi Details of modification: (Please list all changes being made from the original procedure on a separate sheet)

**Requested Modifications:**

To interview professionals (e.g. clinical nurse specialists) who work with parents of children with a birthmark. This is an extension to the current ethics which was to interview parents only.

**3 Reason for extension/modification request**  
(please explain)

In order to deepen the understanding about the research subject.

**4 Hazards**

Does the modification or extension present additional hazards to the participant/investigator?

YES

NO

If YES, a new risk assessment may be necessary. Subject specific forms may also be necessary; you should therefore contact your Supervisor or School to see whether this is the case.

Signature of Applicant .....L.Gait.....Date 13/02/15

Support by Supervisor.....Date.....

Eiv: Evidence of ethical approval granted for this study

**UNIVERSITY OF HERTFORDSHIRE  
HEALTH AND HUMAN SCIENCES  
ETHICS APPROVAL NOTIFICATION**

**TO** Lucy Gait

**CC** Dr Helen Davies & Dr Helen Ellis-Caird

**FROM** Dr Richard Southern, Health and Human Sciences ECDA Chairman

**DATE** 23/02/2016

Protocol number: aLMS/PGR/UH/02105(2)

Title of study: How do parents adjust after the birth of their child with a facial birthmark?

Your application to modify the existing protocol as detailed below has been accepted and approved by the ECDA for your School.

Modification: To include interviews with professionals who work with parents of children with a birthmark.

This approval is valid:

From: 23/02/2016

To: 01/06/2016

**Please note:**

**Any conditions relating to the original protocol approval remain and must be complied with.**

**Approval applies specifically to the research study/methodology and timings as detailed in your Form EC1 or as detailed in the EC2 request. Should you amend any further aspect of your research, or wish to apply for an extension to your study, you will need your supervisor's approval and must complete and submit a further EC2 request. In cases where the amendments to the original study are deemed to be substantial, a new Form EC1 may need to be completed prior to the study being undertaken.**

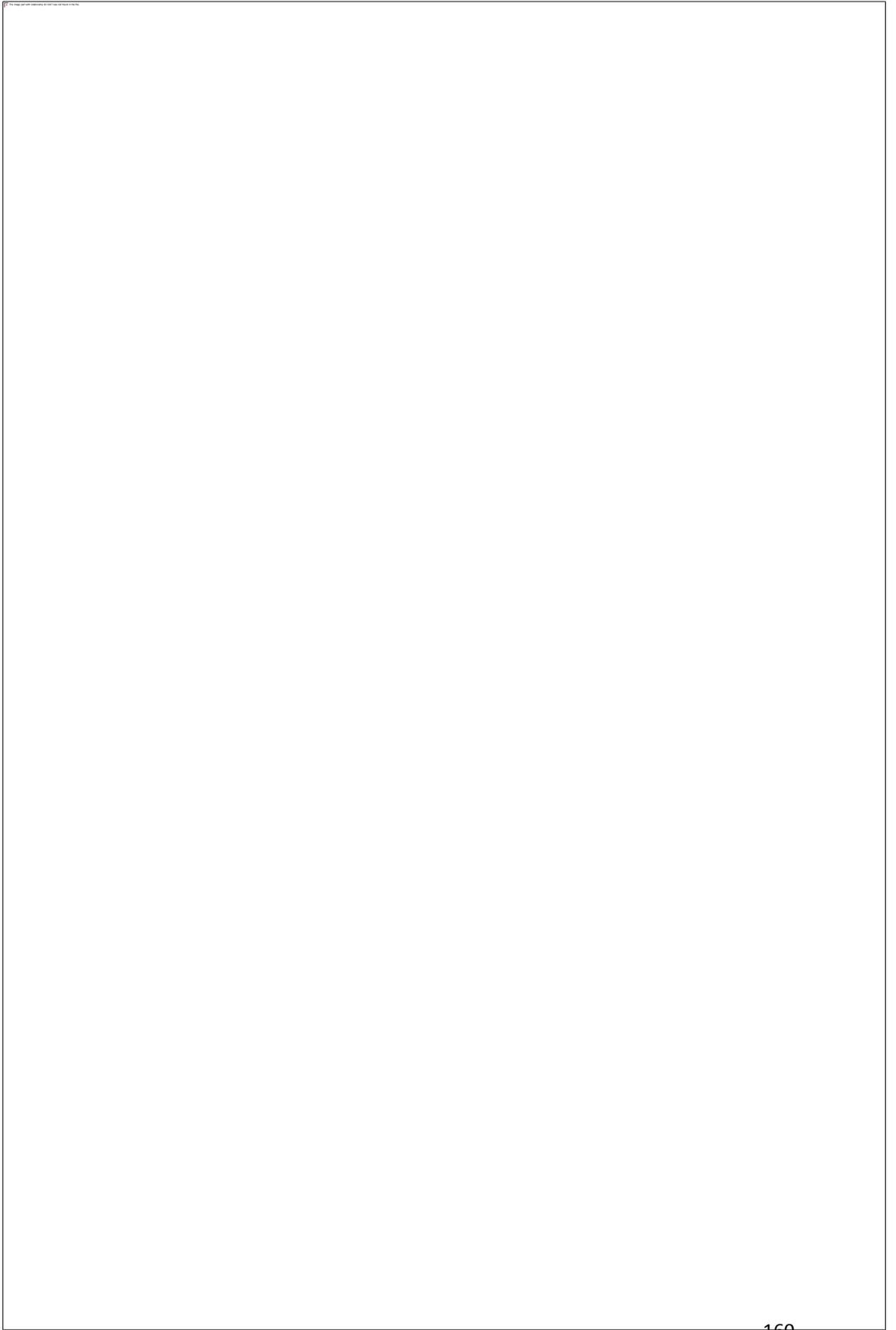
**Should adverse circumstances arise during this study such as physical reaction/harm, mental/emotional harm, intrusion of privacy or breach of confidentiality this must be reported to the approving Committee immediately. Failure to report adverse circumstance/s would be considered misconduct.**

**Ensure you quote the UH protocol number and the name of the approving Committee on all paperwork, including recruitment advertisements/online requests, for this study. Students must include this Approval Notification with their submission.**

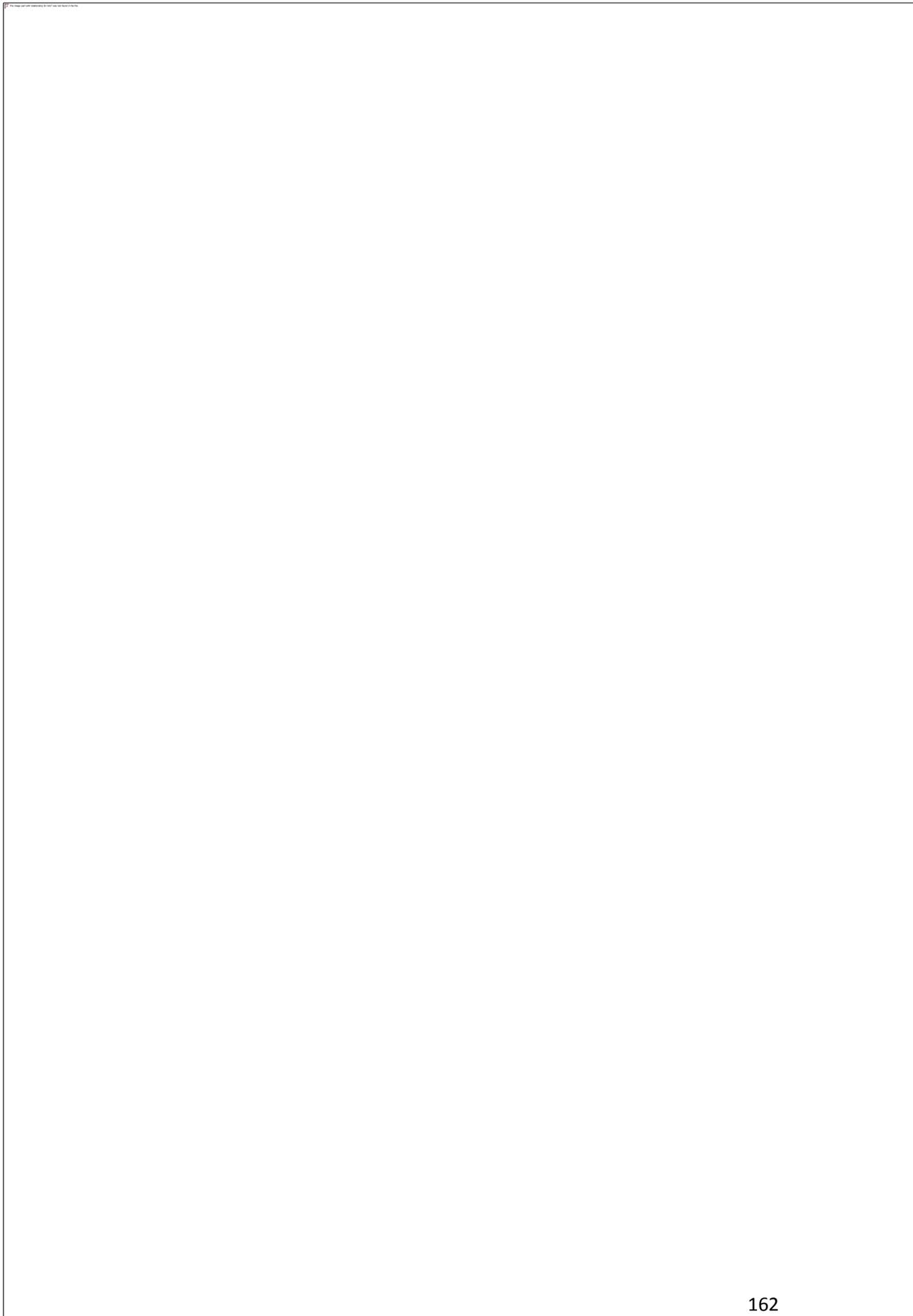
Parental ad. to birthmark

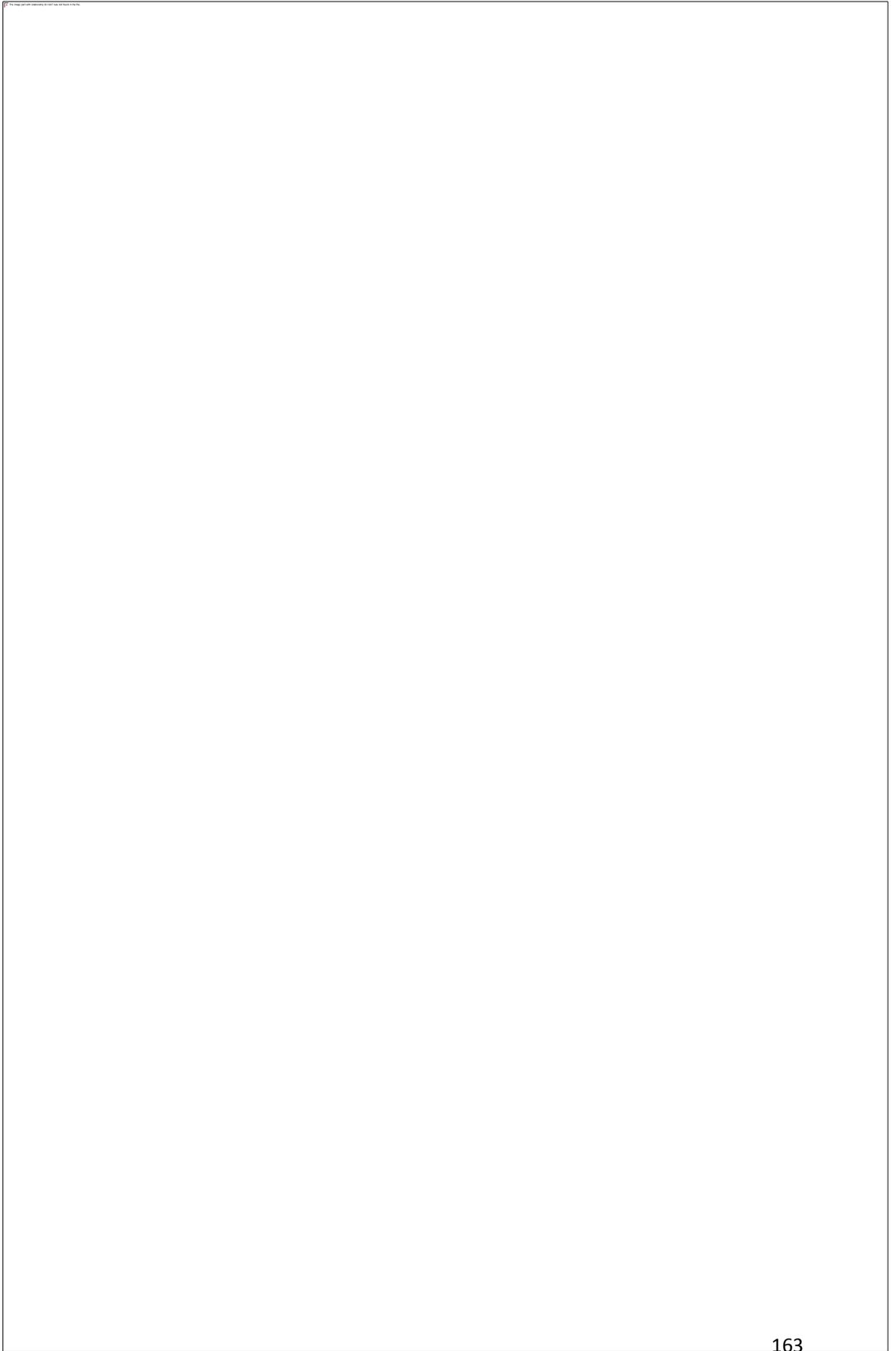
Appendix F i: Transcript exert

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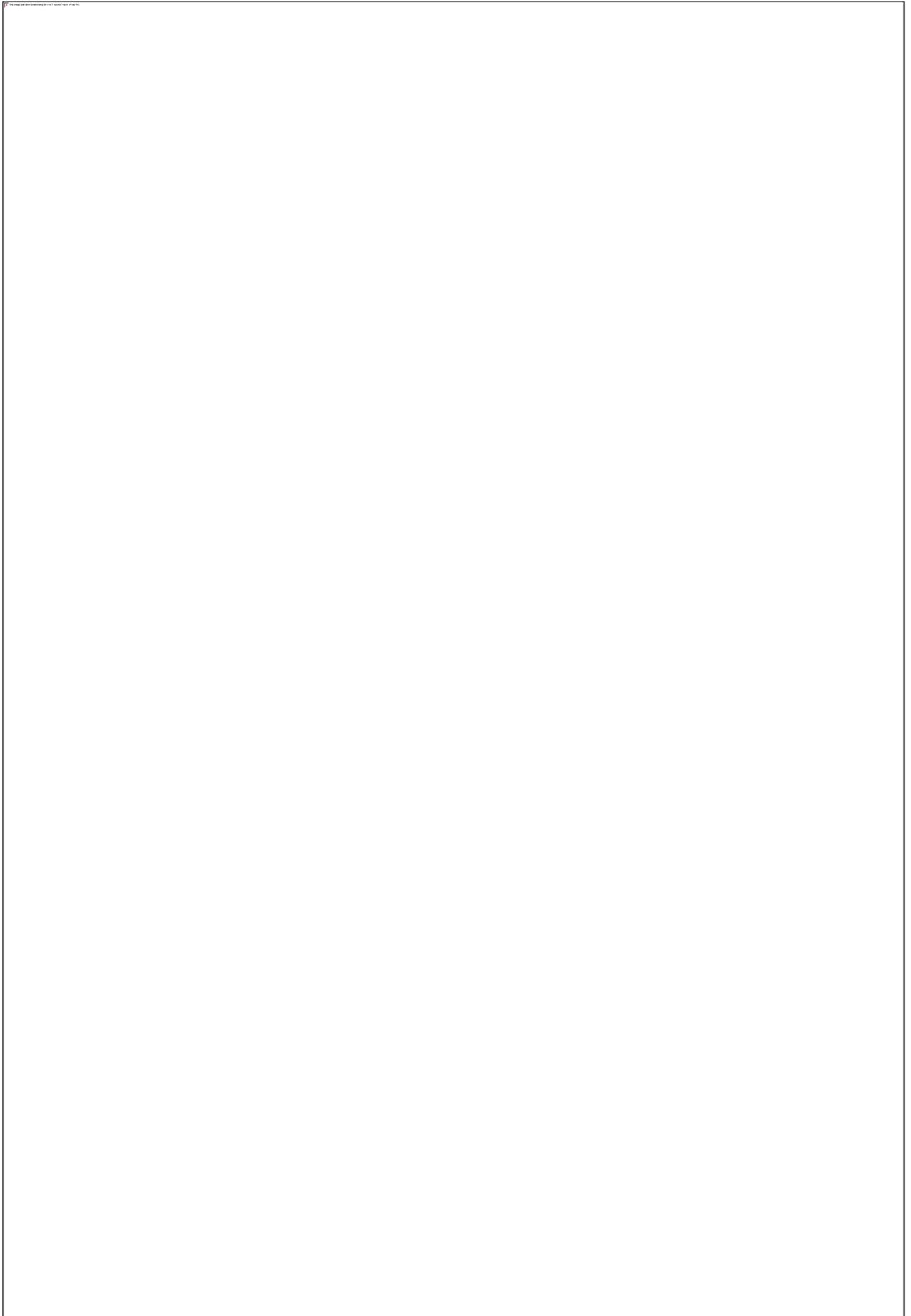


Parental ad. to birthmark





Parental ad. to birthmark





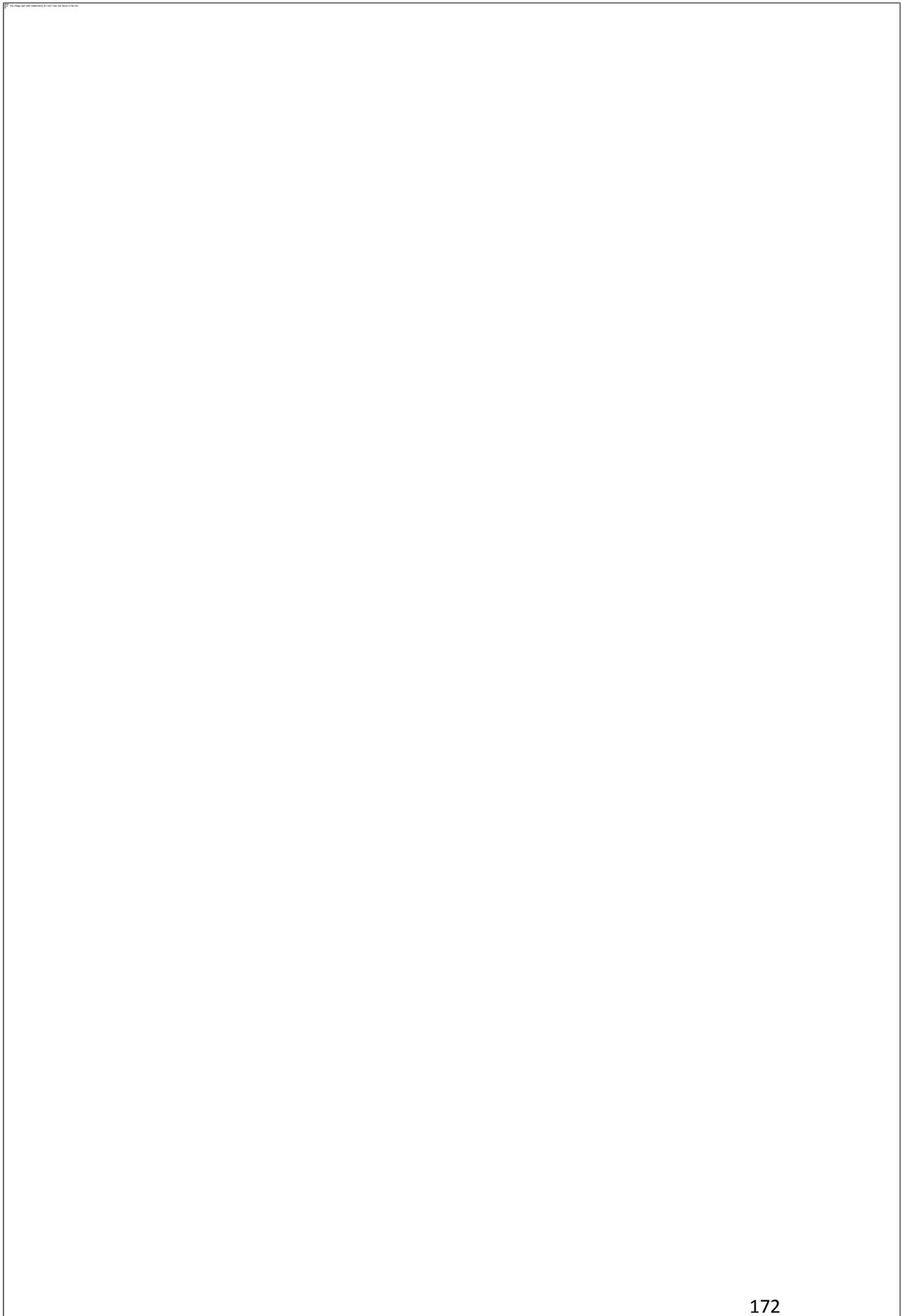
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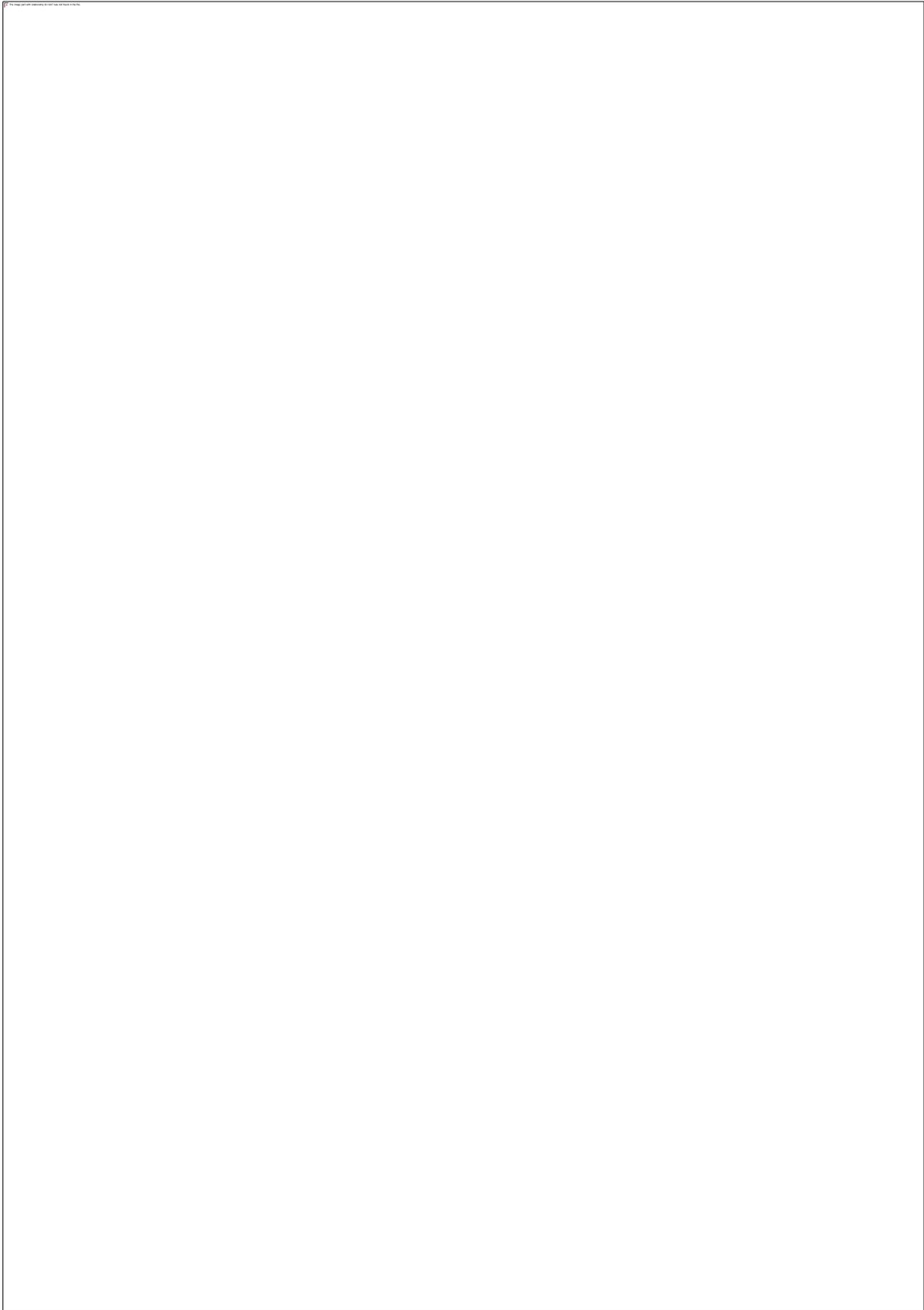


Parental ad. to birthmark





Parental ad. to birthmark



Parental ad. to birthmark



Parental ad. to birthmark

Appendix F ii: Exerts from memos

<p>18.12.15</p>	<p>Medical staff, role of support and coping</p>	<p>All found that medical professional knowledge was unsatisfactory but had a way out, through money or family but acknowledged that's not the same for everyone else.....leading to a story of being 'lucky' seeing others on the group....it could be worse idea.</p> <p>All found the group crucial to coping and managing both for practical advice and support and comfort</p> <p>All found family important for support, emotionally and practically although some variation on amount of discussion with partners about emotional impact of the experience</p> <p>All commented on negative of group is fear mongering, worries about others experiences happening to them – However all expressed the support the group has been. All commented on remarks and stares but felt they had not had to deal with too much of this, but this worry led them to 'be on edge' and feel anxious in public and for Hannah this led her to be socially avoidant.</p> <p>All promoting better medical education for staff on the front line and greater support in making treatment choices</p> <p>Avoidance came up – mentally not thinking about it and practically not going out and for Tom positive thinking was important 'cross that bridge when we get to it' type thinking</p> <p>All spoke of having a good cry early on and were unsure about whether this was normal for post pregnancy or if this was caused or exacerbated by the birthmark. Hannah and Sally had older children and were able to say they thought they were more emotional than with their firsts.</p> <p>Seems to be a process, moving from being panicked, very emotional and needing support early on to moving towards a point of maybe acceptance or balance and at those times the importance of sharing and supporting others was noted.....spreading the word</p> <p>Both talked about how it had made they feel very protective of their children and felt like they want to promote knowledge and education of birthmarks and expressed this was related to wanting to take part in the project.</p> <p>12.1.16 – mixed reactions to medical staff, appears that speed in which the initial stage, answers and certainty can be given the sooner the parents can accept the birthmark or treatment route allowing them to cope better.....the longer they are in a not knowing place, the harder it appears to be for them.</p>
<p>After the interviews</p>	<p>Initial thoughts</p>	<p>Dad main carer and mum worked, which was a new dynamic – dad already felt judged for this and found comments about</p>

<p>with Anna &amp; Jack</p>		<p>the birthmark particularly hard to manage if he felt implicated in some way.</p> <p>Difficulties conceiving so strong family sense of not caring about the baby's problems, may have helped move forwards more quickly. However, I noted strong emotion in mum's voice when talking about her daughter/family. She was heavily pregnant and I wonder if this may have been an influence or perhaps she is finding it harder to manage than she openly said.</p> <p>However, still shock and upset especially regarding the physical problems they can cause and therefore the amount of medical input and appointments to adjust to.</p> <p>Strongest sense of it being a 'special mark' and wanting to strongly instil daughter with confidence to tell people</p> <p>Found the group very helpful, especially showing pictures of others to their daughter to normalise hers.</p> <p>Have experienced some of the most difficult responses from the public of anyone I have spoken to, this has been difficult but they expressed feeling in it together as a team and always share with each other difficult experiences. Dad found it hard to stay with negative emotions and moved quickly back to a place of pride in his daughter for coping.</p> <p>Daughter is 2 and furthest along with treatment – greatest discussions about dilemmas around treatment – who is the treatment for, wanting to know the future, will it be worth it? Will she be grateful for us doing this? Will she be angry with us for doing this? How will the treatment/hospital experiences affect her? Etc.</p> <p>Spent the longest time yet thinking about social reactions and managing social reactions – I wonder whether this is related to having the most difficult social experience of people I've spoken to or whether this is more relevant to her age as she is 2 she is going out more and is able to talk and so there is greater onus on her beginning to understand and explain it herself than with the younger children who have not yet realised, or learned to talk.....may need to explore further.</p>
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<p>Trends so far</p> <p>21.12.15</p>	<p>Shock &amp; Uncertainty</p>	<p>There appears to be an initial shock for parents about the appearance, but at that early stage the first adjustment is to the appearance, but as medical professionals become involved and they learn about the potential physical implications of the birthmarks they are overwhelmed by the shock, the grief for not having the perfect baby and a great deal of uncertainty for the future.</p> <p>Time and good social and emotional support appears to aid the process of adjustment, but the medical input and on-going concerns about potential complications remains, leaving this constant sense of anxiety or uncertainty. This uncertainty appears to be exacerbated by slower medical support and mirrored in social interactions; while most reported thinking/worrying about possible reactions generally had only innocent or curious reactions, but Anna had a horrible experience and all had heard of others having difficult</p>
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		<p>interactions with the public from the group which appeared to leave them with a certain hypervigilance when in public places.</p> <p>There appears to be a split in how this uncertainty is managed, some share with partners and are open about their experiences while others hold their concerns either it would appear use intellectualisation and rationalisation to manage their distress or hold it themselves, feeling quite alone in it so not to worry their partners or because they worry about what that means about them. Eg Penny – judgement of self as a bad mum for having those thoughts.</p>
<p>Trends so far</p> <p>21.12.15</p> <p>12.1.16</p>	<p>Protect, Explain &amp; Promote</p>	<p>It appears to be a dilemma between protecting their children but having to put them forward for treatment which can be an ordeal in its self. Publically, again the need to protect is evident however it appears that remaining calm and explaining and promoting knowledge and good will is deemed important in trying to educate people in the hope that this will lead to greater acceptance of their child and ease their child's path in life.</p> <p>This is tackled in different ways so far, role play, stories, word patterns, informing people before they meet the child etc.</p> <p>These appear to be more distinct categories. Protect – a lot of concern in the present and for the future, concerns about adjustment, socially, treatment and psychological effect on child.</p> <p>Explain – using words, books, resources, roll play, preparing both the child and also managing to explain socially to others, how to do it, preparing phrases, allowing/inviting people to ask, being polite about it when people do comment.</p> <p>Promoting – actively when people ask, trying not to react defensively, working to normalise it and then promoting in terms of giving back to the group and supporting other people in the same situation.</p>
<p>Trends so far</p> <p>21.12.15</p>	<p>The treatment dilemma</p>	<p>There appears to be mixed emotions about treatment, the unknown outcome and whether it's right to put their child through treatment, would the child want it, what would the child say in years to come if they don't, how will they explain it to them? Is it right to put a child through elective surgery for cosmetic reasons? And the outcome, they have now adjusted to the birthmark, what would they look like without it, how would they feel about that difference?</p>
<p>Trends so far</p> <p>21.12.15</p> <p>13.1.16</p>	<p>Forward thinking: Attempts to pre-empting everything</p>	<p>Need for planning, strategies for themselves about what to say to people about it, prepared word patterns and responses.</p> <p>Also planning for the child, how to tell them, resources to use, preparing them to deal with the comments</p> <p>Thinking about siblings and other family and preparing them before they see it and preparing siblings to know what to say to other children as well</p>
<p>Coding Penny</p>	<p>Not knowing – the process of</p>	<p>Initially isolated, not knowing, not great medical support results in questions about self, possible explanation of the</p>

27.12.15	sense making	birthmark, blaming self, feeling guilty/low/lonely/not understood by others. Not sharing true inner struggles and feelings in case people will judge her. Finding support group is a relief, gain knowledge, share experiences and feel understood by others in the same situation. However, seeking information uncovers further worries and risks of PWS, leading to further worry to hold on to. However, once information and answers (as much as possible) have been given, this allowed transition from shock/worry/uncertainty in to a state in which the process of acceptance was possible.
Coding Penny 27.12.15	Protecting partner	Despite isolation, not sharing, noticing apparent coping by partner leading to further self-blame and shame as a mother. Also not wanting to share her feelings and findings from researching on the internet for fear of upsetting and worrying her partner.
Coding Penny 27.12.15	Feeling in control	Early stages when there was the shock, poor and unorganised medical support and various referrals, combined with grief of not having her perfect baby and not being able to share = feeling out of control. Slowly gaining information and advice, taking control of appointments and choice of hospital, treatment options, pre-empting reactions and having planned word patterns = feeling in greater control and feeling knowledgeable
Coding Penny 27.12.15	Comparison with others	Comparison with other PWS's on the internet = sense of relief, feeling that O's is not so bad. Comparison with other children more widely, thinking about LD and other health difficulties and miscarriage and still born babies resulted in feeling grateful for their son and put the birthmark in perspective.
Coding Penny 27.12.15	"I breast fed him which feels ridiculous"	Given her challenges to come to terms with the birthmark she felt that the closeness of breast feeding was ridiculous. She hinted at feelings of wishing to reject the baby because of its appearance and finding it difficult to voice such feelings. Perhaps there was a wish to create distance with him, however perhaps the intimacy of breast feeding supported the development of a bond that was strained from the beginning
Coding Penny 27.12.15	Reaction of others	Generally positive, family and friends positive and social reactions generally positive, one instance in a coffee shop of a child shouting, but well managed, disliking mothers shielding their children. Wanting to share and show and familiarise other children in the hope to normalize difference. Children's worries that it hurts him, but no unpleasant reactions have reduced parent's worries and anxieties, although still some anxiety for his future and his school years. Strongly dislike a sense of pity from others "that's a shame" reactions. Noticing stronger reactions from older generations than younger people/children.
Sarah 28.12.15	Initial thoughts	Mother has autism which made it difficult for her to connect with her feelings. She had a similar experience to others in terms of little medical knowledge or support with diagnosing the birthmark. Researched greatly herself....her coping mechanism for managing her anxiety/autism was to google

		everything. Difficult home life as other children had a variety of ASD and genetic conditions. Concerns that the baby may also have similar difficulties and unsure if these could be birthmark related for more similar to siblings problems. Little social experience/staring/comments so was not able to elaborate on that. Interview was ended prematurely due to her having to take a phone call. Did not have chance to explore her support systems in depth although she referred briefly to her partner, mother and sister in law suggesting some support was available. Good relationship with a paediatrician appeared to also be a good source of support for her. Resilience having faced difficulties with other children may also have helped prepare her
7.1.16	Preparation preparation preparation	(Sally) Being prepared and educating and providing tools and word patterns for her children to answer comments on the birthmark. Also preparing herself in how to respond to the public. (similar to Penny). Having resources, books, teddy bear and role play were important.
29.2.16		Being prepared appears to be important in terms of managing social reactions and preparing the family and the child for coping in public and in terms of managing treatment routines.
8.1.16	BSG	The support group appears to be an important resource for parents to share advice, get support and give back to others. It has been described as a safe place for parents to share difficult experiences and get support, information and advice.
8.1.16	Promoting difference & sharing	Wanting to promote difference and educate others appear to be important for mothers. Additionally supporting others on the group and sharing advice is important.
8.1.16	Feeling Lucky	Everyone has spoken about feeling lucky about their situation. It appears that later, after the initial shock, once any syndromes have been ruled out, parents consider themselves lucky that their child's is not worse. Additionally seeing photos of other children and hearing other stories (comparing their situation to others) on the group appear to lead to parents feeling lucky their child's is not worse. This represents a change in the group they choose to compare themselves to.
10.2.16		Greater nuance in this category – lucky for not being worse, for not having syndrome, lucky for having family support, lucky for support group help, lucky in instances when they have had good treatment opportunity/medical staff
8.1.16	Public reactions	Appear to be age/generational differences in expectations and acceptance of response. E.g.. Older people appear most likely to make what is felt to be inappropriate responses, while child curiosity is accepted. Responses from the mothers of curious children appear to differ. Open responses appear to be preferred to reactions such as shushing the child or ushering them away. Shying away from public and avoiding going out was reported by Hannah, Sally and Penny reported anxiety about public reactions but mentally prepared themselves. Perhaps personality type or preparation plays a role here.
10.2.16		

		Definite cultural impact on reactions, Anna, Jack and Kate have had significant differences in reactions from western vs Asian people who have responded.
8.1.16  10.2.16	Medical professionals	<p>Mixed experiences of medical professionals, often poor knowledge, and disorganised and slow referrals process. This seems to impact on their level of early distress, speed of adjustment and trust in what the doctors tell them. Medical professionals were not even able to give enough advice on wound dressings for Hannah, only print outs from the internet as advice.</p> <p>This continues throughout all of the transcripts and appears to not be related to geographical locations. Personal links to medical professionals (Sally) and (Sarah) appear to help. As does financial wealth allowing for private consultations (Hannah)</p>
8.1.16 16.2.16	Missing out on parts of babyhood	Early adjustment and in the case of Hannah & Amy intensive treatment experience led to feelings of missing out on parts of the child's first few months, especially with HMP being prem. Focus being on birthmark for some lead to less attention on other aspects of the child's personality and development in these cases and with PWSO where anxiety about birthmark was very high. Perhaps moving forwards parents need to manage their feelings about this as a loss as well as adjustment to the birthmark and treatment processes learning to manage social encounters.

Appendix F iii: Table of codes and categories

Category	Sub-category	Focused code	No. transcripts code occurs in	Examples of quotes making up the code
Accepting the birthmark	Accepting birthmark	Learning to love the birthmark	8	<p>Penny " I love his birthmark and I almost want people to see it and like, I want to show it off and stuff.</p> <p>Anna "but all the kind of family friend reactions, they've just accepted it as part of her face" .... " It's never going to disappear and I think it you've got to like it or lump it, you know what are you going to do, you've got this gorgeous little baby that you've just made, that you've waited a long, long time for and is wonderful, you're going to like it, aren't you, eventually."</p> <p>Charlie "I think we got used to it because we saw it every day and it just didn't bother us at all. It was part of her you know."</p>
Accepting the birthmark	Noticing the birthmark less	Noticing the birthmark less	11	<p>Penny "I looked at it the other day in M&amp;S with my mum and I was like oh my god its disappeared, like it isn't there.".... "most of our family now look at him and can't see it and I genuinely believe that they do look at him and can't see it as we can't."</p> <p>Sally "I do still see it but not all of the time, you don't notice it."</p> <p>Kate "we got the picture on the wall with her birthmark clearly visible before we actually started with treatment, ummm but as I said, it's all part of her and you don't really notice it."</p>
Accepting the birthmark	Permanency of birthmark	It's only temporary	6	<p>Hannah "I mean we're lucky, the propranolol worked and it's going down."</p> <p>Amy "the Propranolol medication which has worked wonders now, I mean you can still see it but it's nowhere near as blue, as scary and it doesn't block her ear canal anymore."</p>
Accepting the birthmark	Permanency of birthmark	Marked for life	8	<p>Anna "we had no idea how it's gone from being a bruise which obviously is something very temporary to being something like long that could affect not just her appearance but everything about how she grew up."</p> <p>Jack "We know it's going to be there forever on her face in one way or another, she'll have a birthmark that is visible."</p> <p>Kate " I suppose the key message is that they are your child and it's just a mark, in a sense, it you know, wherever it is on their body it's just a mark."</p>

Parental ad. to birthmark

Accepting the birthmark	Noticing the birthmark less	Pride in baby	8	<p>Penny " I ended up with this like, I'm really proud of him and I love is birthmark."</p> <p>Tom "He is smiling, he is breathing, he is laughing, doing everything you should as a baby, He's absolutely perfect."</p> <p>Sally "I think he is a very attractive baby."</p> <p>Jack "it gives me a sense of pride the way she deals with people asking her questions that others don't have to deal with."</p>
Accepting the birthmark	Permanency of birthmark	Visibility of birthmark	13	<p>Penny "I mean, he's half red, like how often do you meet a baby that's half red."</p> <p>Kate "we were lucky that it's so small and wasn't raised and it didn't affect anything."</p> <p>Amy "when she is pretty side up loads of people would come up and say aww, what a pretty baby....um, and the when she's haemangioma side showing less people come up."</p>
Accepting the birthmark	Leaving a mark	Birthmark leaving a mark	6	<p>Hannah "it had a massive effect on our lives for such a long time"</p> <p>Kate "it's kind of a chapter in her life and when the chapter's closed we'll just shelve the book so to speak and then just get it out when we need to"</p>
Accepting the birthmark	Promoting birthmarks	Spreading the word (promoting birthmarks)	10	<p>Sally " so I suppose I've become quite evangelical about it really, people thinking it's just a birthmark, so from that point for view I've become a bit.... yeah yeah, spreading the word."</p> <p>Kate "now we know what we are looking out for a bit and should it happen to any others we have umm.... also anybody else we know we can give advice and support."</p> <p>Amy " I've suddenly become quite an expert at diagnosing everyone else's birthmarks."</p>
Accepting the birthmark	Leaving a mark	Feeling like this huge thing	5	<p>Penny "I didn't feel like anyone was taking me seriously with it, I thought maybe I was making a massive deal out of nothing, but to me it felt like this huge thing.".... " If anything I wish I felt like this when I first had him.... that I missed out on all of that really."</p> <p>Hannah "like in a way, it spoilt my enjoyment of her, like I was so excited to hear I was having a girl as I have a boy already and I really wanted a girl so I was really delighted but then, I dunno, I couldn't really enjoy her for those first few months."</p> <p>Kate "don't let that little mark or that big mark whatever it is, grind you down and so you know, so that you miss out on you know, whatever it is that your child is doing at that stage."</p>

Accepting the birthmark	Promoting birthmarks	Requesting better professional support/knowledge	13	<p>Tom "we had to learn ourselves, we haven't had any help from anyone really. It amazes us both how little people know, if we had a sort of group or something we'd definitely go to it."</p> <p>Charlie "Well....I mean....errr ...the biggest concern, and it's not just the birthmarks unfortunately but it is primarily, is the lack of medical knowledge. And errr, that was...that...errr that really worried us because we kept saying to people that there was something wrong or we kept hearing different things from people."</p> <p>Harry "we're not talking about cancer cures here but we're talking about something which is, is not difficult, .....it's simple, you know...it can be, you know, just a bit of education and we can avoid a load of problems."</p>
Feeling alone and uncertain	Isolating emotions	Isolating emotions	5	<p>Penny " I felt horrible guilt that I even felt like that, that felt horrible and you feel really lonely"...."so....so you can't talk, it's really lonely and you don't say it.".... "It feels wrong how you feel, that isn't how you imagine having a baby to feel, it's like you can't go and admit to someone that's how you feel."</p> <p>Hannah "I'm really cross with myself, or I was, not so much now, but at the time, once it ulcerated, I couldn't believe I'd been so stupid.</p>
Feeling alone and uncertain	Not knowing	What is it?	12	<p>Sally "she didn't quite know, said that it could be part of a traumatic birth or something but blatantly wasn't as it took barely an hour and half to have him."</p> <p>Amy "They weren't sure what it was but they thought maybe it was an abscess from the forceps injury."</p> <p>Sarah "Um, I thought it was a burn or something."</p> <p>Jack " they said oh there's bruising on her face and um, we'd had quite a prolonged labour and we thought maybe it was a bruise."</p> <p>Kate " they thought it was possibly a feeding rash as she was dribbling her milk and it was rubbing"</p> <p>Charlie "originally we thought it was....because she was forceps delivered, we thought it was something to do with that."</p>
Feeling alone and uncertain	Not knowing	Was it my fault?	5	<p>Penny "I went through this guilt stage um and sort of thinking oh god what if it's something I did? ... I'd smashed up the car and I thought what if the car crash, like what if I'd bumped it or something."</p> <p>Anna "and then when it isn't a perfect baby you think Ohh, what have I done?" ... "but before it happened I'd been clubbing twice in the time that I</p>

				think I was pregnant and for ages I was like arrgh, what if I've made this happen in some way."
Feeling alone and uncertain	Not knowing	Threat of the possible complications	13	<p>Penny "who knows and what if nobody can get to him in time and it goes on and on and that you know what you mean, so that's where my fear lay in the seizures more than anything"</p> <p>Sarah "it could be birthmark related and it's just not knowing, it's the waiting game to know if he's affected."</p> <p>Anna "He started mentioning Sturge Webber syndrome and Klippel Trenauney and lifelong disfigurement and we just freaked out big time."</p> <p>Kate " I remember thinking gosh does that mean it will affect her jaw, her teeth."</p> <p>Amy "then as the...haemangioma started to grow on the side of her face I guess...um as it started blocking her ear canal."</p>
Feeling alone and uncertain	Not knowing	"It's the not knowing"	10	<p>Penny "I wanted the appointments to come through quicker so I had an answer."</p> <p>Sarah "it could be birthmark related and it's just not knowing, it's the waiting game to know if he's affected.".... "I try not to worry about it and try not to think about it but I just want to know. I need to know, as when I know I can deal with it....and that's the hard bit, the not knowing."</p> <p>Hannah "Just looking at it all of the time, just didn't know what to do."</p> <p>Sally "we were in hospital, waiting for the referrals to come back and he had already contacted his friends who are paediatricians."</p>
Making sense of it	Making meaning	Adding personality	4	<p>Penny "If he gets angry, it's like a little hulk, but red!"</p> <p>Anna "It's a great barometer of her moods and health actually, when she's hot or angry its bright red and if she's cold its pale"</p>
Making sense of it	Putting it in perspective	Challenges of parenting a child with a 'difference' (comparison with others)	7	<p>Penny " I think it makes you realise what parents go through, not just with birthmarks but with other stuff as well"</p> <p>Tom "so obviously when we are looking at pictures on the internet of other kids. On O we can't see it as much as he's ours but we look at other kids and go bloody hell, theirs is bad."</p> <p>Sally "I've read all that Facebook group, awful stories about people who don't want to leave the</p>

				<p>house because of awful people just making it horrendous, it's heart breaking."</p> <p>Amy "I mean it felt odd because I was getting worried about what was wrong and I've had friends who have had far worse with their children."</p>
Making sense of it	Putting it in perspective	Feeling lucky	10	<p>Penny "We realise now how lucky we are that he's just got this, that he's just like that"</p> <p>Tom "I think you realise what parents go through a bit more, not, not with birthmarks but other stuff as well. We realise how lucky we are that he's just got this, that he's just like that."</p> <p>Kate "I think looking at photos on the support group where your seeing other children with it...it puts it in perspective how small and almost...ummm...what's the word...my brains gone."</p>
Making sense of it	Normalising experiences	Normalising the challenges	8	<p>Penny "she said whether or not you get a positive or negative result from an MRI you're still going to sit waiting because if they tell you he has got Sturge Webber, you'll look for a seizure and look for this and that but if he hasn't you're still going to look for it."</p> <p>Sally "say like I've had a really bad day and I've just been in tears because of these awful people making comments and it just normalises it."</p>
Making sense of it	Normalising experiences (linked to support groups)	Being in the same boat	7	<p>Penny " that's why joining the sites, the parents have given me like more information than anyone else because they are in the same boat as you."</p> <p>Anna "she knows that her facial appearance is a bit different to some other people but being on the Facebook group is great because I can show her pictures of other people who have similar special marks."</p> <p>Sally "people just realise that they are not the only ones, they are not isolated, they can put anything down really and people won't be judgemental."</p> <p>Kate " I mean you start talking to people and they say my so and so has one of those on this part of their body and they've left it and it was absolutely fine, kind of thing...you think oh it's one of those things that if only people talked about more."</p> <p>Charlie "because everyone's in the same boat and it was sort of comforting and quite a help that one of the nurses we see at GOSH was one of the founders."</p>
Making sense of it	Making meaning	What do we call it?	8	<p>Sally "We don't say special, your special mark because when you have more than one child, what's special about them?"</p> <p>Anna "well lots of um 'oh look at your beautiful</p>

				<p>mark' and talking positively about her mark."</p> <p>Kate "and they used to say that it means you've been kissed by an angel.</p> <p>Amy "I describe it as a tumour"</p>
Resources	Personal coping styles	"Not dwelling on it"	4	<p>Tom "I've taken a step back, I've sort of like, I don't know if I try not to think about it as much"... "I've just shut off to it as I feel he is what he is."</p> <p>Sally "I just thought, oh he'll be fine, I'd convinced myself there was nothing wrong, so I wasn't actually, maybe because I just wasn't letting myself think about it, denial or whatever."</p> <p>Kate "just kind of went with the flow and thought there's nothing that we can do to change it." .... "and there's nothing anyone can say to us that is going to make it any better, she's got it and we'll just have to deal with it."</p>
Resources	Support groups	Linking into support groups	9	<p>Penny "I found out most of the information from joining the Facebook sites. The sites I have been on have been amazing."</p> <p>Sally "and being told about that support group was definitely really helpful" ...."there are so many people on there err and people talking.....and everyone's just really helpful....and you're not just the only one....it's like any support group really, people just realise that they are not the only ones, they are not isolated, they can put anything done really and people won't be judgemental."</p>
Resources	Child factors	Child's personality	5	<p>Sally "he is such an engaging toddler, very smiley and people, you know, he does attract people, he's got that sort of.... charisma"</p> <p>Anna "She's such a robust character now that I know her, that she'll probably be fine. She will have some tough times but she...I think she's got the resilience to kind of....and the confidence to bounce back from it."</p>
Resources	Parent factors	Going privately	2	<p>Hannah "by then I had already found Dr G and booked an appointment with her at her private clinic on Saturday" ...." I was in such a state by then to be honest I said I just want to see Dr G as soon as possible, I don't care if we have to pay."</p> <p>Harry "So it wasn't until it really, really got very nasty and infected that suddenly we....sought, through a different angle, we went the specialist angle and we went private and saw a specific person in a posh hospital in London. And that was kind of ...right ok actually this is, this potentially we'll have to do something about....other than just wait till it goes away."</p>

Resource s	Medical system	Being passed around (disjointed care)	11	<p>Anna " by the whole thing of like having an optometrist, a dermatologist and a paediatrician, it's all a bit dis, can be a bit chaotic and disjointed."</p> <p>Sarah" she said from a skin point of view she's got no reason to refer him [for an MRI] and I said what about the blinking, what about the neurological symptoms and she said that's a paediatrician's job, that would be down to the paediatrician to do, to refer you, so now he's been referred in to the doctor's service."..."so he's nearly 6 months now and we've been on 2 waiting lists and seen by two separate people so it's quite difficult really."</p> <p>Penny " Then I wanted the appointments [paediatrician, dermatologist, ophthalmologist, geneticist] to come through quicker so I had an answer and that's when I saw the geneticist in the end... yeah 4.5 months is quite a long time to do before you get to a point and you sort of stop worrying about the Sturge Webber."</p> <p>Amy "it was all very frustrating when it felt like the midwife, the GP and A&amp;E all agreed there was something wrong and someone needed to look into it and someone needed to take ownership but no one seemed to agree who owned the problem."</p>
Resource s	Medical system	They just don't know about birthmark s	13	<p>Sally "She [health visitor] is there to pick up on things so I'd expect her to say your baby has extensive birthmarks, have you been to see someone as it might be an indicator for this or for that and she didn't say anything, appalling really."</p> <p>Hannah "it's really frustrating that people don't listen, why don't GP's know about this, why don't health visitors know about this? They are the first line coming into contact with these babies."</p> <p>Kate "then the health visitor was the first one who said it could be a birthmark but he'd only just qualified so he didn't have a lot of experience of what to do.... umm...so the doctors .... weren't particularly thinking it was a birthmark."</p> <p>Amy "we kept asking people in the hospital about them [birthmarks], they didn't really give many answers...um....indeed an obstetrician told us with complete confidence that they were forceps marks</p> <p>George "Well....I mean....errr ...the biggest concern, and it's not just the birthmarks unfortunately but it is primarily, is the lack of medical knowledge. And errr, that was...that....errr that really worried us because we kept saying to people that there was something wrong or we kept hearing different things from people."</p>
Resource s	Medical system	"She knew what she	7	<p>Sally "she was a very experienced midwife, she delivered my first one, so erm yeah, it helped that she was there and she knew what she was talking</p>

		was talking about"		about"  Harry "and whilst we were let down by, we felt a little bit that we were let down a little bit by the maternity unit, by the hospital, the people we had gone to see since we had genuine and very solid trust in."
Resources	Medical system	Poor medical professionals bedside manner	4	Anna " there was a consultant stood at the end of the bed peering into her cot, um...he wasn't a people person and he said all the wrong things."  Penny" so then I went to see a dermatologist at children's... the lady was terrible and I left there probably feeling the worst I'd felt because everyone else seemed to want to help... but this dermatologist, she was terrible, I sat down and she said what have you come to see me for? And I thought, you think to yourself, my babies half red, what do you mean what have I come to see you for?  Hannah "(Dr) just wafted in...she didn't really talk to me".... "I felt a bit un consulted....under consulted"
Resources	Medical system	Getting the ball rolling	10	Sally " so the paediatrician was good in terms of going straight off to get more information, get referrals straight away"... "Erm so yeah, the ball got rolling quite quickly in terms of getting appointments for things"  Kate "I put a photo on the birthmark support group and said somebody help us find what it is and someone from GOSH came through and said this is what it is I want to see her".... "I had a lot of contact with her over the Easter weekend...um...then went back to a GP who specialises in dermatology and said we want to be referred and she took one look and said OK and it was done and dusted."  James "I knew it was a port wine stain, er...pretty clear it's a port wine stain and then within err...I think it was probably about five minutes later...umm...I sent pictures to my friends who are paediatricians...umm, just to get confirmation."... "he said there were two things to look out for, the Sturge Webber or ummm, what's the other one...I keep forgetting it."
Resources	Personal	Parent factors	5	Sally "I'm a bit older, I had him when I was 40, nearly 41 actually".  Sarah "as an experienced mum like, I know if he is developing well, I don't need to see a doctor for that."  Anna "We'd never seen a freshly born baby anyway so it was all a bit new to us."

Parental ad. to birthmark

Resources	Personal	Personality types	7	<p>Sally "I'm a practical person"</p> <p>Hannah "I'm a bit of a panicker".... "I'm quite a control freak"</p> <p>Amy "My husband and I are quite intellectual."</p>
Resources	Support groups	Birthmark related resources	3	<p>Anna " We got his book called Sam's birthmark"</p> <p>Sally " we try like more of the role play and we might get that book called Sam's Birthmark ad we have a teddy from that Changing Faces charity.....a teddy bear with a pink bit on its face."</p>
Resources	Support	In it together (parents)	10	<p>Charlie "I would say we have got stronger as a couple ummm because we are both dealing with it at the same time."</p> <p>Hannah "R is a lot calmer than me and he is good at calming me down and giving me a hug or whatever and telling me I was doing the right thing."</p> <p>Harry "just talking about it quite a lot, that helped just because,.....you know...its only something that she saw or I saw, we both had a look at it, do you think it's getting bigger, do you think it's....do you think it's improving...it's improving, yeah all that kind of stuff its just kind of talking about it quite a lot."</p> <p>James "I guess it's not all the worries it's all the details of the different syndromes particularly. Why didn't I tell her? Because I know she would just worry and we didn't know and there was no purpose behind it."</p> <p>Harry "I think we, we did talk about it a lot, we tried to sort of .....just be as .....be as, open about it as we could in terms of our own feelings about it and yes we were upset, but no we both were very much, this is what we've got to do isn't it ...and I, I don't think either of us ever questioned it ...but...you know...talking about it also just reinforced it that we sort of ...can't...can't not, you know."</p>
Resources	Support	Feeling supported by friends	5	<p>Penny "They were really good like that was....like your real friends talk openly to you about it."....."They just loved him instantly and it didn't matter and that was lovely,"</p> <p>Sally.... "being able to talk about it normally with them"</p>
Resources	Support	Having family support	6	<p>Sally "in the early days I'd speak to my sister and be quite upset about it. ".... "she was really supportive, really helped me when I was feeling weepy she would panda to me."</p> <p>Kate I mean obviously my parents have been really supportive and wanted, obviously to get her sorted."</p>

				Charlie "mum being 20 mins down the road was quite comforting"
Resources	Support	Not as supportive as they could have been	5	<p>Kate "His parents weren't quite so, which is quite surprising as his mum's a nurse and she kind of pooh poohed it a little bit."</p> <p>Amy "she [mother in law] keeps talking again about putting make up on her when she is older, she made a comment about, oh can she take a photo of the haemangioma to show her sister but don't worry she won't show anyone else and obviously it won't end up in the family album."....."I'm not really sure who has been my support."</p>
Seeking to understand	Having answers	Putting one's mind to rest	9	<p>Penny "the glaucoma was fine; I was alright considering he'd given me an answer. Then he told me yeah, he probably will....so then I could deal with that.... "but when I spoke to the genetic lady she put my mind to rest with the Sturge Webber."</p> <p>Kate – "once we knew it was a birthmark it was like OK"</p>
Seeking to understand	Information searching	What to do? (waiting for appointments etc.)	9	<p>Penny – "So then we went to a dermatologist at children's...I'd had to chase that up as nobody got back to me."</p> <p>Hannah "I didn't know what to do, so I tried phoning A&amp;E and GOSH and H."</p>
Seeking to understand	Information searching	Googling	12	<p>Penny "I just googled all of that again"... "I've left hospital with a piece of paper and just googled everything"</p> <p>Tom "can only look at other kids on the internet and see pictures on the internet and stuff like that."</p> <p>Sarah "That's my thing at the moment, googling things and research them quite extensively."</p> <p>Hannah "I thought oh my god, I didn't know what to do and at that point I started googling it crazily."... "I was on the Facebook group and googling all the time and constantly looking on the GOSH website."</p> <p>Kate " just, you know, get straight in there and probably for me is to research things a bit more."</p>
Seeking to understand	The 'knowing' dilemma'	Knowledge empowers	9	<p>Penny "We've learned loads, yeah I'm like a doctor now"</p> <p>Sarah " I'd already read into birthmarks and how if they are midline they can affect you more so straight away I was worried about it being near the brain."...." I'm just really clued up on things and I find that the best way to be really, to be mentally prepared."</p> <p>Hannah....Because now I know much better that</p>

				things you would do for a normal wound you can't do with a haemangioma."
Seeking to understand	The 'knowing' dilemma'	Knowing increases worry	9	<p>Penny "I think I knew more about it because I researched more and the less you know about something the better."</p> <p>Sarah " I'd already read into birthmarks and how if they are midline they can affect you more so straight away I was worried about it being near the brain.".... "and when he was born with this birthmark on his face on his midline I was worried that he was affected neurologically, so um.... straight away when he was born and no one could tell me what it was, so now I was panicking."</p> <p>Hannah "I found myself spending hours on, I, people were, I was beginning to get a bit affected by the scare stories, winding myself up a bit."</p> <p>Charlie "I mean the internet is full of things to scare you."</p>
Social challenges	Societal influence	Gender differences	8	<p>Sally "I think it helps that he's a boy to be honest and not a girl bearing in mind appearances."</p> <p>Hannah "I know it's only aesthetic, I know that but, she's a girl and aesthetically its important, like she's a girl."</p> <p>George "because I am concerned about the cosmetic side, you know, I don't think a girl would want to grow up with something like that."</p>
Social challenges	Feeling Judged	Parents feeling self-conscious	6	<p>Hannah "I didn't want to go out or take her into shops in case of what people might say"</p> <p>Sally "at the moment we don't get a great deal of comments, apart from double takes in the street and things like that, it does make you feel a bit weird, yeah, it's strange." ...."we went to Centre Parcs over the summer and we were in the pool at the time and I um, no, I don't think anyone said anything actually which was good, I think I was on edge so yeah relieved."</p> <p>Amy "I felt very self-conscious about it, I remember feeling nervous about going to meet people at work."</p> <p>George "you see people seeing and thinking Oh my God what have the parents done to this poor child. So we do get a lot of people wondering if this is an injury, wondering if it's because of parents."</p>
Social challenges	Feeling Judged	Fearing other's reactions	9	<p>Penny "you have completely irrational thoughts and then it was fear of what other people thought."</p> <p>Jack "It can be quite a powerful thing for them to hear, it can be scary I suppose just because you</p>

				<p>don't know what is going to come out of the stranger's mouth."</p> <p>Anna "I think it's harder for B when people insinuate he's injured or hurt her in some way."</p> <p>Harry "It's not, it's not so much wanting to fit in or, not wanting people to think you are different. It's more a .....(sigh)....what can I say.....it's not wanting, its wanting people to think that you, well not wanting people to question your, how much you care for your child. Because you know you care beyond any....you know, ...any tangible... any absolute....and it's the thought that people might think that you might do anything which might have led to this."</p>
Social challenges	Experiencing unwanted reactions	Experiencing unwanted reactions	13	<p>Penny - "like the older generation are funny as they will almost feel sorry for him, which if someone my age said that to me, I don't know how I'd react, well I do but I can't say it!"</p> <p>Jack " one time when a kid asked me what I'd done to her and practically accused me of I don't know what, I didn't really stop to ask."</p> <p>Sally "it's the nosiness and peering kind of interest I can't stand."</p> <p>Amy " one guy who peered in and at the time it was a very rare time when she was haemangioma side up and he looked in and went ugh and just walked off."</p>
Social challenges	Societal influence	An Asian thing	2	<p>Jack "We've had some weird conversations with people from different cultures that are always quite interested, like people from...I'm going to say India....as a continent Asia covers it. I've had 2 of 3 people say it's really good luck to have a facial birthmark.... but then J had a man of Pakistani decent or something similar and he was really rude about it."</p> <p>Anna "I don't know what language she spoke but it wasn't English and her son who was in his 40's stopped me, sort of putting his hand out like I need to talk to you. It started out about a mile a minute and he was translating and telling me.....But then she said she looks like the Phantom of the Opera ad I was just really upset."</p>
Social challenges	Experiencing unwanted reactions	Natural reactions (its human nature to look)	4	<p>Penny "it's a natural reaction for a child to go err if they see something they don't like and I haven't had a child say that to me"</p> <p>Sally "even though we would double take if we saw something, it's not malicious most of the time, just instinctive really."</p>

Social challenges	Societal influence	Thinking you should want it gone	8	<p>Penny "They almost think that we should want it gone, we don't want it gone, well, at the beginning....and they almost talk to you as if you don't want it there."</p> <p>Anna " when a kid asked me what I'd done to her and practically accused me of I don't know what, I didn't really stop to ask."</p> <p>Charlie "We had photos taken went she was.... just as it...before we started treatment and the photographer was like saying I assume you want me to air brush the birthmark out and it's like no, it's part of, part of her, we don't touch that at all."</p> <p>Amy "do you want us to airbrush the birthmark for you and....I find that sort of reaction really annoying."</p>
Social challenges	Societal influence	Considering 'difference' in society	9	<p>Penny "I think it's a generational thing and it's that lack of knowledge and that's it." .... "I hope things like this are more accepted in society anyway".</p> <p>Tom " Schools have moved on a lot now and um, like all nationalities going to primary school and high school"...." like, I never seen an Indian boy until I went to high school.... Maybe they'll just think he's like a different shade!"</p> <p>Anna "Every time someone said it's nothing, its fine, it actually made me feel okay, society is receiving her more positively than I anticipated."</p> <p>Amy "Well for a start it's harder to cover up on your face, I think something about society that you look at facial defects as being ugly."</p>
Social challenges	Societal influence	Social concerns influencing treatment choices	10	<p>Tom "I just think what difference does it mean being half red? And we've said that we wouldn't do anything.... wouldn't try the laser if he could go from 6 to I dunno 20 and get past the teenage years."</p> <p>Amy "I would have a strong motivation to do it [treatment] for my own personal reasons so I don't have that going into work and everyone judging me and saying what did you do wrong during pregnancy...um...how have you ended up with a freak child."</p>
The shock of it	Feeling shocked	Needing to release emotions	5	<p>Sally "In the early days I was almost hoping someone would say something so I could go grrr you know, get really irate." ..... "I think in the first few weeks when I was really tearful about it, um, and then you feel a bit weepy, well I did feel quite weepy in those first few months."</p> <p>Hannah "I mean I cried and cried, he wasn't there, he was doing something with my husband and they go back and I was holding her just sobbing."</p>

The shock of it	Expecting perfection	Expecting perfection	5	<p>Penny "We knew he'd be big so you've got a vision in your head and he came out and he wasn't what I considered at the time my perfect baby"...."everyone says when their born you've never felt anything like, like their perfect. I looked at home and to me he wasn't and I think that was really hard."</p> <p>Sally "not that you want perfection, but you know"</p> <p>Anna " I think everyone expects to have a perfect baby don't they?"</p> <p>Amy "and I was just going silently well objectively no she's not perfect because of ..you know...there's not a lot to judge a baby on, you can only really judge them on their physical appearance, they don't have a personality, not at, you know, six weeks before their due date, they're quite slug like."</p>
The shock of it	Expecting perfection	Finding the words	5	<p>Penny "I looked at him and to me he wasn't [perfect] and I think that's really hard, like that you can't really...I've never said that before because you're not meant to say that.".... "so...so you can't talk. It's really lonely and you don't say it."</p> <p>Harry "I think we, we did talk about it a lot, we tried to sort of .....just be as .....be as, be as open about it as we could in terms of our own feelings about it and yes we were upset."</p>
The shock of it	Feeling shocked	(Initially finding out) Overwhelming feelings	7	<p>Penny "There wasn't an obvious emotion. It was sort of everything"</p> <p>Hannah "I was in such a state by then to be honest I said I just want to see Dr G as soon as possible."</p>
The shock of it	Expecting perfection	"Coming to terms"	10	<p>Penny " it's coming to terms, I don't know if this sounds terrible...but you picture your baby and what they will look like....and when he came out he wasn't what I considered at the time my perfect baby" .... "I think that took me time to get used to, he wasn't when he was born"</p> <p>Hannah "we weren't expecting that to have happened, it was like oh my goodness."</p> <p>Anna "you've got this gorgeous little baby that you've just made, that you've waited a long, long time for and is wonderful, you're going to like it, aren't you, eventually. It's a bit of a shock, don't get me wrong, it is a shock but I wouldn't say it's a disappointment, it's just a surprise we weren't prepared for."</p>
The shock of it	Feeling shocked	Feeling shocked	7	<p>Sally "so yes when he came out the midwife said oh it looks like he's got a mark on his face and it was like oh and obviously all over his body as well and err, yeah that was a bit of a shock."...."as soon as he was handed to me I looked at him and errm, yeah, you don't yeah, it was a bit of a shock."...</p>

				"but I suppose it was a bit of a shock as you weren't expecting it and didn't know what it was.".... " I was a bit stunned really, I think. Later on you get this kind of...it used to shock me a new each time I saw it"
Thinking ahead	Protectionism	"Getting waves of protectionism"	6	Penny "I just thought that's it, maybe I should home school him and then you think hold on I can't shelter the poor kid from everything."  Sally "I don't know how bad or good it will be; I sometimes get waves that kind of.... protectionism.".... "but um, yeah you do think what if it gets worse, what do you do? So you do get very protective."
Thinking ahead	How to talk about it	"Getting in there first" (explaining to others)	5	Penny " you find yourself sort of, when people meet him just going oh yeah and obviously he's got birthmarks, just to...to.... I dunno, it's just to sort of break the ice."  Anna "I rang my mum and the first thing I said was id had a little girl and the second thing was she's got a facial birthmark because I thought I don't want mum to come in and go urgh what's that?!"
Thinking ahead	How to talk about it	Allowing people to talk about it	3	Penny " You can talk about it, like we want you to talk about it. It's obvious, do you know what I mean, he's half red and that's fine."
Thinking ahead	Managing others reactions	Managing children's reactions	7	Penny "just let them ask, their kids, if they say why is he red you just explain what you can and if they say he's red, I say Oh yes he is, they don't mean it nastily."  Tom " we get lots of kids asking and you spend the time to tell they exactly what it is"....."You try to tell them as much as you can"  Kate " these were special needs boys with behavioural problems so they could have been quite cruel...as soon as you were just, they could tell you were being honest and you put it in child friendly terms that they could understand and that curiosity was over and done with."
Thinking ahead	Managing others reactions	Managing unwanted reactions	10	Jack - " I just blurted back that I hadn't done anything and that it was a birthmark and that she was born with it."  Sally - "It just makes my husband turn around and give them a funny look.".... " A couple of people have peered in and said different things and I've just stayed even tempered and walked away."
Thinking ahead	How to talk about it	What am I going to say?	8	Penny " I used to sit there on the night planning conversations and things that people might say when I'm out and thinking about how am I going to react to that."  Sally "I do sometimes visualise in my head, you know, what if this happened or what if that happened, you start thinking about all of your prepared responses and things like that and just

				hope that you don't end up saying well look at yourself you ugly git or something really childish like have you looked in a mirror recently!"
Thinking ahead	How to talk about it	Planning how to prepare the child ("there are stupid people")	6	<p>Tom "Let him know that people at times may say stuff about it and the other thing we've considered is drilling into him that no one is perfect."</p> <p>Anna " We got his book called Sam's birthmark"...."we've learned it off by heart so whenever anybody asks us at playgroup or anyone asks A she knows what to say."</p> <p>Sally "its explaining to both of them because I've usually got them both and T is older you know, that there are stupid people who don't know what they are saying and all that kind of stuff."</p> <p>James "I think we've talked about just not making a big deal of it, making clear it's part of him, it's just who he is and we've all got birthmarks and its immaterial really....ummmm...at the end of the day...ummm...if people have got a problem with it it's more their problem than it is his. Ummm...he is who he is and he needs to just enjoy who he is, at the end of the day is...I think our view on it."</p>
Thinking ahead	Strangers mistakes	Strangers mistakes	4	<p>Sally "there are sometimes those people say oh my cousin had that and it faded and blah blah blah and I have to say no it's not true in S's case."</p> <p>Jack "they will always ask the question, is that a strawberry haemangioma or oh that's going to fade isn't it, like my brother had one of those and it faded and no it's like no, it's not going to fade. That's an awkward conversation to have."</p>
Thinking ahead	Worrying about the future	Wondering what will happen	13	<p>Penny "I think your biggest worry is what his future is going to be like.".... "I think the anxiety of the first day of school is going to be completely different for us" .... "I think I will give him something for others to pick on him for, but I do know we can't spend the rest of his life worrying about it."</p> <p>Hannah "I was starting to feel a bit like, was it going to work and I was really worried it was going to scar, just didn't know what would happen."</p>
Treatment	To treat or not to treat?	Making decisions in baby's best interest	12	<p>Penny "the choices that you make are for his future and when he's 16 and turns around to me and goes mum why didn't you try that".... "what if you wait and then it's too late.... well not too late but you lose the opportunity when it is most effective."</p> <p>Anna " The consultant kind of said it's a no brainer to go with it, he never meets a teenager who wishes their mark was bigger or darker."</p> <p>Anna2 "it's hard to reconcile what becomes your perfect baby when you adjust your thinking and</p>

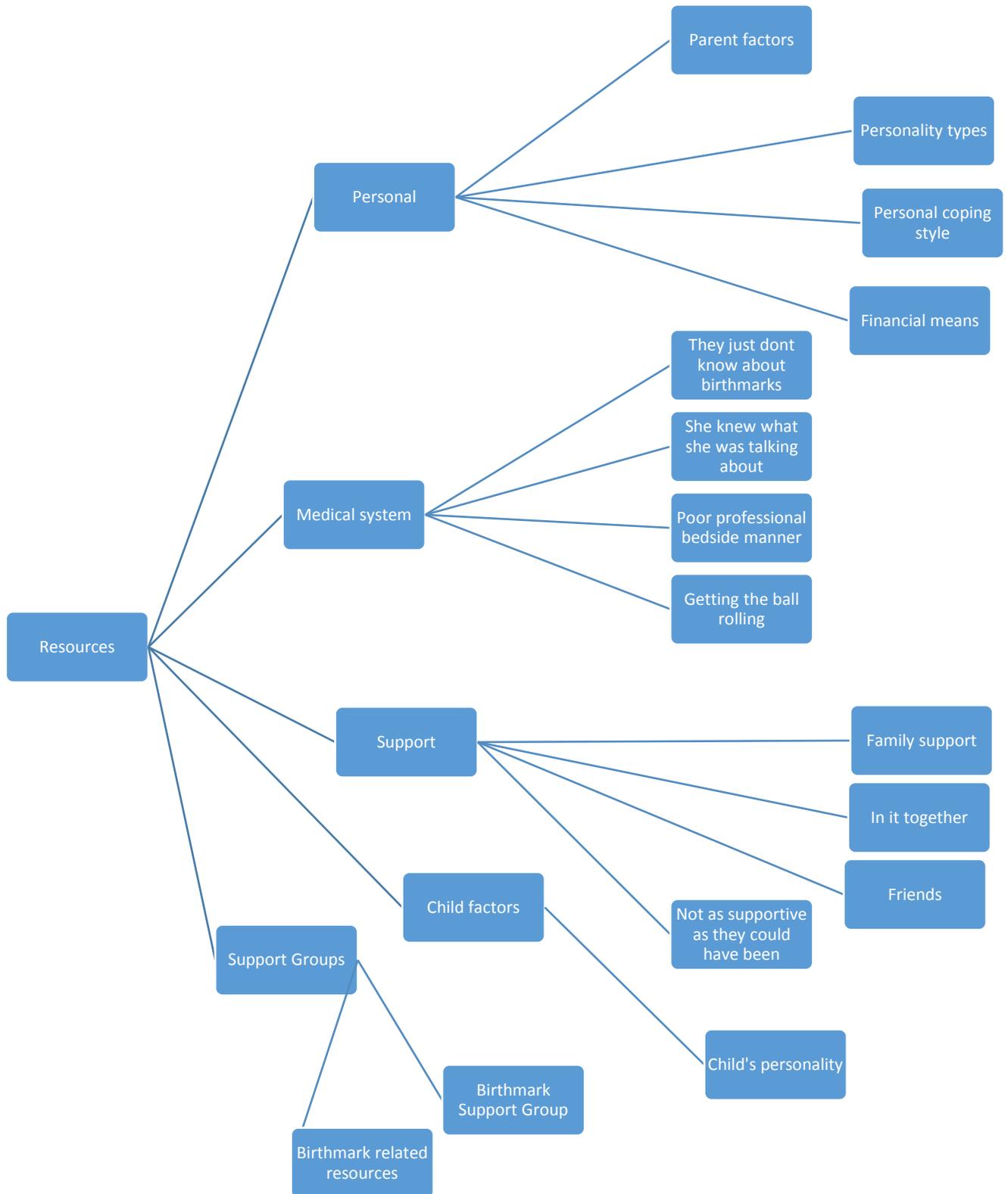
				<p>you realise she is perfect and that's exactly how she is supposed to be with the fact that you're trying to do something medical to change her."</p> <p>Amy "there was a functional reason why we had to take the medication for T's birthmark because we really should have taken, if it was cosmetic, maybe we wouldn't have....since haemangiomas are temporarily cosmetic issues we would not risk medication."</p> <p>James "I think that would be good, just so it's about ummm....only on the face he'd have it done, he'd have laser. So I think that would be good to have it because that's the bit that, you know, is....in your face....ummm.....so ummm....Yeah I think it would be good because it would may minimize what people's initial reaction would be to it."</p>
Treatment	Treatment expectations	Hopes for treatment	10	<p>Penny "We look at the laser not as getting rid of it but maintaining it so it doesn't thicken or darken, that's my hope for the laser."</p> <p>Sally "it will fade"</p> <p>Amy "better to give her less invasive treatment when she was younger to save her having to go through something a lot more traumatic when she is older."</p>
Treatment	To treat or not to treat?	Taking 1 step at a time	8	<p>Tom "now that we get to see a cosmetic surgeon and that will be able to start making some real decisions about it, about whether were going to do it."</p> <p>Kate "they've talked about laser when she's older if there's a need, little red veins left.... but cross that bridge when we come to it."</p> <p>Charlie "but you just cross that bridge when you get to it, you think right, let's take one step at a time and see what happens."</p>
Treatment	To treat or not to treat?	Risk of treatment	12	<p>Anna "but for us really the idea of her having a general anaesthetic for cosmetic reasons is quite hard to swallow."</p> <p>Charlie "cos when they sort of said it's a beta blocker...and it, the treatment that they gave was a beta blocker you instantly then, if you start researching what a beta blocker is well that's to do with heart attacks or something because it slows down the blood vessels. You think oh what, what should we really be putting this on our baby."</p> <p>Amy " I had concerns about Propranolol, the treatment they use has only been going for six years so there no long term studies into the impact."</p> <p>James "I think the only ummm thing is the anaesthetic risk for a child that young, that's all</p>

Parental ad. to birthmark

				.....ummm....he's not going to remember much else to be honest."
Treatment	Treatment management	Treatment management and routine	9	<p>Charlie "It became a routine, it was like right, she needs the treatment on 2 times a day, so morning and night"</p> <p>Hannah " I got into a good routine in the morning with the bandages and I got a good relationship going with then girls in Boots, they would always order me in an extra bottle."</p>

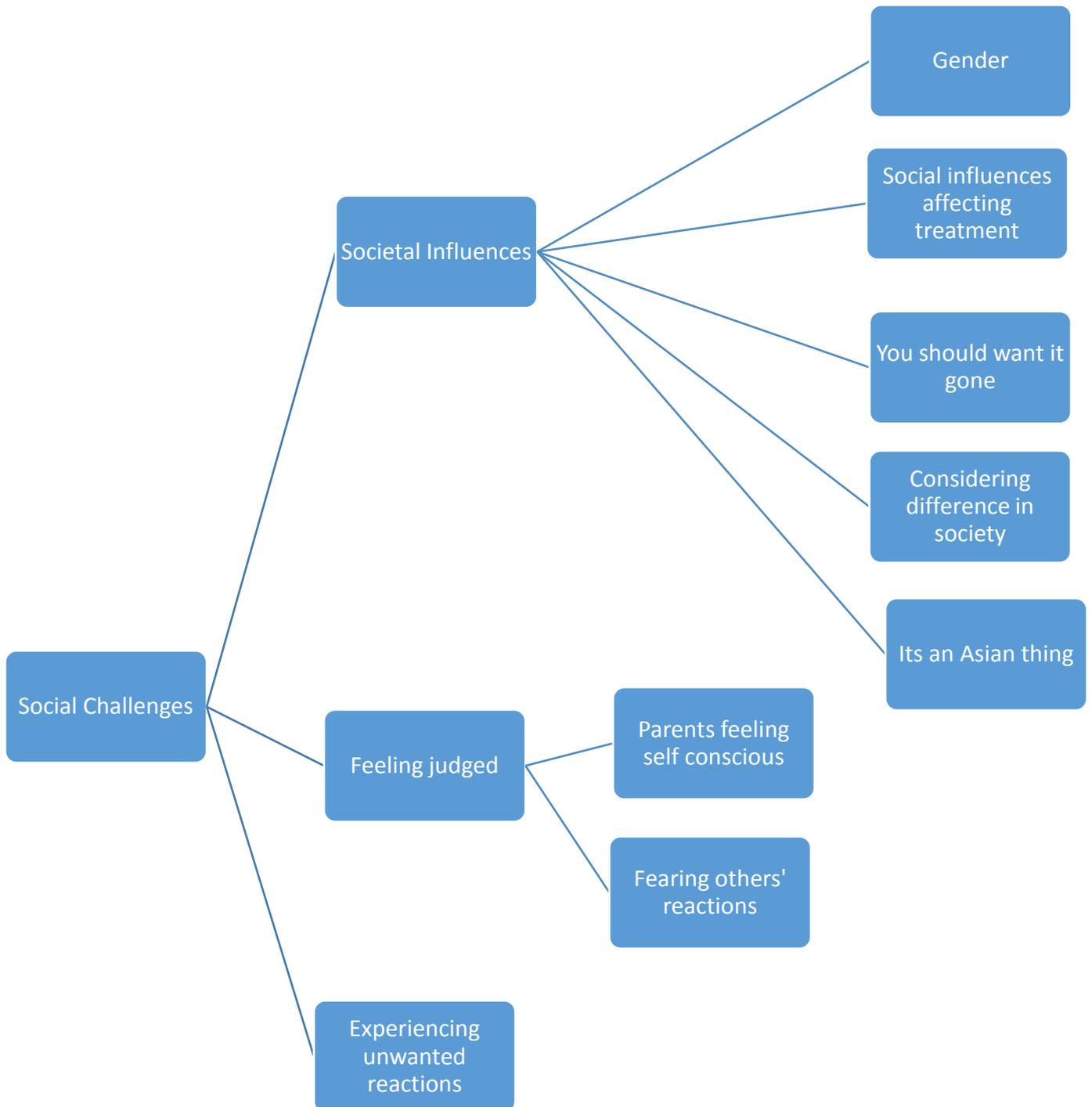
Appendix F iv: Models of categories, subcategories and focussed codes

Resources

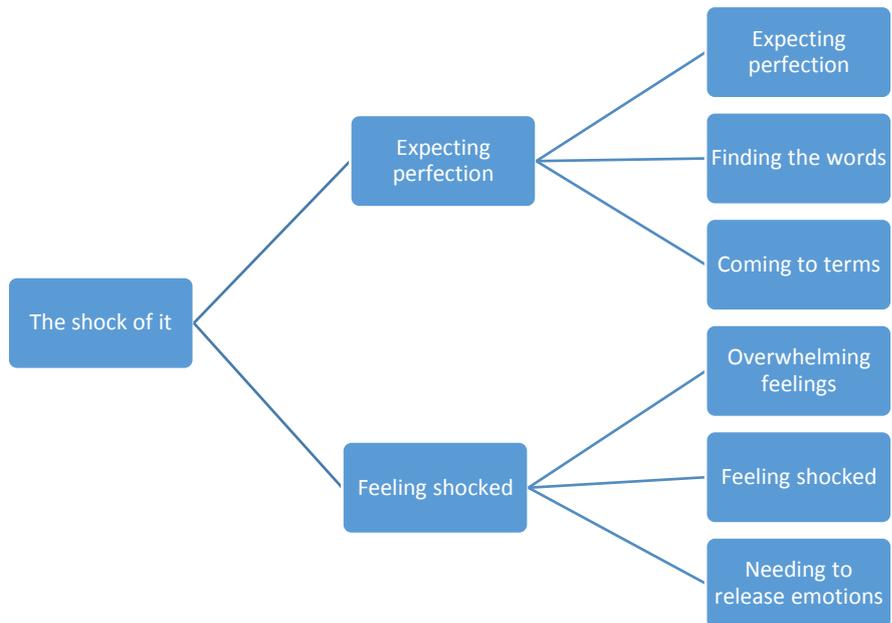


Parental ad. to birthmark

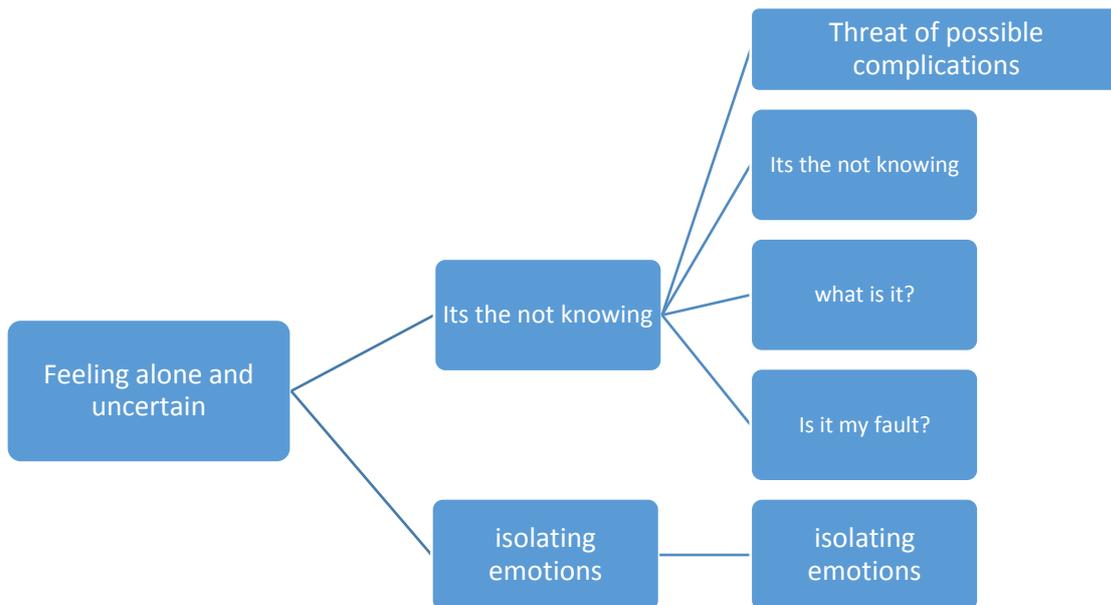
Social Challenges



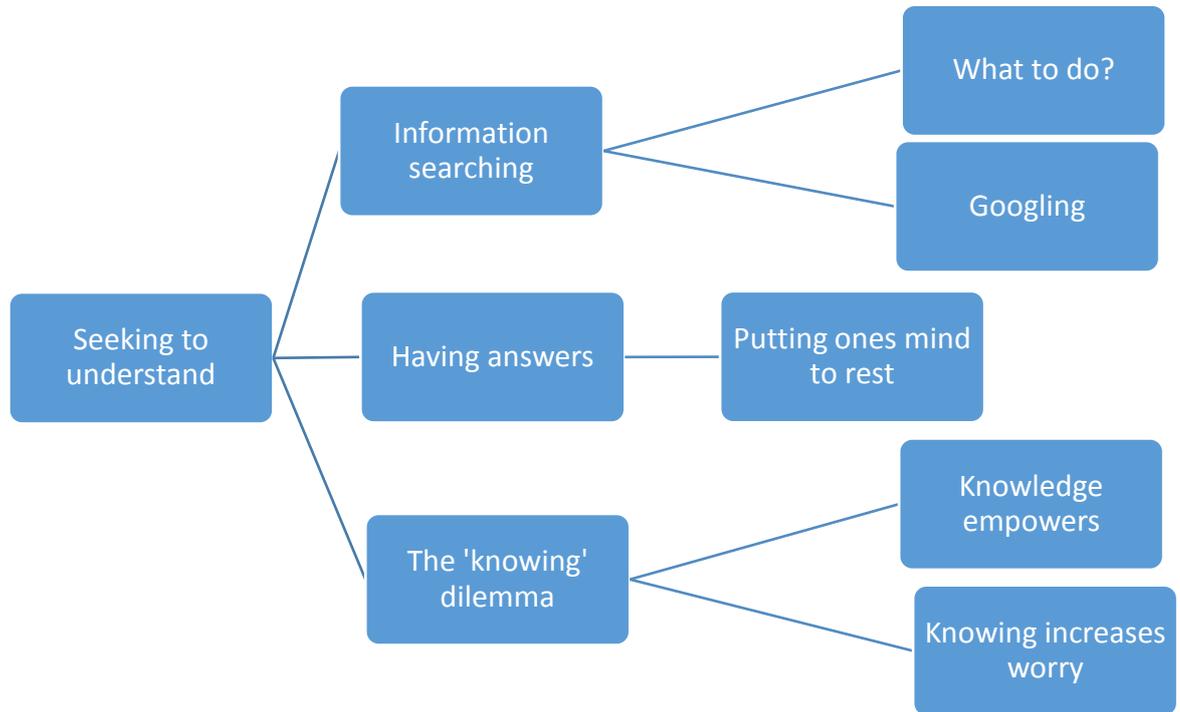
The shock of it



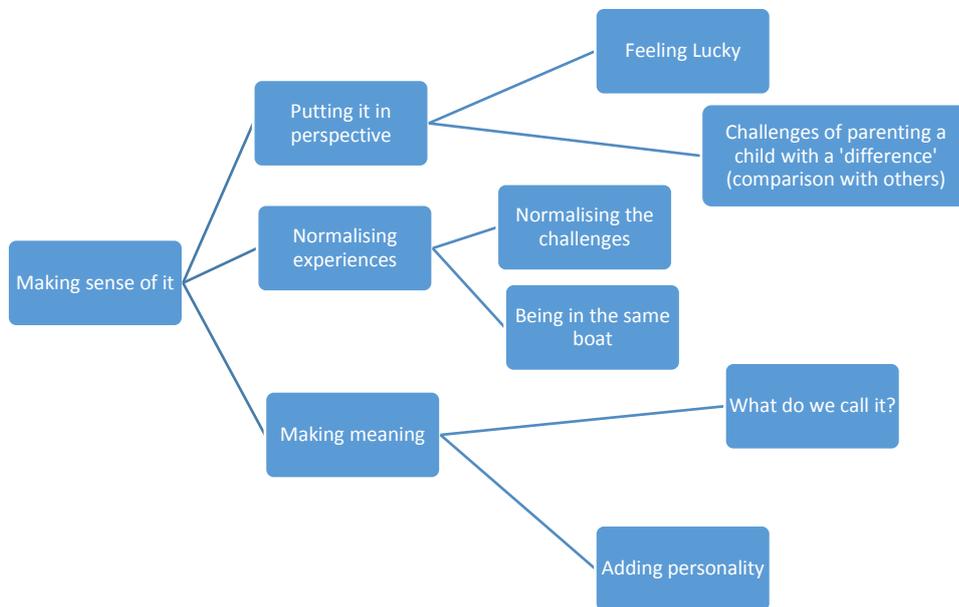
Feeling alone and uncertain



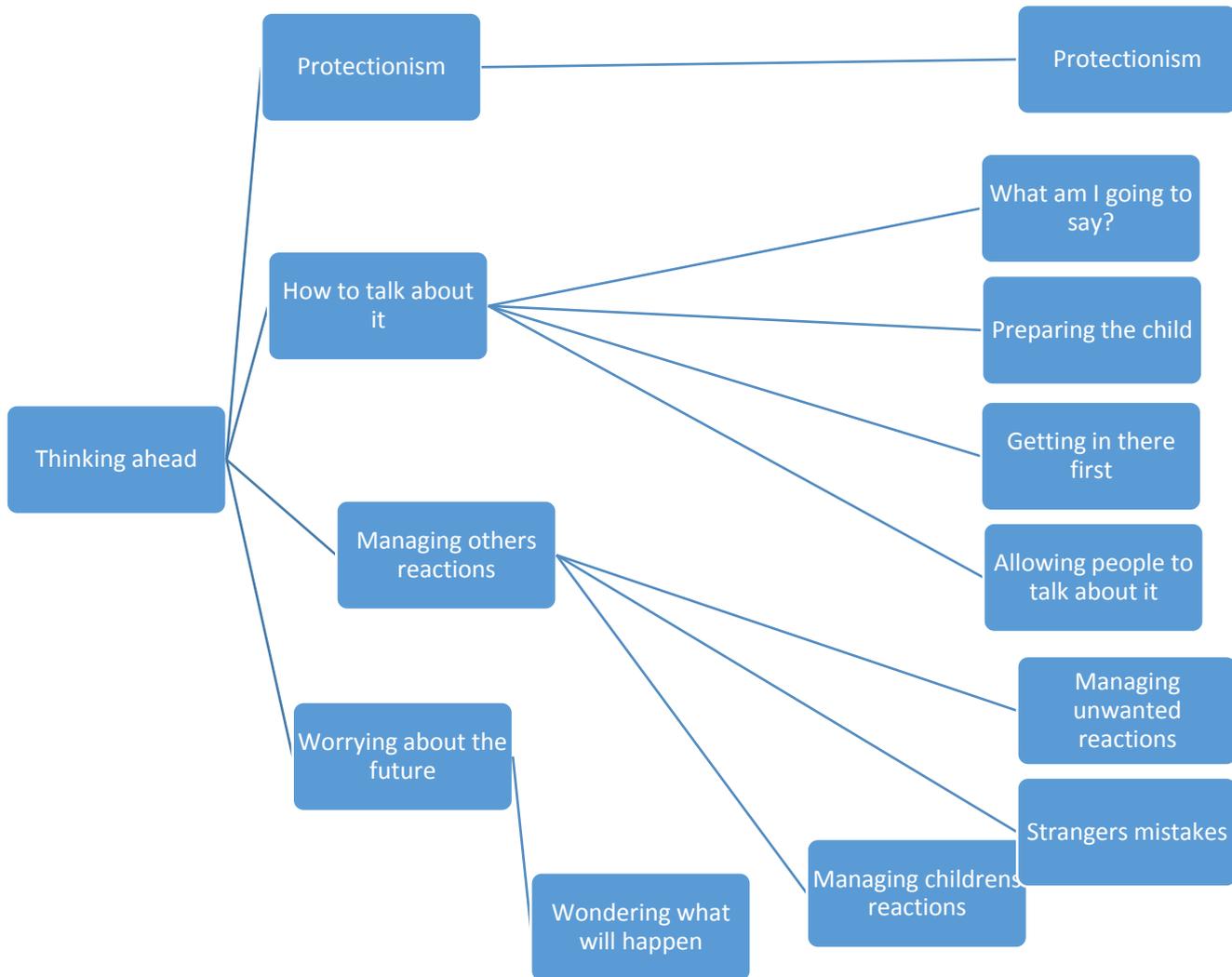
Seeking to understand



Making sense of it

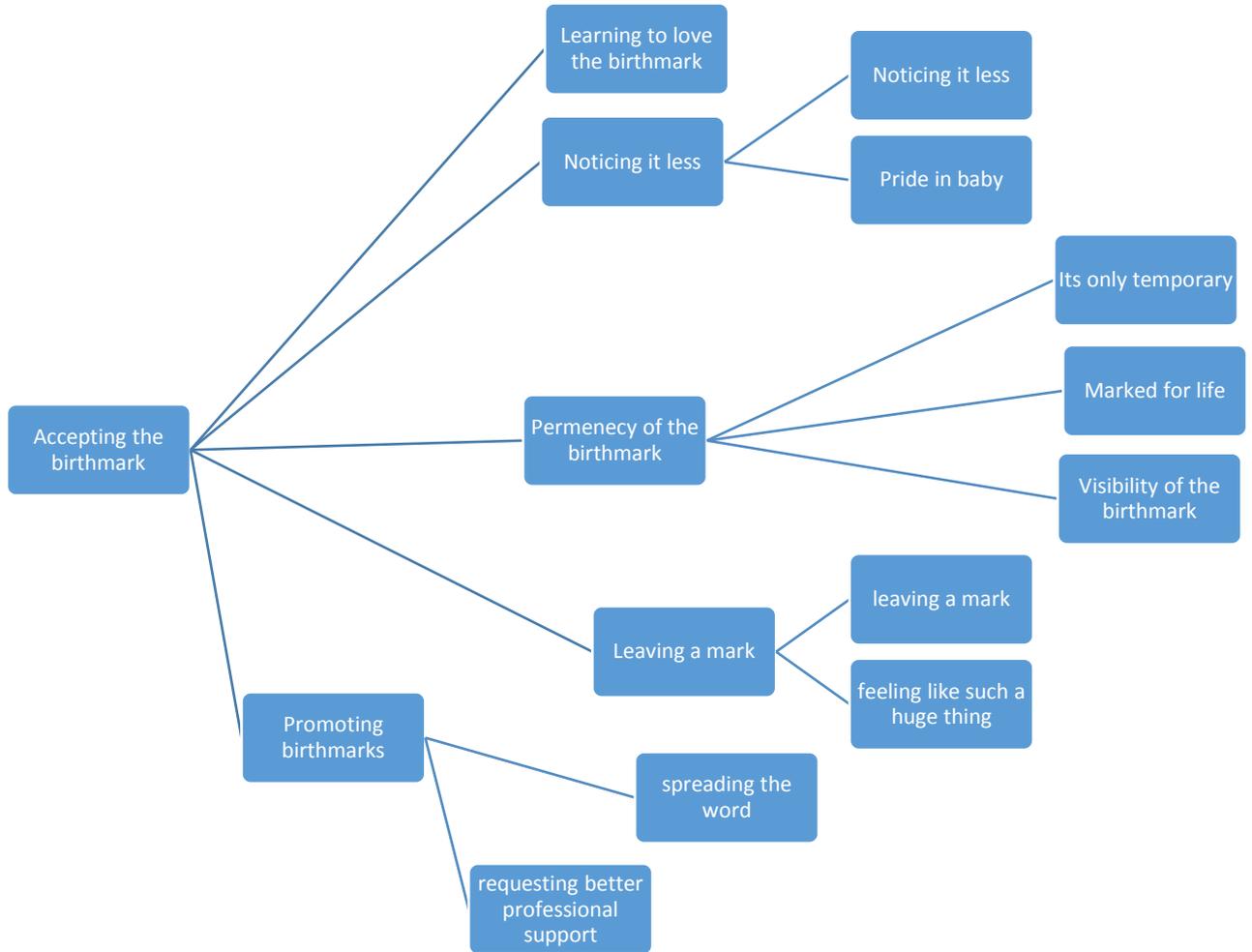


Thinking ahead

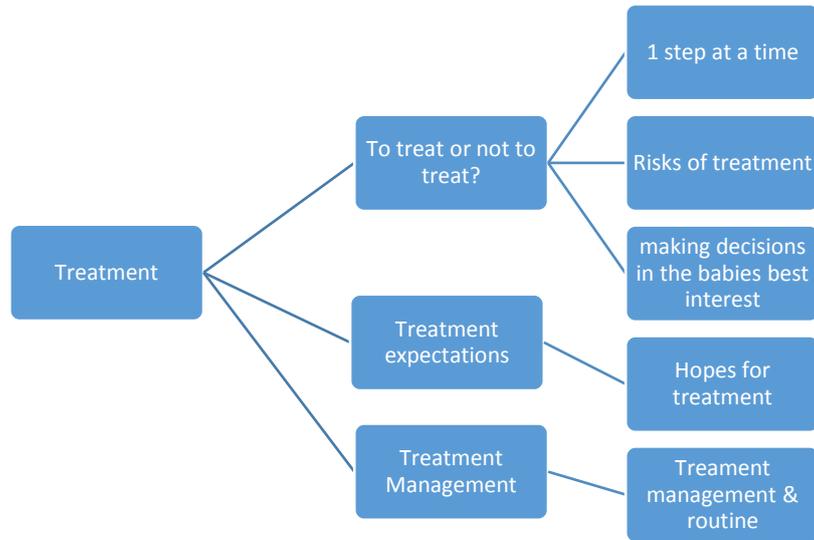


Parental ad. to birthmark

Accepting the birthmark



Treatment



## Appendix G: Participant's Journeys

I will outline the journey of two parents from different families in order to illustrate the model.

### Participant 1

Sally's child was the second child born to her and her husband. She described herself to be a bit older than other mothers as she was 41 years old. She was married to a doctor who had friends who were Paediatricians. She had a good relationship with her sister who was a source of support for her.

When her son was born, she was quickly informed by a midwife that he had a PWS and this was swiftly confirmed by the husband's medical contacts. She initially found this hard to come to terms with and reported feeling "shocked anew" each time she saw it. The questions that she had about the birthmark and the uncertainty she faced was eased by having referrals to appropriate professionals made swiftly. In addition to their medical contacts, getting in touch with the Birthmark Support Group enabled a clearer understanding of the birthmark and the associated syndrome risks. She was able to put the birthmark in perspective and did not face many social challenges, apart from the occasional stare or comment, however she reported this to be "heart sinking" when it happened. She described herself to be a practical person, which was an asset to her as she began to think ahead and plan how to support her son and his sister to manage any remarks from others that they may encounter as he grows up. She was also in the early stages of treatment and was considering the meaning of this for their son. She described herself to be very proud of him and spoke of noticing the birthmark less over time, which may indicate that she had accepted the birthmark. She spoke of feeling evangelical about the birthmark and wishing to promote wider understanding of birthmarks. She also spoke about wishing for medical professionals to be better educated about birthmarks.

### Participant 2

Hannah's daughter was her second child and first daughter. She developed a severe haemangioma within a few days of birth. She was told that the red marks visible at birth were insignificant and these were not marked down on the child's medical notes. The haemangioma grew within a few weeks, which shocked her but she was slow to seek medical advice as the doctors had suggested it wasn't a concern. She had many questions and felt thoroughly unprepared so she sought advice from the internet and found the Birthmark Support Group. Following this it ulcerated and when she finally was given a diagnosis, through accessing medical specialists privately, she was given the wrong treatment. She became stuck in the 'seeking to understand' as she was initially given conflicting advice and found it difficult to make sense of the

birthmark. She found the Birthmark Support Group a great source of support, normalising her experiences, however, at times she found that the stories that others were posting would affect her and make her quite fearful of what could happen to her own daughter. It was apparent that she became quite socially isolated during this time and felt very self-conscious about the birthmark, fearing how others may judge her for it. She described herself as a panicker and a control freak and it may be that these personality traits factored into the way that she coped, isolating herself and becoming quite socially avoidant. She described her husband to have been a great source of support to her throughout. Unlike many other parents who spoke about planning ahead and thinking about how to cope with the reactions from members of the public, it appeared as though this mother did not do this. This may be related to why she found it difficult to move forwards. The treatment regime for this child was also intensive and the Propanolol was an additional source of concern for this mother. Her information searching may have helped, but also hindered her as she agreed at times it became excessive. It seems as though this mother did not ever fully adjust to the birthmark, instead she was left with feelings of guilt that she had not acted quicker and blamed herself that her daughter will now have a scar from the ulceration even after the birthmark has resolved. Despite her daughter's birthmark having nearly healed, she described how she was still very active on the Birthmark Support Group and intended to continue to promote understanding of birthmarks with friends to make sure no one she knows is given the wrong information like she was.

Appendix H: Summary of Interview with a Clinical Nurse Specialist.

Background of the participant:

She is a Clinical Nurse Specialist working in a specialist centre for birthmarks, skin conditions and vascular disorders since 2002.

Key findings from her clinical practice:

1. In her experience, many of the parents she has encountered have been given inaccurate advice about what the birthmark is and that doctors such as GP's are not aware that they can refer to specialist centres, instead they think they need to refer to local hospitals. Doctors need to be more aware of referral pathways. She also highlighted that specialist centres will offer consultation to GP's and hospitals but this is not widely known.
2. Parents struggle to know how to respond to the comments that people get from the public.
3. As a professional in the field she emphasised the need for parents to have resources to support them to cope. She suggested having a card, such as a business card with an explanation of the birthmark to hand out to people.
4. Has a lot of parents who want a "quick fix" crying down the phone, being upset in clinic, struggling to accept their child's birthmark is life-long (PWS).
5. Debate whether laser treatment for babies/young children is appropriate as the child may have been through many surgeries for little difference in appearance and reduction of redness (PWS specific). Described a recent change in practice of now doing a maximum of 4 treatments and then reviewing progress before continuing treatment.
6. Majority of parents in her experience want to get rid of their child's birthmark, aim to "get rid of it" before the child starts school to reduce concerns about bullying and social stigma. But she had not noticed any patterns about which parents were more or less likely to want treatment vs those who chose not to treat.
7. She described the trend in parents with children with haemangiomas are now choosing to treat even when it is not necessary (e.g.. not ulcerated) just because treatment is now available.
8. She also highlighted her concern about the Birthmark Support Group where parents give other parents advice that would not be advised by professionals, or in the worst case, could cause greater harm.

Comments of the model:

1. Agreed that the shock parents of children with haemangiomas experience might be different to parents of children with a PWS as initially they have their "perfect" baby but within a few days it has changed and now has a growing birthmark. Perhaps they experienced

a greater sense of loss than those who are confronted with a PWS at birth.

2. Suggested greater recognition of coping strategies, perhaps evolving the resources category further.
3. Praised the idea of 'moving forwards' being captured and highlighted that in her experience it can be reassuring for parents and offering purpose.

## I. Exerts from my reflective journal

18<sup>th</sup> December

Feeling a little worried today as I still haven't had confirmation for my 5<sup>th</sup> interviewee yet. Hoping she agrees as I was excited to interview her as she has a child with the same type of birthmark as me. Haven't really been that moved by the interviews so far and I wonder whether it being the same type of birthmark would affect me differently. Otherwise, the introduction I have, on Helens, advice slimmed it down considerably and I now need to write the literature review section. It felt quite miserable to have to cut out so many words but I know it will be better in the long run. As far as transcripts go the first 3 are all ready to be coded and just 1 more to finish off transcribing so that feels like a little success!

29<sup>th</sup> December

So I started coding today, I re read by transcripts so far which sparked off lots of ideas which I've memo'ed. Coding feels a little alien, on the one hand, from reading the book and looking at her examples, it looks straight forward, but I'm finding the uncertainty of the subjectivity of it a little disconcerting. Otherwise, the concept is easy to grasp and the insights feel relevant and useful, so it feels like a bit of cognitive dissonance, easy but hard.

2<sup>nd</sup> January

Happy New Year! I can't believe it is officially the 6 month count down. The end is feeling uncomfortably close and I am noticing I am feeling the need to hurry up with things. I am meeting with Helen on Thursday and need to share this with her, I think I might need her help to not panic over time. It will be good to chat things over and get a bit of a timeline and a few self-imposed deadlines agreed. Also to make sure my coding is ok so I feel confident to continue with it.

13<sup>th</sup> January

I have been doing lot of coding and it has made me reflect on my own journey of having a birthmark. I have noticed that the data has not be resonating with me as much as I imagined it would and I having been thinking about why that is. I wonder if, following on from my surgeries a few years ago I physically no longer have the birthmark as it has been removed and although I am left with scars, they are not a birthmark and are not the same shape. This thought process has led me to the realisation that perhaps I know longer identify as someone with a birthmark, instead I identify as having a scars and a visible difference. Also, seeing people at the beginning of their journeys with

birthmarks has allowed me to reflect on what I have been through and to feel proud of all that I have accomplished in spite of the challenges growing up with a birthmark caused me. It also makes me thankful for the support I've had throughout my journey.

15<sup>th</sup> January

Meeting with Helen went well we did some coding together which was so helpful and she has sent me some examples of things to look at so that should help. She reminded me to keep hold of the essence of the transcripts and not to lose the emotion, otherwise the model could be quite generic. This was helpful and I have noticed myself today digging a bit deeper into the transcripts and working to capture the meaning behind in more emotive language. She also encouraged me to use more NVIVO codes which has been helpful. I'm feeling less panicked by time as well so finding it easier to slow down.

31<sup>st</sup> January

I have don't my final two interviews this weekend which feels like a mini triumph, I now have all of my data and I can carry on working on transcribing and coding. My last interview felt like it was the thinnest of all of them as the dad I was interviewing just didn't seem able to connect to the emotion of it which was a bit disappointing. I will be keen to see what it is like when I listen to it back when I transcribe it.

12<sup>th</sup> February

I was supposed to have meeting with Helen but she cancelled which was a bit frustrating. Hopefully she'll get my intro back to be soon so I can crack on with that. In the meantime, it's not like I don't have plenty of transcribing to be getting on with.

19<sup>th</sup> February

I have had my intro back from Helen and generally feel pleased with it and her comments, I have some changes to make but her comments all feel relevant and helpful so I will have to spend some time on that.

20<sup>th</sup> February

As expected, the interview with that dad was the hardest one to transcribe, I found myself getting annoyed at him being so superficial and even more so as

I could hear myself working hard in the interview to help him connect with it more but he was not willing to. It was quite a short interview and yet it has taken me a lot more sittings and I'm not even finished yet, probably because of how little I think I will get out of the interview, so it feels frustrating to be spending so much time on it.

29<sup>th</sup> February

I can't believe it's the end of Feb already and we had the milestone of end of lectures last week so now it really is that crunch time that has been looming for months. Time to really buckle down and crack on. Having said that, I am on track and where I wanted to be. I have my ethical extension through for interviewing the clinical nurse specialist and I am a fair way into my coding. But I have taken a few days annual leave so hopefully will make some serious progress this week. Coding is coming on well and I have 1 more interview to transcribe so all being well, my goals for this week are to have finished making changes to my intro, to have transcribed and coded all of my interviews and to have my coding table with the sentences making up the focussed code written in and to begin to conceptualise my model.

8<sup>th</sup> March

Finally got around to finishing the transcription on the last interview, by far the hardest to do which feels weird as it was the last one and the shortest. Think it was more to do the content, he really didn't engage well and I found it frustrating listening to how hard I was working and how little he was giving back. Feels strange that he seemed to lack empathy for the project and for his son. He is a doctor so perhaps he found it harder to connect with the emotion and is more matter of fact and more medically minded about it all. Also, the older son was in the room and this may have affected his responses. Glad it's done though and I can move on to finishing the analysis which is going well so far, model needs honing but the general ideas and concepts are all in my mind, just trying to put it down on paper in a way that makes sense.

Also pleased as ethics for the interview with the nurse are through, sadly she's on holiday so need to wait a week or so.