

This report will be submitted for possible publication in a journal. We are not therefore able to publish it fully here at the present time but we list the chapter headings, provide an overview and give some conclusions. For further information on the report contact the project coordinator.

SCR4Cleft Theoretical Report

A report based upon a literature review identifying the key issues faced by children with a cleft, summarising the difficulties of access to specialist care and presenting evidence showing the effectiveness of providing support to parents, empowering them to provide certain elements of care at home. A particular strength of this report is the synthesis of evidence relating to speech, language and psychological adjustment.

1) Overview

- I. Europe: The Context*
- II. Incidence of Cleft Lip and Palate in Europe*
- III. Brief summary of the challenges faced by children and young people affected by cleft*

2) Communication Skills & Key Challenges Associated with Cleft

- I. An Introduction to prelinguistic skills, language and speech*
- II. Key Challenges Associated with Cleft:*
 - prelinguistic skills, language and speech*
 - aspects of social communication skills*
 - the family environment*
 - the wider environment: social & educational processes*

3) Key Goals of Cleft Care: Developing Effective Communication Skills & Psychosocial Resilience

- I. The need for speech & language therapy*
- II. Achieving psychosocial resilience*

4) Current provision of specialist support for patients and families

- I. Disparities in care & the access to specialists (in particular, speech and language therapists & psychologists)*
- II. The impact of financial and social inequalities on access to care*

5) The Potential to Improve Outcomes in Speech & Language and Psychology

- I. The key role of parents in the development of speech and language, communication skills and psychosocial development*
- II. The potential to enhance outcomes through training for specialist & non-specialist healthcare professionals and parents*

6) Summary & Conclusions

Overview

Clefts of the lip and or palate come in all shapes and sizes and may be accompanied by associated conditions/syndromes. Affecting appearance and key areas of functioning (including feeding, speech and language and in some, hearing and ENT issues, physical and cognitive development), the challenges of growing up and living with a cleft can be considerable for those affected, their parents and other family members. These challenges include threats to psychological/emotional wellbeing (mood; mental health) self-perceptions (body-image; self-construct; self-esteem), social experiences and relationships (communication skills; first impressions, friendships and intimate relationships), educational and occupational performance. Treatment from birth into adulthood also brings with it a considerable burden of care, including appointments with health care professionals from several disciplines and also regular checks involving a multidisciplinary team. Nelson et al's (2012) study of parents and patients highlighted the stress attributed to time away from school, from the disruption to normal family routines, and for many, the considerable expense of attending appointments. Parents also reported concern regarding their child's readiness to begin school as well as their frustration and anxiety regarding poor access to speech and language therapy and the lack of consistency in the staff available to offer support (Sweeney et al in press).

Current understanding of the extent of these challenges for patients and their parents has been summarised elsewhere (Rumsey & Stock, 2013; Crerand, 2020). It is now understood that in those affected, psychological adjustment to cleft is multifaceted and that there is considerable variability in individual levels of wellbeing and distress (Stock et al, 2018). While in comparisons with the broader population, some studies have reported poorer long-term outcomes for those affected by cleft and their families in the domains of social engagement, educational and vocational attainment, and physical and mental health, other research has reported few differences compared to contemporaries without cleft, and/or more favourable scores on a range of psychological outcomes (e.g. Berger and Dalton, 2009; Berger and Dalton, 2011d). In the light of this, there has been a shift away from a focus purely on the problems and difficulties associated with cleft towards a growing emphasis on efforts to identify factors and processes involved in psychological resilience in the face of these challenges (Rumsey & Harcourt, 2012; Stock et al 2018; Ridley 2020). In a synthesis of qualitative studies, Stock and Feragen (2016) reported a range of challenges associated with growing up with a cleft, but also highlighted findings of stronger relationships, positive growth heightened empathy for others and an increased appreciation of diversity. The variability of the results reported in the literature is testament to the elaborate interplay of physical, cultural, psychological and social factors contributing to adjustment, also highlighting that in any one person, wellbeing will fluctuate over time, experiences and contexts (Stock et al 2018).

Progress in fully understanding the key factors in psychological adjustment has been hampered by a range of methodological challenges, including lack of agreement on key constructs, the large variety of measures used in studies reported in the literature and a lack of longitudinal study designs. However, it is now well understood that the severity of a cleft is not a key predictor of adjustment (Feragen and Stock, 2018). Instead, psychological and social factors play a crucial role. Key elements of adjustment in adulthood, for example, include dispositional optimism (the degree to which a person has a positive or negative outlook on life), the degree to which a person's sense of self-worth is dominated by feelings about their appearance, levels of concern about the possibility of negative reactions from

others, effective communication skills and feelings of social acceptance (Clarke et al, 2013). In Ridley's research (2020), adults who had incorporated their experience of a cleft and its treatment into an overall life story in which the presence of the cleft is acknowledged, but not given 'centre-stage' were more positively adjusted than those for whom the cleft had become a prominent 'hook' on which to hang the bulk of their negative experiences. These and other findings have highlighted the need to include positive psychological adjustment as a key goal of cleft care and the imperative to engage in more research to fully understand the precursors of psychological adjustment as they emerge in childhood.

Additionally, as the majority of researchers in this field have to date been working within their own disciplinary silos, the vast majority of published studies have focused on piecemeal elements of the overall functioning and experience of people affected by cleft. Yet, as the growing number of qualitative studies in this field illustrate, everyday experience involves a broad sweep of psychological, social and functional aspects (Stock et al, 2018). Thus, although elements of the complex jigsaw of the person's total experience are available in the literature, more interdisciplinary co-working is needed to piece these elements together. No area illustrates this better than a key element of everyday functioning – communication. As a cleft can result in differences in the structure of the mouth and nose which persist beyond treatment, residual functional deficits in speech and language development (and for some, hearing) may also endure, whether or not intervention has been accessible. As the child grows older, additional challenges to effective communication may include reactions to their appearance and speech, their own temperament, self-perceptions and social skill (Rumsey & Harcourt, 2004; Stock et al 2020). Feragen et al (2017) have highlighted the need for an awareness that children with communication difficulties may be psychologically or socially vulnerable, but as yet, few interdisciplinary studies have been undertaken that clarify the components and inter-relationships of the contributory factors.

In summary, while acknowledging the need for more interdisciplinary research, there is agreement spanning disciplinary boundaries about two key challenges to effective functioning in everyday life for people negatively affected by cleft – firstly, developing the capacity for effective interpersonal communication, and secondly, the development of psychological resilience (Lyons and Roulstone, 2018; Crerand et al, 2020). This project focuses on these key challenges in the belief that a synthesis of knowledge and understanding from the disciplines of speech and language therapy and psychology has great potential to improve outcomes for people affected by cleft and their families.

Conclusion

Long-term outcomes for infants born with a cleft of the lip and/or palate are very variable and not well predicted by the eventual aesthetic or functional results of treatment. To reach the goal of positive adjustment and successfully navigate the challenges associated with living with a cleft, key developmental tasks for children and their families include the acquisition of effective communication skills and the development of psychological resilience. Parents are uniquely influential in achieving these goals. Ideally, they should be supported in their efforts by appropriate specialist care from their local cleft team, however, across Europe, access to specialist care is very variable.

With the aim of filling the gap left by these inequalities in care provision, this project focuses on the development of training materials designed to empower health care professionals to

impart the knowledge and skills to parents and caregivers necessary to optimise the likelihood of positive outcomes for their children. Acknowledging both the multifactorial nature of adjustment, and the prevailing tendency for researchers in this field to work in their own disciplinary silos, this project harnesses the strengths of an interdisciplinary approach. Synthesising existing and emerging findings from speech and language therapy and psychology, as well as expertise from other relevant fields, these training materials will promote the acquisition of effective communication skills and psychosocial resilience in children affected by cleft.