Parents’ experiences of caring for a child with a cleft lip and/or palate: a review of the literature

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Abstract

This review brings together for the first time the existing quantitative and qualitative research evidence about the experiences of parents caring for a child with a cleft. It summarizes salient themes on the emotional, social and service-related experiences of parents and critiques the literature to date, comparing it with wider, selected literature from the field of children’s long-term conditions, including disability. The review suggests that there are similarities and differences between the literatures, in terms of research focus and approach. Similarities are found across children’s conditions in the perspectives of parents on emotional, social and service-related aspects, although much of the cleft literature is focused on the early stages of children’s lives. However, the quality of cleft research to date about parents’ experiences has also been variable, with a narrow emphasis on cross-sectional, deficit-orientated psychological approaches focused mainly on mothers. Despite a substantial literature, little qualitative research has examined parents’ perspectives in-depth, particularly about their child’s treatment journey. This contrasts with the wider children’s literature, which has traditionally drawn not only on psychological approaches but also on the broader perspectives of sociology, social policy, nursing and health services research, using both qualitative and quantitative methods, often in integrated ways. Such approaches have been able to highlight a greater range of experiences from both mothers and fathers, about caring for a child with a long-term condition and views about treatment. The review identifies a lack of comparable research in the cleft field to examine parents’ experiences and needs at different stages of their children’s lives. Above all, research is needed to investigate how both mothers and fathers might experience the long-term and complex treatment journey as children become older and to elicit their views about decision making for cleft treatments, particularly elective surgeries.

Introduction

Clefts of the lip and/or palate1 are among the most commonly occurring congenital impairments (Mossey & Little 2002) and may affect a child’s ability to feed, chew, breathe and hear, as well as cause disturbance to dental, facial, speech and language development (Clinical Standards Advisory Group 1998). Other possible consequences include effects on emotional and social well-being, behavioural and learning difficulties (Hunt et al. 2005). In the UK, a long-term programme of surgery, orthodontic treatment and speech therapy is offered by multi-disciplinary teams of specialist clinicians, to address the functional and appearance-related consequences of having a cleft.

1 In this paper, the term ‘cleft’ refers collectively to clefts of the lip, clefts of the palate or clefts of both the lip and the palate.
Biomedical and psychological perspectives have predominated in cleft research to date (Rumsey & Harcourt 2005) and have focused mainly on the identification of risk factors and measurement of deficits (Eisermann 2001). Some research has also investigated people's views of service-related issues, although qualitative approaches to gauging in-depth perceptions have so far been relatively rare (Nelson 2009). This paper will present a narrative overview of the literature that examines the experiences of parents caring for a child with a cleft, making comparisons with wider, selected literature from the field of children’s long-term conditions and disability. It will draw out key issues to highlight similarities and differences, as well as gaps in knowledge.

**Literature search and appraisal**

The literature was searched comprehensively to identify publications that have examined the experiences of parents of children with clefts and more selectively for articles about parents and long-term conditions including disability. The search strategy is presented in Table 1, while Table 2 presents a summary of the 57 publications found.

**Table 1. Literature search strategy**

### Cleft literature search

**Databases searched:**
- British Nursing Index
- CINAHL Plus
- EMBASE
- Health and Psychosocial Instruments
- Maternity and Infant Care
- MEDLINE
- PsychINFO
- Social Sciences Index
- Sociological Abstracts
- ISI Web of Knowledge

**Search strategy** (combination of free text terms in title or abstract and Subject Headings):
- **Free text terms:**
  - (parent* or mother* or father* or family or families) OR (child* or infant)
  - OR (adolescen* or young people or young person* or teenage*)
  - OR (paediatric* or pediatric*) AND (cleft lip or cleft palate or cleft lip and palate or craniofacial or cranio-facial)

**Limitations:**
- from 1980 to present; human subjects; English language

**Critical appraisal:** All studies appraised for quality using criteria for the critique of both qualitative research (Popay et al. 1998; Seale et al. 2004) and quantitative research [Greenhalgh 2001; Critical Appraisal Skills Programme (CASP) 2007]

### Long-term conditions literature search

**Databases searched:**
- British Nursing Index
- CINAHL Plus
- EMBASE
- Health and Psychosocial Instruments
- Maternity and Infant Care
- MEDLINE
- PsychINFO
- Social Sciences Index
- Sociological Abstracts
- ISI Web of Knowledge

**Search strategy** (free text terms in title or abstract):
- **Free text terms:**
  - (parent* or mother* or father* or family or families) OR (child* or infant)
  - OR (adolescen* or young people or young person* or teenage*)
  - OR (paediatric* or pediatric*) AND (chronic disease or chronic illness) OR (long term condition or long-term condition) OR (disab*) OR (congenital)

**Limitations:**
- from 1990 to present (because of changes in policy/theoretical perspectives since 1990s); human subjects; English language

Emotional experiences of having a child with a cleft

**Early experiences and needs**

Discourses of 'loss', 'mourning' and 'correcting' have been common in research perspectives surrounding the diagnosis of a child with a cleft, informed by the assumptions of earlier theoretical perspectives (Olsansky 1962; Solnit & Stark 1962; Drotar et al. 1975). Both pre- and post-natally, across countries and cultures, parents' feelings of shock, anger, grief and worry have been documented both in surveys and in qualitative studies (Bradbury & Hewison 1994; Rey-Bellet & Hohlfeld 2011).
Table 2. Details of cleft lip and palate studies included in the review

<table>
<thead>
<tr>
<th>Author and year</th>
<th>Focus of the study</th>
<th>Methods</th>
<th>Sample size (parents)</th>
<th>Strengths and limitations</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Mixed-methods studies</strong></td>
<td></td>
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</tr>
<tr>
<td>Benson et al. 1991</td>
<td>Coping/adaptation (social support)</td>
<td>Questionnaires Semi-structured interviews</td>
<td>72</td>
<td>Matched comparison child group; validated measure (Social Support Questionnaire – Revised) Response rate/parent characteristics missing; fathers under-represented</td>
</tr>
<tr>
<td>Bradbury and Hewison 1994</td>
<td>Coping/adaptation</td>
<td>Semi-structured interviews Questionnaire</td>
<td>59</td>
<td>Recruitment details/sample characteristics missing; qualitative data collection/analysis not fully described 100% response rate; longitudinal</td>
</tr>
<tr>
<td>Cadogan et al. 2009</td>
<td>Information (diagnosis)</td>
<td>Questionnaire incorporating both closed and open questions</td>
<td>31</td>
<td>Small sample; qualitative analysis details missing</td>
</tr>
<tr>
<td>Cleft Lip and Palate Association 2007</td>
<td>Services (care provision)</td>
<td>Questionnaire with some open-ended questions</td>
<td>227</td>
<td>40% response rate Sampling strategy/characteristics missing and mainly from parent support group</td>
</tr>
<tr>
<td><strong>Qualitative studies</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cartwright and Magee 2006 Voluntary sector report</td>
<td>Information (needs)</td>
<td>Focus groups One-to-one interviews</td>
<td>3</td>
<td>In-depth focus; framework analysis Response rate missing; sample small and from support group only</td>
</tr>
<tr>
<td>Chucharoen et al. 2009</td>
<td>Information (needs)</td>
<td>Semi-structured interviews</td>
<td>15</td>
<td>Longitudinal; independent interviewer; content analysis Response rate/recruitment details/sampling strategy missing</td>
</tr>
<tr>
<td>Farrimond and Morris 2004 Unpublished undergraduate dissertation</td>
<td>Information (diagnosis)</td>
<td>Semi-structured interviews</td>
<td>10</td>
<td>In-depth/flexible approach; reflexive stance; interpretive Phenomenological analysis Response rate missing</td>
</tr>
<tr>
<td>Johansson and Ringsberg 2004</td>
<td>Coping/adaptation (social support)</td>
<td>Semi-structured interviews</td>
<td>30</td>
<td>In-depth focus; use of phenomenology Response rate missing; sample not diverse</td>
</tr>
<tr>
<td>Klein et al. 2006</td>
<td>Parenting</td>
<td>Semi-structured interviews</td>
<td>9</td>
<td>In-depth focusResponse rate missing; sample from parent support organization only</td>
</tr>
<tr>
<td>Martin 2005</td>
<td>Information (prenatal diagnosis)</td>
<td>Structured interviews</td>
<td>10</td>
<td>100% response rate; mothers and fathers Ages of children missing; little flexibility in study instrument; analysis techniques/reflexive stance missing</td>
</tr>
<tr>
<td>Author and year</td>
<td>Sample size (parents)</td>
<td>Focus of the study</td>
<td>Methods</td>
<td>Strengths and limitations</td>
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</tr>
<tr>
<td>Nusbaum et al. 2008</td>
<td>20</td>
<td>Information (diagnosis)</td>
<td>Semi-structured interviews</td>
<td>63% response rate; in-depth/flexible approach; includes comparable and contrasting findings; reflexive stance</td>
</tr>
<tr>
<td>Owens 2008</td>
<td>20</td>
<td>Services (feeding support)</td>
<td>Narrative interviews</td>
<td>Recruitment/sample details missing; few details of analysis techniques; no treatment of negative cases</td>
</tr>
<tr>
<td>Quantitative studies</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Andrews-Casal et al. 1998</td>
<td>61</td>
<td>Coping/adaptation</td>
<td>Questionnaires</td>
<td>52% response rate; some validated instruments (Parenting Stress Index, Hollingshead Index of Social Position)</td>
</tr>
<tr>
<td>Baker et al. 2009</td>
<td>103</td>
<td>Coping/adaptation</td>
<td>Questionnaires</td>
<td>Gender mix of sample unclear</td>
</tr>
<tr>
<td>Barden et al. 1989</td>
<td>10</td>
<td>Mother–infant interaction/attachment</td>
<td>Observation</td>
<td>Reasonable sample size (multi-centre); validated instruments (General Well-Being Scale, Brief COPE, Satisfaction with Appearance Questionnaire)</td>
</tr>
<tr>
<td>Berger and Dalton 2009</td>
<td>143</td>
<td>Coping/adaptation</td>
<td>Questionnaire</td>
<td>37% response rate; unrepresentative sample; fathers under-represented</td>
</tr>
<tr>
<td>Black et al. 2009</td>
<td>98</td>
<td>Adaptation</td>
<td>Questionnaire</td>
<td>Sample size reasonable and representative</td>
</tr>
<tr>
<td>Broder et al. 1992</td>
<td>495</td>
<td>Services (outcome of care)</td>
<td>Standardized interviews</td>
<td>Good sample size</td>
</tr>
<tr>
<td>Broder and Trier 1985</td>
<td>37</td>
<td>Information (diagnosis)</td>
<td>Questionnaire</td>
<td>46% response rate; sample small and characteristics missing</td>
</tr>
<tr>
<td>Byrnes et al. 2003</td>
<td>98</td>
<td>Information (diagnosis)</td>
<td>Questionnaire</td>
<td>Reasonable sample size</td>
</tr>
<tr>
<td>Campis et al. 1995</td>
<td>77</td>
<td>Coping/adaptation</td>
<td>Questionnaires</td>
<td>95% response rate; validated instruments (Child Behavior Checklist, Beck Depression Inventory, Spielberger Trait Anxiety Scale, Parenting Stress Index, Social Support Questionnaire – Revised)</td>
</tr>
<tr>
<td>Canady et al. 1997</td>
<td>96</td>
<td>Services (continuity of care)</td>
<td>Questionnaire</td>
<td>73% response rate; few sample or study instrument details</td>
</tr>
<tr>
<td>Cleft Lip and Palate Association 1996</td>
<td>102</td>
<td>Services (care provision)</td>
<td>Questionnaire</td>
<td>100% response rate</td>
</tr>
<tr>
<td>Davalbhakta and Hall 2000</td>
<td>90</td>
<td>Information (diagnosis)</td>
<td>Questionnaire</td>
<td>78% response rate; sample size reasonable</td>
</tr>
<tr>
<td>Endriga and Speltz 1997</td>
<td>116</td>
<td>Mother–infant interaction and attachment</td>
<td>Observation</td>
<td>Matched control group</td>
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</tbody>
</table>

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<table>
<thead>
<tr>
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</tr>
</thead>
<tbody>
<tr>
<td>Field and Vega-Lahr 1984</td>
<td>Mother–infant interaction and attachment</td>
<td>Observation</td>
<td>24</td>
<td>Recruitment/response rate missing; small sample; control group matched on some variables but not fully described</td>
</tr>
<tr>
<td>Jeffery and Boorman 2001</td>
<td>Services (care provision)</td>
<td>Questionnaire</td>
<td>341</td>
<td>72% response rate; good sample size; Sample characteristics missing</td>
</tr>
<tr>
<td>Kramer et al. 2007</td>
<td>Quality of life</td>
<td>Questionnaire</td>
<td>130</td>
<td>84% response rate; validated instrument (Impact on Family Scale); Gender mix of sample unclear</td>
</tr>
<tr>
<td>Krueckebegg and Kapp-Simon 1993</td>
<td>Coping adaptation</td>
<td>Questionnaires</td>
<td>52</td>
<td>Comparison group; validated instruments (Parenting Stress Index, Modification of the Block Child Rearing Practices Report, Social Relationship Scale, Four Factor Index of Social Status, Social Skills Questionnaire); Response rate missing; sample relatively small; gender mix of sample missing</td>
</tr>
<tr>
<td>Locker et al. 2002</td>
<td>Quality of life</td>
<td>Questionnaire</td>
<td>93</td>
<td>Validated scale (Family Impact Scale); Recruitment/response rate and sample characteristics missing</td>
</tr>
<tr>
<td>Murray et al. 2008</td>
<td>Mother–infant interaction and attachment</td>
<td>Observation</td>
<td>190</td>
<td>75% response rate; control group; home environment; some validated instruments (Behaviour Screening Questionnaire, Structured Clinical Interview for DSM Diagnoses)</td>
</tr>
<tr>
<td>Noar 1991</td>
<td>Services (outcome of care)</td>
<td>Questionnaire</td>
<td>30</td>
<td>Instrument piloted; Recruitment/response rate and sample details missing; small sample</td>
</tr>
<tr>
<td>Noor and Musa 2007</td>
<td>Services (care provision/outcome)</td>
<td>Standardized interview</td>
<td>60</td>
<td>Validated instrument (Cleft Evaluation Profile); Recruitment/response rate details missing; gender mix of sample unclear</td>
</tr>
<tr>
<td>Oliver and Jones 1997</td>
<td>Services (feeding support)</td>
<td>Questionnaire</td>
<td>100</td>
<td>64% response rate; Children's ages/sample characteristics/study instrument details missing</td>
</tr>
<tr>
<td>Pannbacker and Scheuerle 1993</td>
<td>Decision making</td>
<td>Questionnaire</td>
<td>42</td>
<td>56% response rate; Small sample; children's ages/sample characteristics/study instrument details missing</td>
</tr>
<tr>
<td>Pelchat et al. 1999</td>
<td>Coping/adaptation</td>
<td>Questionnaires</td>
<td>74</td>
<td>91% response rate; matched comparison groups; validated instruments (Stress Appraisal Measure, Parenting Stress Index, Emotional Distress Index-Quebec Health and Social Survey); longitudinal</td>
</tr>
<tr>
<td>Pope et al. 2005</td>
<td>Coping/adaptation</td>
<td>Questionnaires</td>
<td>47</td>
<td>Longitudinal, validated instruments (Parenting Stress Index-Short Form, Child Behavior Checklist); Response rate unclear; sample characteristics and gender mix unclear</td>
</tr>
<tr>
<td>Rey-Bellet and Hohlfeld 2004</td>
<td>Information/counselling</td>
<td>Questionnaire</td>
<td>29</td>
<td>82% response rate; Parents' socio-economic characteristics missing; analysis details missing; unclear how many fathers participated</td>
</tr>
<tr>
<td>Sank et al. 2003</td>
<td>Coping/adaptation (social support)</td>
<td>Questionnaires</td>
<td>145</td>
<td>98% response rate; validated instruments (Interpersonal Support Evaluation List, Beck Depression Inventory); No fathers</td>
</tr>
<tr>
<td>Semb et al. 2005</td>
<td>Services (care provision/outcome)</td>
<td>Questionnaire</td>
<td>81</td>
<td>65% response rate; questionnaire piloted; longitudinal Sample characteristics of parents missing</td>
</tr>
</tbody>
</table>
In-depth qualitative research, however, has been able to reveal a wider range of experience, often highlighting parents’ elation at a child’s birth, or perceptions of a child’s cleft as unremarkable or unique (Eisermann 2001; Farrimond & Morris 2004; Johansson & Ringsberg 2004; Klein et al. 2006).

Feelings of parental guilt, self-blame and associated anxiety have also been described (Strauss et al. 1995; Byrnes et al. 2003; Nelson et al. 2009) with recognition that parents may to wish share their feelings and get emotional support from experienced professionals at the time of diagnosis (Martin 1995; Strauss et al. 1995; Byrnes et al. 2003; Johansson & Ringsberg 2004; 2004; Martin 2005; Black et al. 2009; Cadogan et al. 2009; Chuchcharoen et al. 2009). In-depth qualitative research, however, has been able to reveal a wider range of experience, often highlighting parents’ elation at a child’s birth, or perceptions of a child’s cleft as unremarkable or unique (Eisermann 2001; Farrimond & Morris 2004; Johansson & Ringsberg 2004; Klein et al. 2006).

### Table 2. Continued

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<tr>
<th>Author and year</th>
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<th>Methods</th>
<th>Sample size (parents)</th>
<th>Strengths and limitations</th>
</tr>
</thead>
<tbody>
<tr>
<td>Speltz et al. 1990</td>
<td>Coping/adaptation USA</td>
<td>Questionnaires Observation</td>
<td>55</td>
<td>Matched control group; validated instruments (Parenting Stress Index, General Well-Being Scale, Social Health Battery, Locke-Wallace Marital Adjustment Scale) Response rate missing; small sample</td>
</tr>
<tr>
<td>Speltz et al. 1993</td>
<td>Coping/adaptation USA</td>
<td>Questionnaires</td>
<td>33</td>
<td>Control group; validated instruments (Dyadic Parent-Child Interaction Coding System, Nursing Child Assessment Teaching Scale, General Well-Being Schedule, Social Network Reciprocity and Dimensionality Assessment Tool, Dyadic Adjustment Scale, Child Behavior Checklist); longitudinal</td>
</tr>
<tr>
<td>Speltz et al. 1994</td>
<td>Mother–infant interaction and attachment USA</td>
<td>Questionnaires Observation</td>
<td>51</td>
<td>100% response rate; control group Clinic conditions; cross-sectional</td>
</tr>
<tr>
<td>Speltz et al. 1997</td>
<td>Mother–infant interaction and attachment USA</td>
<td>Questionnaires</td>
<td>115</td>
<td>Control group; validated instruments (Mental Health Index, Parenting Stress Index, Dyadic Adjustment Scale, Family Environment Scale, Questionnaire on Social Support); longitudinal design Response rate missing; laboratory-based</td>
</tr>
<tr>
<td>Strauss et al. 1988</td>
<td>Services (outcome of care) USA</td>
<td>Standardized interviews</td>
<td>102</td>
<td>Study instrument piloted Recruitment/response rate and parent sample characteristics missing</td>
</tr>
<tr>
<td>Strauss et al. 1995</td>
<td>Information (diagnosis) USA</td>
<td>Questionnaire</td>
<td>100</td>
<td>76% response rate; reasonable sample size; survey instrument piloted Fathers under-represented</td>
</tr>
<tr>
<td>Strauss et al. 2007</td>
<td>Experiences of stigmatization USA</td>
<td>Questionnaire</td>
<td>153</td>
<td>Reasonable sample size; response rate missing; convenience sample; fathers excluded</td>
</tr>
<tr>
<td>Thomas et al. 1997</td>
<td>Services (outcome of care) UK</td>
<td>Questionnaire</td>
<td>62</td>
<td>70% response rate Parent sample characteristics missing</td>
</tr>
<tr>
<td>Turner et al. 1997</td>
<td>Services (care provision/ outcome) UK</td>
<td>Standardized interview</td>
<td>130</td>
<td>76% response rate Recruitment/parent sample details missing</td>
</tr>
<tr>
<td>Turner and Milward 1988</td>
<td>Services (care provision) UK</td>
<td>Structured interviews</td>
<td>60</td>
<td>Recruitment/response rate/details of study instrument missing; gender mix of sample unclear</td>
</tr>
<tr>
<td>Williams et al. 2001</td>
<td>Services (care provision/ outcome) UK</td>
<td>Questionnaire</td>
<td>438</td>
<td>76% response rate; robust sample size; multi-centre Parent sample characteristics missing</td>
</tr>
<tr>
<td>Young et al. 2001</td>
<td>Information (diagnosis) USA</td>
<td>Questionnaire</td>
<td>40</td>
<td>100% response rate Sample unrepresentative; fathers under-represented</td>
</tr>
</tbody>
</table>
Rey-Bellet & Hohlfeld 2004). The quality of both quantitative and qualitative studies in this body of research is variable, however, with half omitting to provide response rates and a quarter to document children’s ages. Most exclude fathers and focus on the collection of cross-sectional data alone. In addition, the quantitative studies comprise samples as small as 25 and use few validated measures, while little of the qualitative research fully presents its approach or findings.

Studies of the early developing relationship between parents and children with clefts have been strongly informed by attachment theory (Bowlby 1965) and suggest that infants may be as securely attached to their mothers as those without clefts (Speltz et al. 1990, 1993, 1997; Endriga & Speltz 1997; Slade et al. 1999), with a potentially less sensitive interplay between the two because of possible disturbances in communication cues (Field & Vega-Lahr 1984; Barden et al. 1989; Speltz et al. 1990, 1994; Endriga & Speltz 1997; Murray et al. 2008). Although the majority have used validated instruments, their findings are based on relatively small and diverse samples, comprised of between $n = 10$ and $n = 55$. Only four took a longitudinal approach to track developments over time, but their groups were inconsistently matched, sometimes including children with associated developmental difficulties as well as a cleft (Speltz et al. 1993, 1997; Murray et al. 2008). Most notably, only one study (Murray et al. 2008), which was also the most robust in terms of sample size at $n = 190$, observed mothers and infants in their own homes, the remainder having been conducted in artificial laboratory conditions. Attachment theory has been widely criticized outside the field on the grounds that it takes no account of social or environmental context on the developing parent–child relationship (Knestrick 2002) and reinforces stereotypical maternal roles, attributing responsibility for a child’s emotional and social difficulties to women alone (Contratto 2002).

Although feeding difficulties have been reported among parents of infants with clefts in one UK survey ($n = 100$; Oliver & Jones 1997), research to explore parents’ emotional experiences of feeding their child has been relatively rare. Only one qualitative study has offered some insight into the emotional impact on mothers of pressure to successfully breastfeed (Owens 2008), but it provides little information on its sampling and recruitment strategy or details of whether any cases might have contrasted with the analysis presented.

**Emotional ‘strain’**

Several psychological studies based in the USA have focused on the assessment of stress levels among parents caring for children with clefts, investigating outcomes such as anxiety, depression and poor psychological ‘adjustment’, collectively referred to here as emotional ‘strain’. Some studies have found evidence of raised levels of emotional strain among parents in their child’s toddlerhood (Speltz et al. 1990, 1993; Pope et al. 2005), but findings generally suggest that by pre-school years, levels are equivalent to those in parents of children without clefts (Krueckeberg & Kapp-Simon 1993; Campis et al. 1995; Andrews-Casal et al. 1998; Pelchat et al. 1999; Slade et al. 1999; Berger & Dalton 2009). Most of these studies have used relatively small sample sizes (average $n = 58$) along with diverse research instruments, making inferences difficult to draw. Further, accounts from parents in their own words are rare and the focus of research has primarily been on the negative experiences associated with caring for a child with a cleft, to the relative exclusion of potentially positive aspects (Eisermann 2001; Baker et al. 2009).

Some studies have suggested that a child’s cleft treatment and clinic attendances may impact adversely on families’ quality of life (Locker et al. 2002; Baker et al. 2009), and outside the UK a small number of in-depth qualitative studies have highlighted parents’ feelings of anxiety about surgery and their perceived need for emotional support through treatment for both themselves and their child (Turner et al. 1997; Eisermann 2001; Johansson & Ringsberg 2004; Klein et al. 2006). However, as far as is known, no previous studies have aimed to explore the emotional experiences of parents in connection with their child’s treatment pathway.

**Coping**

Research in the field on parental coping is subsumed in the studies outlined in the preceding section; however, it is important to highlight that the focus of this work has been mainly on the cross-sectional assessment of poor ‘adjustment’ in deficit-oriented ways (Baker et al. 2009) with no longitudinal work to track parental coping as a child becomes older. Contextual factors have largely been excluded in this body of work too, with studies taking little account of issues other than a child’s cleft that may be affecting families’ lives (Krueckeberg & Kapp-Simon 1993). The incorporation of ‘resilience’ models in the investigation of coping in cleft research has been highlighted as a desirable goal (Broder 2001; Kapp-Simon & Gaither 2009), yet little published research of this kind exists to date. However, influenced by models of resilience and positive coping, a recent British study ($n = 103$), using validated study instruments including the Coping Response Inventory and the Stress Related Growth Scale, investigated coping strategies and
perceptions of positive outcomes among parents (Baker et al. 2009). The study found a high degree of positive coping and outlook, particularly in parents of children with more extensive clefts.

Studies using mainly qualitative methods have also offered insights into the positive and/or rewarding aspects of caring for a child with a cleft that have been identified by parents themselves. Such rewards include recognition of their own personal strengths, stronger relationships, appreciation of diversity and others’ good intentions, tolerance, a sense of community and hope for the future (Bradbury & Hewison 1994; Eisermann 2001; Klein et al. 2006). Parents have also reported recognizing their child’s strengths, such as a determined attitude, perseverance and sociability (Klein et al. 2006). In one study, half of parents reported that they would not remove their child’s cleft if given the opportunity (Eisermann 2001).

In terms of coping resources, it has been suggested that because of the stigma potentially associated with having a visible facial difference, parents of children with clefts may lack adequate social support (Benson et al. 1991; Speltz et al. 1993; Pelchat et al. 1999; Sank et al. 2003). Some studies have found low levels of social support among such parents (Benson et al. 1991; Speltz et al. 1993; Campis et al. 1995; Sank et al. 2003), while others find no particular differences between parents of children with and without clefts (Krueckeberg & Kapp-Simon 1993). Yet others find variation across families, regardless of their child having a cleft (Bradbury & Hewison 1994; Johansson & Ringsberg 2004; Baker et al. 2009). However, these studies have not clearly delineated different types of social support and how it might be experienced by parents, distinguishing, for example, the relative impact of formal and informal support.

Investigation of the coping strategies used by parents does not appear to have been a priority in cleft research to date, although a small amount of qualitative work has indirectly revealed some of the strategies which parents may use. These include cognitive or problem-focused strategies such as discussing a child’s needs with school staff and thinking of ways to help them establish friendships/handle teasing (Klein et al. 2006). Parents have also reported using emotion-focused strategies such as maintaining a hopeful attitude for the future and a belief in their own competence as parents (Johansson & Ringsberg 2004; Klein et al. 2006).

**Social experiences related to having a child with a cleft**

A smaller body of research has provided insights into parents’ social experiences. It is known that children and their families may be ‘stigmatized’ because of a cleft’s effects on appearance and/or function (Goffman 1963; Partridge 1997) and their social experiences characterized by discomfort, anxiety or rejection because of perceived ‘differences’ (Rumsey & Harcourt 2005). Research which has been mainly qualitative in nature has revealed the challenges that parents may face in managing others’ reactions to their child’s cleft. Mothers have described their heightened sensitivity to the reactions of friends and family (Farrimond & Morris 2004; Johansson & Ringsberg 2004), the verbal and non-verbal expressions of distaste encountered in public and attempts to conceal their baby or shun social situations in order to avoid feeling ‘different’ (Bradbury & Hewison 1994; Johansson & Ringsberg 2004; Klein et al. 2006).

Parents have also reported in qualitative studies their worries about social issues, including concerns about a child’s acceptance by peers, experiences of teasing, finding a life partner and securing employment (Cartwright & Magee 2006; Klein et al. 2006). Klein and colleagues’ in-depth study recorded the distress caused to parents by their child’s reported experiences of teasing or bullying at school, or when entering new and unfamiliar settings. This study also uncovered the tension parents experienced in efforts to protect children, while promoting their independence. One survey from the USA (n = 153), reported perceived stigmatizing experiences to be prevalent among both mothers and their children (Strauss et al. 2007). In a number of others across countries, between 15% and 68% of parents reported feeling their child’s self-confidence to have been affected by having a cleft and between 50% and 68% felt their child had been teased (Noar 1991; Turner et al. 1997; Semb et al. 2005; Noor & Musa 2007). Overall, this research has been more balanced in terms of its methods, with quantitative surveys and qualitative studies more equally employed, although fathers’ perspectives remain rare.

**Experiences of cleft services**

**Information and decision making**

The majority of cleft studies have focused on parents’ information experiences and needs at the point of diagnosis, with fewer studies addressing these issues across children’s age ranges. However, a lack of readily accessible information at diagnosis has been reported (Martin 1995; Cleft Lip and Palate Association 1996; Young et al. 2001), as has parents’ need for accurate and balanced information about clefts and their causes (Davalbhakta & Hall 2000; Young et al. 2001; Nusbaum et al. 2008). Studies have also suggested that most parents prefer to receive
their information at this time verbally, from specialist practitioners (Strauss et al. 1995; Byrnes et al. 2003).

While the involvement of parents and children in decisions about their care has been encouraged in UK health policy, research about parents’ experiences of decision making for their child’s cleft treatment is sparse. One small survey from the USA (n = 42) found that over a third of parents wanted to be more involved in decision making about treatment (Pannbacker & Scheuerle 1993), and surveys in the UK have also suggested that some parents do not feel involved in decisions and would like more involvement (Turner et al. 1997; Jeffery & Boorman 2001).

Experiences of service organization, delivery and outcomes

‘Satisfaction’ with the organization and delivery of cleft services has been investigated in a number of surveys whose results suggest high ratings among parents both in the UK and elsewhere (Turner et al. 1997; Jeffery & Boorman 2001; Williams et al. 2001; Semb et al. 2005; Cleft Lip and Palate Association 2007; Kramer et al. 2007; Noor & Musa 2007). Satisfaction has often been treated rather simplistically in this research, however, as most papers fail to properly define how it is being conceptualised. Some refer to satisfaction with ‘care and attention’ (Turner et al. 1997; Williams et al. 2001; Noor & Musa 2007), some to the ‘level’ of care (Jeffery & Boorman 2001) and others to the ‘manner’ of care provided (Semb et al. 2005). Despite the positive ratings of parents reported, areas of concern have also been identified, such as poor access to and co-ordination of services (Martin 1995; Cleft Lip and Palate Association 1996, 2007; Oliver & Jones 1997). However, these surveys comprise widely differing sample sizes (between n = 30 and n = 495) and are almost exclusively cross-sectional and apart from the largest study (Williams et al. 2001), from single centres. Further, study instruments in this body of research are diverse and validated measures largely absent.

Research suggests that parents have confidence in cleft clinicians because of their specialism (Johansson & Ringsberg 2004; Semb et al. 2005; Cleft Lip and Palate Association 2007) and that they have concerns about the lack of knowledge among generic healthcare staff (Turner & Milward 1988; Cleft Lip and Palate Association 1996; Oliver & Jones 1997; Johansson & Ringsberg 2004; Martin 2005; Cartwright & Magee 2006). Parents have highlighted the importance of having practitioners who communicate well and show sensitivity (Broder & Trier 1985; Cleft Lip and Palate Association 1996, 2007; Jeffery & Boorman 2001; Byrnes et al. 2003; Semb et al. 2005; Cartwright & Magee 2006) and have identified continuity of care from specialist cleft clinicians as especially valuable (Cleft Lip and Palate Association 1996; Canady et al. 1997).

Some literature has also focused on parents’ satisfaction with the outcomes of their children’s cleft treatment, using the key variables of facial appearance, function and psychosocial well-being. These studies too have commonly reported high ratings of satisfaction with treatment (Strauss et al. 1988; Noar 1991; Broder et al. 1992; Thomas et al. 1997; Turner et al. 1997; Williams et al. 2001; Pelchat et al. 2004; Semb et al. 2005; Noor & Musa 2007; Berger & Dalton 2009). Some have also found broad agreement between parents and their children (Strauss et al. 1988; Semb et al. 2005; Noor & Musa 2007), while others have revealed dissimilar views on different variables (Noar 1991; Broder et al. 1992; Thomas et al. 1997; Williams et al. 2001; Berger & Dalton 2009). On outcomes for psychosocial well-being, findings have also been inconsistent, with some suggesting children with clefts to experience low levels of self-confidence and high levels of teasing (Semb et al. 2005; Noor & Musa 2007). Others report children to be less emotionally and socially affected by their cleft than their parents estimate (Noar 1991), or to experience more social, but fewer emotional effects (Turner et al. 1997). Differences in these findings may be due to the diverse samples and study instruments used, a lack of longitudinal data or multidimensional approaches to researching satisfaction with treatment results in a particularly sensitive context, or differing personal/cultural expectations about the outcomes of care.

Discussion

This literature review has contributed knowledge about the emotional, social and service-related experiences of parents caring for a child with a cleft, but has highlighted the variable quality of research to date. It has demonstrated that quantitative research in the cleft field has comprised mainly cross-sectional surveys, with relatively small sample sizes focused mainly on mothers. Little qualitative research, either standing alone or as part of mixed-methods designs, has been carried out to investigate parents’ perceptions and experiences and varies widely in the extent to which it takes an in-depth approach. Similarities as well as differences are found in the wider literature on children’s long-term conditions.

Comparison and contrast with the wider literature

Parents may encounter particular emotional and social challenges because of a cleft’s visible and/or audible effects on
their child’s facial appearance and speech – both of central importance to interpersonal relationships and communication (Rumsey & Harcourt 2005). None the less, similarities have been found among the parents of children with other long-term conditions in relation to their emotional, social and service-related experiences. Recognition of the potential emotional impact of a child’s diagnosis and the need for adequate early emotional and social support is comparable (Grootenhuis & Last 1997; Sloper 1999; Case 2000; Piggot et al. 2002; Trulsson & Klingberg 2003; Barr & McConkey 2007). Parental experiences of social stigma are also reflected in the long-term conditions literature, as is the tension parents may experience between protecting children while fostering their independence (Ray 2003; Rehm & Bradley 2005; Duguid et al. 2007; Lassetter et al. 2007). Also found are parents’ concerns about accessible, well-coordinated services for children (Mitchell & Sloper 2001; Watson et al. 2002; Law et al. 2003; Beecham et al. 2007) and needs for information (Mitchell & Sloper 2002; Lawoko 2007). A need for knowledgeable, sensitive practitioners with good communication skills is also reflected here (Davies et al. 2003; Farrant & Watson 2004; Hallstrom & Elander 2007), as is the value placed on long-term relationships between families and practitioners (Trulsson & Klingberg 2003; Lalor et al. 2007). Finally, in common with the cleft literature, high levels of parent satisfaction with treatment outcome are reported in studies of children’s surgery (Bridwell et al. 1999; Smith et al. 2006) with similar inconsistencies of opinion between parents and children (Bridwell et al. 1999; Pratt et al. 2002; Rinella et al. 2004; Smith et al. 2006).

Marked differences can be found in the children’s long-term conditions literature, however, in terms of both research approaches and focus, particularly in relation to the emotional and service-related aspects of parents’ experiences. This literature draws on broader perspectives from sociology, social policy, nursing and health services research, more commonly recognizing the value of both quantitative and qualitative methods to study parents’ experiences, often in integrated ways (Lassetter et al. 2007). Observation of naturally occurring interactions between parents and their children in everyday life and interviews to explore both mothers’ and fathers’ perceptions of the evolving nature of relationships over time are found (Anderson 1981; Lauritzen 1997; Kirk et al. 2005; Santos & McCollum 2007). The importance of the role of expectations and the need for multiple approaches to investigating parents’ satisfaction with services, including in-depth qualitative studies, has also been emphasized here (Callery & Luker 1996; Smith et al. 2006; Green et al. 2008).

The variation in parents’ reactions has been more widely acknowledged outside the cleft field in relation to diagnosis (Quine & Pahl 1987; Lane Tanner et al. 1998; Case 2000; Vehkakoski 2007; Bainbridge 2009), and the ambiguity of parents’ feelings has been more evenly described (Larson 1998; Kearney & Griffen 2001; Nelson 2002; Carnevale et al. 2006). The ability of parents to cope and adapt is highlighted in the wider literature, informed by a change in perspective in the 1990s from pathologizing approaches to ‘ecological’ and ‘resilience’ models (McCubbin & McCubbin 1993; Beresford 1994; Wallander & Varni 1995; Sloper 1999; Rolland & Walsh 2006). Conceptual frameworks have documented that parents caring for disabled children through treatment may use a wide range of coping strategies, both problem- and emotion-focused (Burr & Klein 1994; Graungaard & Skov 2006), and the rewards of caring have also been more widely recognized, with evidence of parents’ personal satisfaction and growth (Gregory 1994; Kearney & Griffen 2001; Nelson 2002; Barnett et al. 2006; King et al. 2006; Lassetter et al. 2007). Although rarely seen in the cleft literature, the emotional impact on parents of children’s surgery has been a focus of research in the wider literature (Sobo 2005; Amin et al. 2006; Ben-Amitay et al. 2006; Joseph et al. 2007; MacLaren & Cain 2008), and parents’ views of their children’s treatment outcomes as often uncertain have also been brought to light (Lane Tanner et al. 1998; Kearney & Griffen 2001; Brinchmann et al. 2002; Vehkakoski 2007).

In contrast to cleft research, the wider literature draws heavily on the notion of flexible, holistic services to support families in different ways according to their needs as they move through treatment (McConachie 1994; King et al. 1997; Mitchell & Sloper 2001; Watson et al. 2002). Theories of ‘respectful’ or ‘family-centred’ care have long underpinned research on quality in family services in the wider children’s literature (Mittler 1994; Mitchell & Sloper 2001; Trulsson & Klingberg 2003) but are relatively rare in the cleft literature so far. Additionally, theoretical perspectives about family involvement in decision making for treatment comprising ideas of ‘partnership’ between parents and practitioners (Coyne 1997; King et al. 1997; Piggot et al. 2002; Coyne & Cowley 2007) and family ‘empowerment’ (Mittler 1994; Mitchell & Sloper 2001) are central to the wider literature, but absent in cleft research. A substantial body of work in the wider children’s literature has also investigated the experiences and preferences of parents about involvement in decision making about their child’s care, using a range of research methods (Ellis & Leventhal 1993; Pyke-Grimm et al. 1999; Brinchmann et al. 2002; Hallstrom et al. 2002; Hallstrom & Elander 2004; Knopf et al. 2008). This research suggests that preferences cannot be predicted on the basis of demographics alone, but depend on
the context and nature of particular decisions (Knopf et al. 2008).

**Conclusions**

This review is the first to bring together evidence from both quantitative and qualitative research about the experiences of parents caring for a child with a cleft and demonstrates the variable quality of research to date. It highlights that despite a substantial literature, little research has so far examined parents’ perspectives in-depth, with a narrow emphasis on cross-sectional, deficit-orientated psychological approaches focused mainly on mothers. Several gaps have been identified, including the lack of research to examine parents’ experiences and needs at different stages of their children’s lives, as they move through cleft treatment. Above all, research is needed to investigate how both mothers and fathers might experience their child’s long-term and complex treatment journey as children become older and to elicit their views about decision making for cleft treatments, particularly elective surgeries.

**Key messages**

- This review synthesizes evidence from both quantitative and qualitative research about the experiences of parents caring for a child with a cleft.
- The quality of research on this topic to date has been variable.
- There has been a narrow emphasis on cross-sectional, deficit-orientated psychological approaches focused mainly on mothers.
- Despite a substantial literature, little qualitative research has examined parents’ perspectives in-depth across children’s ages.
- There has been a lack of research to examine in particular the experiences of mothers and fathers along their child’s long-term and complex treatment journey including their views about decision making for cleft treatments.

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**References**


