



SCR4Cleft Theoretical Report (Output 2)

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

References

Appendix



1) Overview

i) *Europe – The Context*

Size, demographics, economy

The size of Europe differs according to how you define it. In its broadest geographical sense, stretching from the Ural Mountains that border with Asia in the East, to the Atlantic Ocean in the West, it covers an area of more than 10 million square kilometres, comprising 48 states of which 28 belong to the European Union (EU). Europe has a population of over 730,000 000 of which 505,000 000 reside in the EU states. Over 200 languages are spoken in Europe, mostly comprising Romance, Germanic and Slavonic languages, reflecting huge cultural diversity. There are 24 official languages of the EU.

Europe's GDP is just under 20 trillion USD with huge variances of GDP across the continent, ranging from over 25,000 USD per capita in highly developed economies to less than 10,000 USD in the poorest economies. Germany has the highest national economy in Europe, ranking fourth in the world, followed by France and the UK, ranking fifth and sixth globally. Unemployment rates average at 10% but vary enormously, ranging from less than 6% to over 23% nationally and far higher regionally.

Source; Eurostat, United Nations Development Programme

Health

On average, life expectancy within the EU is 77.5 years for males and 83.1 years for females. However, there is a large variation in national rates with Latvia and Lithuania having the lowest life expectancy for males at around 68. (*Eurostat*) Overall, the population profile of the EU is ageing which is presenting very significant difficulties in future healthcare planning. Healthcare systems differ widely. The amount of public funding spent on health care (as a proportion of all healthcare funding) ranges from 96.9 % (UK) to 8.6% in Georgia with a median of 76.9 % (WHO).

ii) *Incidence of Cleft Lip and Palate in Europe*

Childhood and adolescence are periods of extensive physical, psychological and social development, many aspects of which can be impacted by a congenital condition. In Europe, around one in 700 babies is born with **cleft lip and/or palate**, the most common congenital anomaly of the head and neck region. The incidence is approximately 1.6 per 1,000 live births, but there is some variance across Europe. Estimates indicate there are over 900 000 individuals (babies, children and adults) with clefts in Europe - a significant figure, especially when one considers that not only the patients but also their families are affected in terms of psychosocial adjustment and having to endure the



Speech Communication, Resilience – supporting children and parents with clefts
burden of a long treatment pathway. Treatment involves multidisciplinary health care from birth until early adulthood and sometimes beyond.

In some cases, the cleft may be associated with other problems which need specialist management and these need to be identified early. Accurate diagnosis (antenatal or post natal), the provision of appropriate information and support for the family, and the establishment of a structured care pathway, especially in the early months, have the potential to ensure that these infants thrive and develop optimally.. Access to good treatment varies widely throughout Europe, meaning that many children born with clefts are never given the opportunity to realize their full potential. The concept of a comprehensive specialist-team approach to care is not universal. Furthermore, babies with clefts are still institutionalized in some countries in Europe, putting them at even greater risk of developmental issues. Speech and language difficulties are most likely to occur and where there is a cleft of the palate – this accounts for around 80 per cent of all clefts.

This report offers a synthesis of the current understanding of the key preschool and early school challenges faced by children affected by cleft and their primary caregivers, with a particular focus on the development of effective communication skills and psychological resilience. Children with clefts (and other craniofacial conditions) need help to deal with the challenges associated with their condition and its treatment and to develop into confident adults (Feragen et al 2019), however, there are considerable differences in the availability and access to specialist speech and language and psychological support services across partner countries/the EU. In view of this lack of support for many patients and their families, this report also offers an evaluation of the potential for parents to promote positive outcomes in their children and the role of health care professionals to facilitate parental involvement. This evaluation is based on a synthesis of research literature in the fields of speech and language therapy and psychology.

iii) Summary of the challenges faced by children and young people affected by cleft and their parents

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



Speech Communication, Resilience – supporting children and parents with clefts

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,



Speech Communication, Resilience – supporting children and parents with clefts

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.

2) Communication Skills: Key Challenges Associated with Cleft

i) An Introduction to prelinguistic skills, language and speech

Speech and language are the most complex cognitive functions in humans. The infant is pre-programmed or hard wired to make sense of and acquire language. Language is considered to be the symbolisation of thought and is the ability to take a finite set of elements (such as words), using a set of rules (grammar), and create infinite word combinations, which are creative and comprehensible. It is a learned code, or system of rules that enables us to communicate ideas and to express wants and needs. Reading, writing, gesturing and speaking are all forms of language (Bowen, 1998). For the purposes of this document, we are focusing on prelinguistic skills, verbal comprehension (receptive language) and oral language (expressive language) and speech, which are of most relevance to children born with cleft palate.

The *Prelinguistic stage* is a very important period in which although the parent/caregiver initially plays a central role as the interactive communication partner, very soon after birth the infant is an active partner too. This period, before the onset of oral language, is characterised by the infant establishing the foundations for its development. The infant learns to understand the role of the listener in conversation, develops attention and listening skills, eye contact, intentional communication, and lays down motor patterns which become associated with speech, including gesture and vocalization. Importantly, the child's motivation to communicate develops at this stage.



Speech Communication, Resilience – supporting children and parents with clefts

Receptive language involves the processing and making sense of what people say, through understanding spoken words and the rules of the grammar of the language. Receptive language difficulties occur when the listener does not understand the vocabulary or the meaning of sentences spoken by their communication partner. Comprehension evolves from the early understanding of words in context e.g. the infant understands ‘drink’ as the mother reaches for a cup, into much more sophisticated understanding, where the child is able to make deductions about the world from the language heard.

Expressive language is made up of words (vocabulary). There are rules regarding the structure of words known as morphology e.g. ‘s’ at the end of nouns in English often implies plural, or ‘ed’ at the end of a verb puts the verb into the past tense. Words are combined to form phrases, clauses and sentences according to the grammatical rules or syntax of the language. Sequences of sentences are used to make up conversations and narratives. Expressive language also includes semantics referring to the intended meaning and pragmatics is the use of language in context. This includes the skills necessary to interact effectively. It includes the use of language for different purposes (e.g. to question, clarify, describe or debate) and the knowledge of the non-verbal rules of communication. This involves listening, eye contact, knowing how to take verbal turns and how to change language use to suit the situation. More broadly, it takes into account other people’s perspectives, intentions and the wider context in which the communication is set.

Speech is made up of phonology and articulation and is the vehicle for expressive language. Phonology refers to the speech sounds (consonants) in languages and the sequence rules for how the sounds combine to form words, how they are used in different word positions and function in languages to signal contrasts in meaning (Ball et al., 2010). Articulation refers to the motor activity to produce speech sounds.

Intelligibility refers to how much of an utterance can be understood by a listener (Bauman-Waengler 2004). Although Dodd and Bradford (2000) stated that intelligible speech is the long-term goal for children with speech disorders, intelligibility is recognised as a complex phenomenon. Kent (1994) made the point that intelligibility is variable within an individual speaker, depending on the familiarity of the listener, the setting, the nature of the spoken material, motivation and effort level.

In summary, prelinguistic, speech and language skills are multifaceted and multi-layered, and all elements are needed for effective communication. Communication development is a dynamic process between the child and the environment. The Communication Pyramid (Fig 1) shows the component parts of the communication system, including attention and listening skills, play and interaction, receptive language, expressive language, speech sounds, highlighting how communication is more than just speech/pronunciation. Development occurs in the different areas not sequentially but simultaneously, interacting with one another (Morgan 2018), and as James (1990) says ‘in just five years children move close to an adult level of linguistic and communicative competence’.



Fig. 1 Communication Pyramid

ii) *Key Challenges Associated with Cleft: prelinguistic skills, language and speech*

Children born with a cleft palate +/- lip have an altered speech mechanism with an abnormal opening between the oral and nasal cavities. Where there is a cleft of the hard palate, the tongue has no surface against which to place the tongue to make the consonant sounds of the language. Even with timely surgery in infancy, children are at risk for speech and language difficulties (Chapman, 2011; Scherer, Williams, & Proctor-Williams, 2008). Lancaster et al. (2019) in a meta-analysis of speech and language development in non-syndromic children with cleft lip and palate, from birth to age 8;11 years, concluded that young children experience delay compared to their non-affected peers on multiple speech and language constructs, performing significantly below their peers on measures of consonant inventory, (defined as the number and types of consonants a child is using in words), their accuracy, expressive language and receptive language, all which can persist into the school aged years.

Pre-linguistic Skills

The important foundations for the development of speech and language are altered from the outset. Infants with a cleft condition have been reported to look less at their parents' faces and are less communicative than infants of the same age who do not have a cleft. Mothers are in turn less responsive to the infant's cues and less actively engaged with them (Murray et al, 2018), probably compounded by early feeding problems and when parents are struggling to come to terms with the diagnosis (Russell, 2010). This may mean that parents underrate their child's potential and make fewer demands on them to participate in communication. Attempts by the child to communicate may not be recognized as words and communication not reinforced. The parent-child relationship and early communicative interaction may also be affected by parental anxieties associated with surgery(ies) in the first 12-18 months, hospitalisation and attendance at outpatient clinic appointments.

Infants with an unoperated palate have a different experience of sound production. They have difficulties due to a lack of the necessary intraoral air pressure required for oral consonants. They use instead more nasal consonants (e.g. m, n), glides (e.g. y) and glottal stops (a sound



Speech Communication, Resilience – supporting children and parents with clefts made in the throat), with more sounds generally made at the back of the mouth than at the front, irrespective of the language to which they are exposed.

Infants often have a later onset of babbling, producing fewer ‘canonical’ syllables and with oral consonants, compared with unaffected infants (Chapman et al., 2001; Hardin-Jones, Chapman, & Schulte, 2003; Scherer et al., 2008a). Canonical syllables are the typical ‘repeated syllables consisting of a consonant and a vowel such as "da da da da" or "ma ma ma ma”’, that infants produce around 7-9 months of age, and is recognised as an important milestone in communication development. Smaller consonant inventories in babble and early word productions, with less complex syllable and word structures have been reported (Chapman and Willadsen, 2011; Willadsen, 2007). A lack of oral consonants such as /b, d/ is of concern as these are typically characteristic of the early vocabulary of young children in many different languages (De-Boysse-Bardies and Vihman, 1991).

Often children continue to use the sounds they have developed before palate surgery as their speech and language emerge, reflecting the known continuity between pre-speech and subsequent development (Chapman, 2004; Chapman et al., 2003). This may explain why speech, and possibly language, may be delayed (Hardin-Jones and Chapman, 2019).

Language

Toddlers and young children with non-syndromic cleft lip and palate experience speech and both receptive and expressive language delay, showing delays in mean length of utterance (defined as the number of words used) (Scherer, Oravkinova and McBee, 2013), amount of vocabulary (Frey, Kaiser and Scherer, 2018) and grammar (Young, Purcell and Ballard, 2010). However, these differences appear to decrease with age (Lancaster et al., 2019) with older school-aged children presenting with no statistically significant language delays or deficits compared with their peers without cleft (Collett, Leroux and Speltz, 2010; Chapman 2011; Klinto, Salameh and Lohmander, 2015; Feragen et al., 2017; Boyce et al., 2018). Importantly however, these children may fall into the low average range which can affect children’s academic functioning and peer relationships (Klinto et al., 2015; Boyce et al., 2018).

Speech Difficulties

Children may present with ‘cleft speech’ where they sound hypernasal, present with nasal airflow errors, articulation errors known as cleft speech characteristics (CSCs) (John et al., 2006; Peterson-Falzone et al., 2006; Sell et al 1999; Sell et al., 2009) and/or voice disorders or voice differences (Cavalli, 2011).

Hypernasality is often considered the hallmark speech characteristic synonymous with cleft palate. It refers to a nasal tone of the voice, or excessive nasal resonance, usually due to the palate not adequately shutting off the mouth from the nose during speech, even after the palate has been repaired, a condition known as velopharyngeal insufficiency (VPI).

Nasal airflow errors, sometimes described as abnormal extra puffs of air down the nose on consonants, occur when there is any inappropriate escape of air through the nose on oral consonants such as /p, f, s/ (Sweeney 2011). A distinction is made between nasal emission and nasal turbulence; the former has a frictional, hissy quality and the latter a



Speech Communication, Resilience – supporting children and parents with clefts snorting/turbulent quality. These nasal airflow errors can be accompanied by a nasal or facial grimace.

The articulation patterns associated with cleft palate can be divided into different groups of Cleft Speech Characteristics (CSCs). One group known as passive or obligatory errors relate directly to VPI and are usually indicative of the need for secondary speech surgery. The second group of CSCs, known as active or compensatory errors, tend to be errors of place of articulation, ie where the sound is made. Errors are produced further back in the oral cavity than is usual, sometimes at the back of the mouth (posterior errors), throat or in the nose (non-oral errors). These errors need speech therapy intervention, and do not spontaneously improve (Harding and Grunwell, 1998; Kummer, 2011). It is not uncommon for passive and active errors to co-occur. Another group of errors can broadly be described as distortions often associated with malocclusions of the jaws and dental anomalies, and are described as anterior errors. Figure 2 shows where the posterior, non-oral and anterior errors are made.

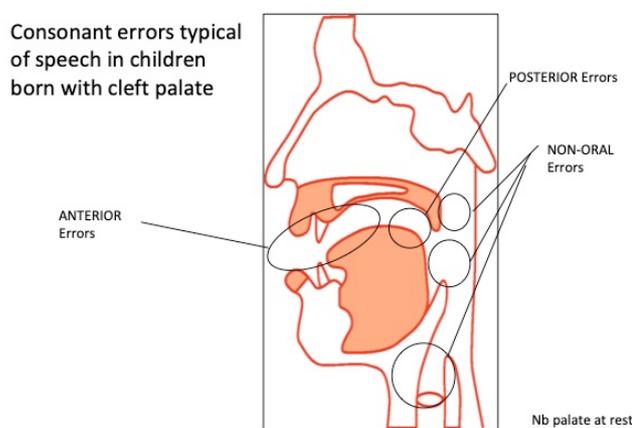


Fig. 2 Place of Articulation for Anterior, Posterior and Non-oral errors

When the CSCs become integrated into a child's speech sound system, they often affect more than one sound, limiting the ability to make meaningful contrasts between words, which is then described as an articulation disorder with phonological consequences (Harding-Bell and Howard, 2011; Howard, Heselwood and Harding-Bell, 2019). These atypical phonological patterns often co-occur with typical developmental immaturities such as 'fronting' (for example, garden is pronounced as 'darden'), or 'stopping' (for example, sun is pronounced as 'dun') (Chapman, 1993; Harding-Bell, 2019; Klinto et al., 2016; Willadsen et al., 2017). Chapman (1993) confirmed that preschool children typically present with a limited system of speech sound contrasts, characterised by atypical cleft speech characteristics (Klintö et al., 2014) compared to their peers. Even when surgery is undertaken to improve structural deficits, such as the problem of VPI, these speech difficulties persist. The consequences of these difficulties with speech is poor intelligibility (Henningsson et al., 2008; Whitehill and Chau 2004; Whitehill et al., 2011). In these cases, speech therapy intervention is indicated.

iii) Components of communication: early social interactions and the development of social processes

In addition to delays and or deficits in the building blocks of speech and language, the young child with a cleft may face other challenges to developing effective communication skills.



Speech Communication, Resilience – supporting children and parents with clefts

Emerging evidence suggests that for some, the quality of early interactions between the infant with a cleft and his/her key caregiver may be compromised, with these interactions can be less mutually responsive than interactions between carers and infants without cleft (Murray et al, 2018). The differences found in interactions between caregivers and children with or without a cleft are thought to disappear by the age of 2 years, however, more definitive longitudinal research is needed to verify whether or not the differences in early experience have lasting effects.

As a cleft can result in differences in the appearance and 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. In addition to deficits in the verbal skills underpinning effective communication, children with speech & language disorders may be further hampered by additional factors, including negative impacts on self-esteem resulting from a communication disability and/or an unusual facial appearance (Lyons & Roulstone, 2018). Several conditions with a higher prevalence in children with CL/P, including autism spectrum disorder; attention and/or hyperactivity disorders; dyslexia; learning difficulties and genetic syndromes may also contribute to delays in the development of speech and language or psychosocial development, further impacting on the quality of the child's social experiences (Feragen & Stock, 2014).

ii) Key Challenges Associated with Cleft: the Family Environment

The important part played by parents and the immediate family environment in the psychosocial development of children with and without cleft is widely acknowledged. In the early years, parents of a baby with a cleft face significant challenges, including the stress associated with the antenatal or postnatal discovery of the cleft and the challenges of caring for the infant during the early months of life. The cleft can result in feeding difficulties; other people's reactions to the condition can be distressing and parents are likely to experience a raft of conflicting emotions regarding early surgery to repair the cleft (Nelson et al 2012; Stock et al 2020). Support and intervention to ameliorate these difficulties have been recommended for parents during the first 18 months of life (Nelson et al, 2012) and peer support may be particularly helpful for parents during the early years (Stock et al 2020).

Feragen & Stock have also highlighted the importance of the wider family environment in the pre-school and primary school years. Indeed, there are indications that this influences the child's wellbeing and adjustment to a greater extent than the objective aesthetic and functional outcomes of treatment during this stage (Feragen 2012). Evidence from body image researchers suggests that parents' feelings about the child's appearance are likely to be transmitted and then assimilated by the child from an early age, influencing developing perceptions of self-worth and the child's own body image (Collins-Donnelly, 2014). Families vary in the extent to which they openly acknowledge and deal with the child's unusual appearance. Although research is lacking, the indications are that for those families in which any 'differences' are openly discussed and in which these differences are framed as 'no big deal', the child is more likely to thrive. Similarly, if parents use strategies to help the child to construct a sense of self-worth and self-esteem based on other factors and encourage their child to develop self-confidence and good social skills, the risks of poorer outcomes are substantially reduced. (Feragen & Stock, 2018). Some parents on the other hand, prefer to 'brush the issue under the carpet' for fear of upsetting the child, or perhaps themselves. In



Speech Communication, Resilience – supporting children and parents with clefts

this less supportive environment, children may also avoid raising any issues/asking questions about their cleft for fear of upsetting their parents (Rumsey & Stock 2013). Parents who openly voice their eagerness for the child to have the next surgical or nonsurgical treatment for appearance, implying that their child will in some way be ‘better’ or ‘more beautiful’ are doing their child disservice, as this may lead to an over-reliance on appearance for the child’s self-esteem and sense of self-worth.

In relation to the child’s feelings about his/her own appearance, awareness of societal appearance ideals can emerge from as young as 5 years of age (Dittmar et al, 2006), although levels of concerns are generally less of an issue before the age of 10 years than these can be for adolescents. Clarke et al (2013) have recommended that in both childhood and adolescence, young people should be encouraged to define themselves by means other than their appearance. A strong emphasis on the need for approval by others should also be avoided (Clarke et al, 2013). If parents and children appear to be heavily persuaded by the messages in social and broadcast media and advertising about the benefits of pursuing an appearance close to appearance ideals, challenges to the beauty myths voiced by health care providers and others may be useful. In particular, a heavy emphasis on the desirability of appearance-related surgery for their child by parents should be avoided if possible, as unrealistic expectations of the likely degree of change in the child’s life as the result of a change in appearance are likely to be unhelpful for both parents and their children (Rumsey & Harcourt, 2012; Stock et al 2020).

A child's communication impairment can impact parents and the extended family. McCormack et al. (2012) explored parents’ experiences of having a child with a speech impairment and identified themselves as experiencing ‘battles’ as well as their children. They reported distress when observing their children struggling to be understood and having difficulty communicating with other children. They also described feelings of guilt, isolation and fatigue. Parents are aware of the role that communication plays in education and expressed concerns regarding their child’s development of literacy as well as social skills (Sugden et al 2019). Barr et al (2008) identified the impact on siblings. They described how siblings take on the task of interpreting for the affected child when they are not understood and protecting them from bullying by others. They reported that siblings were sometimes worried and concerned about their brother or sister. Adult listeners in the child's environment (for example, teachers and grandparents), may be unable to work out what the child is saying leading to frustration for both the speaker and listener (McCormack et al 2010). Parents of children with cleft related speech disorders have reported anxiety regarding the child’s ability to play, start school and make friends, and frustration at not being able to help their child to access the help they need (Sweeney et al 2016-2017; Sweeney et al in press). Lack of access to speech and language therapy not only impacts the child but also other people within the child’s environment.

- iii) *Key challenges associated with the wider environment: social & educational processes*

Much of the literature on the impact of speech and language problems in developmental speech disorders is relevant to children with cleft palate related speech and language difficulties. All children with speech and language delay face potentially serious communication, academic and social consequences (Young et al., 2000). Speech errors can



Speech Communication, Resilience – supporting children and parents with clefts

have a significant impact on a child's intelligibility and quality of life, adversely affecting psychosocial well-being and educational development (Stothard et al., 1998; McCormack et al., 2009; Muir et al., 2011, Chapman 2011, Richman et al., 2012). Lyons and Roulstone (2018) reported that children with speech sound disorders aged nine to twelve years, were frustrated, sad and annoyed when not understood, and bullied and called names because of poor speech leading to feelings of exclusion. This disrupted communication between the child and his/her peers impacts on the ability to form friendships and social communication skills. Speech characteristics are known to influence the impressions the listener has about a person (Allard and Williams, 2008). Peers have been reported as making negative social and personal judgements and comments related to a child's speech intelligibility (Lee et al., 2017; Nyberg & Havstam 2016). Two studies (Nyberg & Havstam 2016; Nyberg et al., 2020) evaluated peer perception of children with cleft related speech disorders. The 2016 study focused on 10-year olds, while the 2020 study focused on 7-year olds. Both studies reported that listeners were sensitive to even mild articulation errors, believing that the speech was 'childish' and that the person was at risk for bullying and rejection in school. In the 2020 study, 7-year olds did not notice mild hypernasality or mild nasal airflow errors, but were sensitive to more severe problems related to nasality. Bettens et al. (2020) studied the attitudes of peers towards children with cleft palate. The attitudes included: positive and negative (such as happy, clever out-going and sad, ugly, sick; affective (such as how the child feels about listening to the speaker); and willingness to be in social contact with the speaker. They found that the better the intelligibility of speech the more positive the attitudes of peers were towards the speaker, while there was a more negative attitude towards speakers with articulation errors and hypernasality. These studies highlight that speech problems are considered a risk factor for teasing and negative impacts on social functioning, and emphasise the connection between speech and psychosocial development and the importance of collaborative care (Perry 2020, Editor, Cleft Palate Craniofacial Journal). They point to the need for an intervention program to empower children to deal with bullying and social rejection by their peers in the context of poor speech.

As the child with a cleft matures s/he will become increasingly aware that her/his appearance and/or speech is different from that of their peers. In addition to problems associated with the intelligibility of the child's speech, the children affected by a cleft lip may have unusual aspects of their appearance. These may attract attention from others in the form of 'double takes' or staring, at times accompanied by direct questions, or indirect questions to others. Whether or not the child takes these responses in their stride, they must learn to deal effectively with a range of reactions from other people on a daily basis (Feragen, 2012). Although children may be teased at a young age, comments from others begins to be assimilated by around the age of 7/8 years. All children experience some degree of teasing, but there are large differences in the extent to which this is experienced as worrisome or distressing. In a study exploring the longitudinal impact of perceived teasing on satisfaction with appearance and depressive symptoms in young people with and without a visible congenital condition, Feragen & Stock (2016) reported the result of routine psychological assessments conducted with 340 young people at ages 10 and 16 years. Experiences of teasing after the age of 10 significantly impacted on appearance evaluations and depressive symptoms in adolescent females. The impact of teasing on adolescent males may have been ameliorated in males by their reports of more positive social experiences than girls. Early identification of perceived teasing in all children to prevent the development of emotional problems and dissatisfaction with appearance is of vital importance.



For some, a cleft may be experienced as an impediment to social relationships such as friendships formation and broader peer relationships. The young person's self-perceptions and experiences of social encounters may act as disincentives to engaging in other social activities too, such as sports, hobbies or relationships. Meeting new people and forming new relationships can be challenging for those who feel particularly self-conscious about their appearance or speech. In correlational analyses, Feragen & Stock (2016) found an association between social experiences and both emotional adjustment and adaptive, pro-social behaviour.

In the educational context, it is well established that a child's speech and language skills support their reading abilities, mathematical skills and behaviour (Hammer et al., 2017). Research from a large epidemiological study of children born with non-syndromic cleft lip and palate (Ness et al., 2015) suggests that poor speech is the single biggest predictor of poor outcomes in education for this population (Grewal et al, in submission). This may be compounded by teachers underestimating the cognitive abilities of children with speech impairments (Richman 1978), having lower expectations of them, which may have a negative impact on their educational achievements (Feragen et al 2015). Interestingly, school aged children have insights into the impact of their poor speech, expressing concern regarding their future academic achievements (Lyons and Roulstone 2018). Nathan et al. (2004) found that non-cleft children whose speech problems had resolved at the time of assessment performed no differently to controls, highlighting the need for therapy to prevent ongoing academic problems.

Changing social groups can present particular challenges to those set apart from others by their speech and/or appearance. One particular transition point during this developmental period – the transition from primary to secondary school - has been highlighted by Rumsey & Harcourt (2004) & Stock et al (2020) to offer particular challenges, as people try to develop new friendships and to 'fit in' with their new peers. This transition provokes anxiety in most young people, but issues with appearance/speech may heighten fears of social rejection and 'difference'.

3) Key Goals of Cleft Care: Supporting Patients in Developing Effective Communication Skills and Psychosocial Resilience

Until recently, researchers and clinicians have expected that the impacts of a condition such as cleft involving both physical and functional differences will be predominantly negative. An additional assumption has been that the more severe a condition is, the worse will be the consequences. In recent years, however, it has become clear that any association between the noticeability of the cleft and the extent of the problems in speech and language and adjustment is at best, weak (Feragen & Stock, 2018; Crerand et al, 2020). An appreciation of the need to shift the focus of health care provision from a sole focus on optimising appearance and correcting structural anomalies optimise speech to a broader, more holistic focus on facilitating the acquisition of the coping strategies to deal with the challenges of cleft, has grown. In parallel, the spotlight of research has also shifted away from an exclusive focus on the problems and difficulties experienced by those affected by cleft to efforts to



Speech Communication, Resilience – supporting children and parents with clefts

learn more about resilience and positive adjustment in the face of these challenges.

Rosenberg & Yi-Frazier (2016), for example, recommend that health care professionals and researchers should focus on how we can help children and young people, as well as their parents, acquire the resources to equip themselves to respond to challenges effectively.

Reflecting this shift in emphasis, this project will focus on a strength-based approach to empower key players in the developing child's environment to optimise the likelihood of positive outcomes in children and young people affected by cleft.

i) Improving communication skills: The need for speech and language therapy:

Due to the complex nature of their communication difficulties, many children with cleft palate require ongoing speech and language therapy, even when they receive surgery in a timely way in the first 18 months of life. Approximately 68% of pre-school children with cleft palate require speech and language therapy (Hardin-Jones and Jones, 2005), while 50% have speech problems at 5 years of age (Britton et al. 2014) and 20% have persistent long-term intransigent speech difficulties requiring long-term therapy (Sell et al., 2001, Sell et al., 2015).

The UK/Irish standard of care is for normal or near normal speech by school entry (Murray 1998). Although the partner countries participating in the SCR4Cleft project, (Italy, Bulgaria, Malta, Estonia, Romania and Serbia) do not appear to have official standards of care for children with cleft palate, all aim to ensure that children have good speech or normal speech by school entry (5 to 7 years). Based on the literature it is extremely difficult to calculate the average amount of therapy these children require, in order to develop normal speech and language skills. Studies have shown significant improvement in articulation using different therapy approaches ranging from 30 to 112 hours of intervention (Hodson et al 1983; Pamplona et al., 1999, Pamplona et al., 2005; Derakhshandeh et al 2016), but studies rarely report the total time required to obtain normal speech and language (Vallino-Napoli 2011).

ii) Achieving Psychological Resilience:

Definitions of psychological resilience vary, but all agree on the central element - having the psychological strength and resources to cope with problems and stress and to bounce back (recover) after periods of stress or a specific calamity. A resilient person can keep his/her cool and achieve positive outcomes despite adversity (Masten 2001) and has a shorter period of rumination and worry after stressful events (Gassling et al, 2012). Rather than wellbeing being characterised by an absence of stress, exposure to greater adversity can be adaptive as it can have a 'steeling effect' and promote belief in a person's ability to cope successfully with any challenges that life may bring.

There is also agreement amongst researchers that resilience isn't inborn. Over time and in the right environment, all children (and adults) have the potential to achieve it. Resilience is perhaps best understood as the process of accumulating a 'toolbox' of adaptive resources across the lifespan (Rosenberg & Yi-Frazier 2016; Ridley, 2020). Different challenges require different mixes of resources and the degree of resilience a particular person is able to draw upon in response to stress will vary from situation to situation – hence the desirability of filling the 'toolbox' with as many resources ('tools') as possible. A definitive recipe for



the components of resilience has yet to be agreed, but there is a broad consensus about key elements.

In a qualitative study of 15 adults with cleft who were asked to reflect on their lives to date, Ridley (2020), found that the strongest indicators of resilience were optimism and the belief in a bright future (in both the child and parents), self-compassion (the ability to be kind to yourself following failure) and a positive body image/view of the self (this construct is in turn associated in the literature with self-esteem; positive affect; life satisfaction and subjective feelings of happiness). Also key in Ridley's work (2020) were differences in the participants' interpretation of their experience of having a cleft and the challenges the cleft and its treatment had posed. The ability to incorporate these experiences into the person's life story as one aspect of their identity, rather than viewing it as a defining characteristic and a 'hook' on which to hang negative experiences and distress was a strong element in positive adjustment.

Other researchers in this field have highlighted the beneficial effects of a sense of purpose (a clear idea about what they would like to be/do) in young people and adults, empathy for others (the ability to tune in to and appreciate other people's thoughts and emotions) (Stock et al, 2016). Lyons & Roulstone's (2019) study focussed on resilience in 11 children with speech and language disorders. Their qualitative data highlighted the beneficial effects on adjustment of higher levels of impulse control and self-regulation, together with a sense of autonomy and agency (the ability to act independently and to experience a sense of control over one's environment - for example, to have a range of strategies with which to respond to teasing and bullying), and positive relationships with family members and peers. In a sample of 268 young people with cleft, Feragen, Borge & Rumsey (2009) also reported that psychosocial resilience in children was also associated with higher levels satisfaction with appearance. Physical characteristics such as visibility of cleft, gender, and additional diagnosis did not contribute to explaining psychosocial resilience.

4) Current Provision of Specialist Support for Patients and Families

i) Disparities in Care and in Access to Specialist Services (Speech and language therapy & Psychology)

The provision of care for children with cleft and their families varies markedly across Europe. The 'gold standard' of multidisciplinary care is available only to a small minority. A survey conducted by EUROCRAN researchers in 2000 found that 201 teams had 194 different protocols for cleft care and there were 17 different sequences of operations. There are also some insurmountable financial obstacles to receiving adequate care (even if the expertise exists) which is true not only in some of the economically poorer states but also in some western states as well. The situation has improved over the last decade with more countries offering more centralised services (e.g. in France, the network of *Centres de Reference* and *Centres de Competence*) but there are still huge gaps in provision across all non-surgical disciplines. This is compounded by poor local, regional and national funding arrangements which mean in some countries only the cost of surgery is funded by the state. The complexities of funding health care, and the varying types of funding arrangements available across different countries makes it difficult to really get a clear overview of exactly



Speech Communication, Resilience – supporting children and parents with clefts

who pays for what, and whether treatment is affordable against any specific measure of affordability. It is clear, however, that even where speech therapy and psychology services are available, they may not be accessible because of cost. For example, The European Cleft Organisation (ECO) has been financially supporting families in Bulgaria to access speech therapy services, providing therapy for over 300 children since 2011. The European Cleft Gateway, a directory of cleft services in Europe, (www.gateway.europecleft.org) lists 103 cleft centres in Europe. Only around a half of these provide speech therapy at no cost to the child/family. In the remainder of centres parents are expected to pay some or all of the costs of speech therapy for their child. In another European study across 8 European countries (www.cleftectp.org) parents were asked whether they thought cleft care was affordable. Nearly three quarters indicated that cleft care was ‘affordable’, another quarter said it was ‘partly affordable’ (i.e. not quite affordable) with 6% saying the care was ‘not affordable’. This figure can be considered to be low, but there is still a need to have mechanisms in place to identify these groups and to ensure they do not lose out on care.

A technical report by the Committee for European Standardisation (Early Care Services for babies born with cleft lip and palate, CEN/TR 16824, 2015) recommended that every cleft centre should include a speech therapy and psychology service as part of its commitment to cleft care. In practice this is still a long way off but the development of the European Network for Craniofacial Anomalies (<https://ern-cranio.eu/about/ern-cranio/>) (one of the 24 networks for rare diseases) set up in 2018 continues to emphasise the need for universal access to speech therapy and psychology for all children born with clefts.

Accessing speech and language therapy is a problem in many parts of the world. In Australia the amount of intervention available for children with speech sound disorders is limited by insufficient funding and increased demand on SLT services (Sugden 2019). In the UK and Ireland, speech and language therapy services have been described as inadequate, inaccessible, inequitable and not evidenced based (Bercow Report – 10 years on, 2018). In England and Wales, the Clinical Reference Group for Cleft Lip and Palate (2016) undertook a survey of the eleven regional cleft specialist speech and language services and seventy five non specialist community based SLTs, regarding therapy services for children with cleft palate speech disorders. They concluded that services were based on costs, not the evidence-base or national recommendations, and that therapy was provided in small doses by non-specialist SLTs with considerable variation in service provision. In the partner countries participating in the SCR4Cleft project in this project (Italy, Bulgaria, Malta, Estonia, Romania and Serbia) access to regular speech and language therapy varies considerably with many barriers such as geographical location, costs, staffing, lack of specialist SLTs and in some cases many different languages spoken in the country (see appendix x). Furthermore, there is limited information regarding waiting times, duration and frequency of therapy, but the overall impression is that the provision of speech and language therapy is insufficient (See appendix x). Hence there is a problem, both in high income countries with established professions of speech and language therapists and low/middle income countries where there are few, for children to be able to access adequate, timely speech and language therapy.

Health care professionals across Europe have highlighted large variation in the availability of psychosocial support for patients and families with visible differences and have identified the need for greater awareness of common problems and appropriate interventions, as well as



Speech Communication, Resilience – supporting children and parents with clefts further training in order to better meet the needs of patients and their families affected by visible differences (Harcourt et al, 2018).

ii) *The impact of financial and social inequalities on access to care*

The context – pressures on healthcare systems in the EU:

Achieving universal healthcare coverage, including financial risk protection for all, is a key target (target 3.8) of the United Nations' Sustainable Development Goals (SDG). Health systems face the challenge of ageing populations and increasing demand, which can also result from non-demographic factors such as the emergence of new (often expensive) treatments. In some European countries, costs and waiting times remain important barriers to accessing healthcare. Against a background of rising demand for healthcare resources, and public budgets which are often under pressure, ensuring universal and timely access to high quality healthcare — whilst also guaranteeing the financial sustainability of health systems — is a challenge which requires increased efforts to improve the efficiency and effectiveness of health systems. (European Commission, *Inequalities in access to healthcare, A study of national policies*, 2018).

A lack of public healthcare coverage, or the provision of only a limited set of services by the public health system, may result in higher costs and affordability problems for some groups. Similarly, some types of coverage (e.g. occupational health insurance schemes) may result in easier or faster availability of healthcare for people in a better socio-economic position. Finally, the different dimensions of access, in particular population coverage, may be affected by the financing structure of healthcare and by the mechanisms linking payments into the system to access to healthcare.

People on a low income have more difficulties accessing healthcare. The share of self-reported unmet healthcare needs (especially due to cost) is usually higher among low-income households. However, other groups may also potentially have limited effective access to healthcare, such as single person households or informal workers. Labour migrants and refugees may have difficulties accessing healthcare. ((Health of Refugees and Migrants, WHO Europe Region, 2018)

Cleft care

It is no surprise that the access to good quality cleft care is subject to the same constraints as access to healthcare in general. However, the impact of not being able to access certain aspects of cleft care can make a difference between a young person that thrives and goes on to lead a fulfilling life and a young person that has been abandoned and institutionalised, with widely recognized consequences, including an increased risk of social and financial exclusion. It is often not a lack of care that is the issue, but the lack of communication (frequently amongst health professionals) that prevents a family from being referred to the right place at the right time. The 2016 report by CEN (see page 15), with an agreed set of guidelines for early cleft care, produced with input from 16 countries, was an attempt to address poor communication, especially around referrals to specialist care.



The European Patient's Forum (EPF) has broken down access to healthcare into the following subdivisions:

- Availability –whether a healthcare service or product is available in the healthcare system of a country
- Affordability –whether seeking healthcare causes financial hardship to patients
- Accessibility–Whether there are barriers, other than financial (e.g. waiting lists, geographical barriers...), that stop patients from accessing healthcare
- Adequacy –the quality of healthcare and involvement of patients in shared decision making with their healthcare professionals
- Appropriateness –whether healthcare meets the need of different groups in the population

(Defining and Measuring Access to Healthcare: *the Patients' Perspective*, EPF, 2016)

An ongoing COST Action, 'CA 16234' www.ecce.eu has adopted these headings to conduct a survey amongst patient groups to determine what the current issues are in terms of access to cleft care across 26 European countries. The results will be published at the end of Action (2021). Early results of a parallel survey amongst health professionals in the same countries has shown that access to speech and language therapy and psychosocial support is severely limited and where it does exist, is often not affordable to families.

It is worth noting that two countries participating in the SCR4Cleft project, Bulgaria and Serbia, have significant Roma populations (8.5% and 6.2% of their populations, respectively). Reaching these groups in terms of cleft care poses significant challenges. In Bulgaria, a very low level of education (often incomplete from elementary onwards) the tendency for Roma to live in big non urban communities, poor family relations, a high rate of unemployment and finally a large number of children in one family, has led to the fact that a large number of children born with a cleft are abandoned and end up in orphanages. The integration of the Roma population is extremely complex; it is a problem concerning the whole of society and can be successfully accomplished only when there is understanding and commitment on the part of both the Roma ethnic population and the majority of the country's population. (<http://www.facevalue.cc>)

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

i) The Key Role of Parents in the Development of Communication skills and in Psychosocial Development

As highlighted above, primary caregivers are considered key agents for change in their child's development and well-being, including communication and speech disorders. The family environment provides a crucial platform for the development of positive adjustment in a child with a cleft. Parents want the best for their children and aspire to optimise their child's chances of successfully dealing with life's challenges, yet often lack the specialist information and advice to help them achieve this goal.



Speech Communication, Resilience – supporting children and parents with clefts

Parental need for information and advice about how to identify early warning signs of challenges and how to address the difficulties is acute. There have been calls from parents and healthcare professionals to develop authoritative information to support caregivers in promoting positive adjustment in their children. Looking and/or sounding different does not have to be a barrier to a happy and successful life. Parents are uniquely influential in shaping their child's experience of being born and treated for a cleft, in the values, attitudes and behaviours their child develops, and in how to interpret social and cultural information. But parents need help to achieve this, particularly if the health care resources available to them are limited.

A study conducted by ECO for another Erasmus+ project (<http://www.ihem.no>) showed that a majority of health care practitioners involved in cleft care believed that parents and family environments were key to a successful overall outcome, and that more needed to be done support families directly.

On factors that contribute to successful outcomes, all felt that family played an extremely important role and if the parents coped well and the child was brought up in a positive environment, outcomes were likely to be positive. Thus parents should be assisted/aided in any way they can to cope best from the point of diagnosis. It was felt that this is best done if the parents feel they are in partnership with the cleft team to achieve the best results and that the team should ensure it is easy for them to access advice, information and support *when needed*. How to know "*when needed*" was difficult but the consensus was that a regular – periodic – chat with parents in a positive way by an appropriate member of the team to see where things are at in terms of social functioning and development would be appropriate.

Given that language is acquired in everyday interactions between children and their parents (O'Toole et al., 2019) 'parents are ideal teachers because of their responsiveness to their child's communicative attempts, their ability to closely monitor the child's communicative attempts based on proximity to the child and their ability to provide language that elaborates those attempts' (Scherer and Kaiser, 2010 pg. 431). Bowen (2010) reasoned that engaging with well-informed parents would 'tap a special therapeutic resource' (pg 410). Early intervention often involves parent training on how to promote speech and language development (Barton and Fetting 2013).

Early intervention for speech and language problems is advised in infants known to be at risk, as is the case with cleft palate. Hardin-Jones and Chapman (2019) highlighted the need for early intervention when children with cleft palate present with any of the following: delay in receptive language (understanding), delay in use of words and gestures, reduced vocalisations, a lack of plosive sounds (such as p,b,t,d) and canonical babble by 11 months. Early intervention approaches for children with cleft palate have focused on parent training/education, with the emphasis on speech sound development, phonological interventions and increase in vocabulary, with programmes that simultaneously intervene in speech and language areas (Scherer and Louw 2011).

- ii) *The potential to enhance outcomes through training for specialist and non-specialist HCPs & parents*



Speech Communication, Resilience – supporting children and parents with clefts

The work of Scherer and colleagues provides strong evidence for early interventions in children with cleft palate speech and language delay using Enhanced Milieu Training and focused stimulation (Scherer, 1999; Scherer et al., 2008b). Enhanced Milieu Teaching (EMT) involves modelling children's communication attempts and arranging the child's environment in order to develop his/her communication skills (Hancock and Kaiser, 2007). Focused stimulation refers to 'targeted parent stimulation to facilitate the child's consonant sound practice and provide feedback regarding the child's attempt at the sound' (Scherer et al., 2008b p. 26) These naturalistic approaches aim to improve communication 'because the teaching of speech and language occurs in response to the child's interest and intent to communicate' (Kaiser et al 2017, pg 808).

In the Scherer et al. (2008b) study the parents of children, aged 14 to 36 months received two to four 45-minute training sessions and worked with their children using EMT and focused stimulation. Results indicated that children in the parent-trained group showed a significant increase in vocabulary, consonant inventories, sound accuracy and a reduction in the use of glottal stops (sounds made in the throat). Ha (2015) compared the speech and language skills of two groups of children with cleft palate aged 13 to 29 months, a parent-delivered intervention group and non-intervention group. Parents were instructed in language stimulation skills and communication strategies during a 3 to 4 hour training session. She found that the children in the intervention group showed a significant increase in the vocabulary size, and mean length of utterance (average number of words in the utterance) compared to the control group. Dobbelsteyn et al. (2014) evaluated a parent-led intervention for speech disorders in older children aged 4 to 16 years. Parents completed a four hour training session, received a training manual and a workbook. Results indicated that the children made significant gains in speech production. These three studies all indicated that the parents can be trained to work on speech and language skills and increase the number of speech sounds, consonant accuracy, vocabulary and mean length of utterance post-intervention.

Recently, a mixed methods randomised controlled trial compared routine speech and language therapy with parent led, therapist supervised, articulation therapy for children with cleft related speech disorders, aged 3-7 years (Sweeney et al., in press; Sweeney et al., 2016-2017). In this study the parents completed an in-depth two-day training course. The therapist provided child specific therapy programmes, and parents carried out speech activities for 10 to 15 minutes a day, five days a week, supported by a specialist cleft SLT using mainly telehealth. The control group received therapy similar to routine care. Results indicated that both groups made statistically significant improvements in speech, activity and participation. Parents reported satisfaction with the programme and feelings of empowerment. Although a small sample size, this is the first intervention study in cleft related speech disorders in which parents have undertaken in-depth training and outcomes have been reported for impairment, activity and participation (see section on ICF-CY below).

This review shows there is a growing body of evidence showing how trained parents can effectively deliver therapy for children with cleft-related speech and language problems. Gibbard (1994) warns however that some mother/child dyads are more suitable than others for parent-led intervention. The parent needs to understand their role in therapy (Glogowska and Campbell 2000) and to see themselves as the agent of change (O'Toole et al., 2019; Sugden et al., 2019). This contrasts with the more traditional model where therapy is



Speech Communication, Resilience – supporting children and parents with clefts

delivered by a therapist, with the parent observing and being given small homework tasks to carry out at home. In this model, parents usually have minimal training and usually see themselves in a more supportive role (Sugden et al., 2019a).

Although communication is recognised as a basic need (Sell et al., 2011) the lack of access to speech and language therapy is a global issue. The problem of accessing therapy is hugely challenging in low income, and probably middle-income countries where the speech and language therapy profession and services do not exist. In others, training courses are new, and services and the profession are embryonic in their development. The challenges of providing services is impacted further by social, cultural and economic characteristics and failure to follow up on care is compounded by this non-availability of trained professionals (Balasubramanian et al 2017).

As a result of this dearth of speech and language therapy professionals, there have been initiatives to use other health care staff to provide advice and intervention, particularly in low income under-resourced countries, where most of the studies have been conducted (Sell et al., 2008). One example of this is China, where speech and language therapists are especially scarce (WHO Report, 2002). Oral maxillofacial surgeons and nurses have been trained to deliver speech and language therapy services for children with cleft lip and palate (D’Antonio 2003). Reports of effectiveness are unavailable.

Landis and Cuc (1972) first reported an approach, in which a counterpart in Vietnam was trained in the principles of articulation therapy. There are several examples of speech camps, in which intensive therapy is delivered over a number of days by a trained SLT, simultaneously training and supervising intervention delivery by non-specialists, such as physiotherapists, occupational therapists, nursery nurses, and auxiliary nursing midwives (Wirt et al, 1990 a and b, Pamplona et al 2004, 2005, Makarabhirom et al., 2015; Hanchanlert, et al 2015). In the intensive therapy programme in the Sri Lankan Cleft Lip and Palate Project (Lambadusuriya et al, 1988), five non-specialists were trained by two overseas therapists who worked on articulation skills in children and young people with cleft palate over a four month period. Ongoing therapy, once the Western SLTs, had left proved not to be sustainable (Wirt et al, 1990 a and b). Prathanee et al., (2014) combined a community-based speech therapy model delivered by a SLT, together with a week’s speech camp in Thailand. Trained speech assistants (one occupational therapist, four physiotherapists and a nurse) delivered therapy within the community over a nine month period. Although the results indicated that the number of speech errors reduced significantly following intervention, the methodology of the study was weak. For example, no information was given regarding the speech sample used, how speech was tested, who tested speech following the intervention and how reliable the testers were, making generalization of results difficult. The overriding message from the studies is that speech camps rely on there being a trained SLT.

Community based programmes are led by non-specialist community-based workers who bring together the efforts of the patient, their families and communities, along with appropriate health education, vocational and social services (D’Antonio and Nagarajan 2003). These models of speech and language therapy have been proposed in places such as India and Sri Lanka (Sell et al., 2008; Balasubramanian et al., 2018). Subramaniyan (2019) evaluated the effectiveness of community-based workers who implemented speech therapy for children with cleft palate, aged 5 to 12 years, in India. Eight community-based workers



Speech Communication, Resilience – supporting children and parents with clefts

(CBWs) attended 24 hours of training over one month. Training included information related to cleft palate and the speech characteristics, principles and methods of correction of articulation errors and skills to identify errors in placements of sounds such as /p, t, k/. Seventy four patients received therapy, 37 with the trained CBWs and 37 with an SLT. Seventy percent of the children seen by the CBWs reached the set criterion of 70% consistency in correct sound production, although it took 2 - 3 more sessions to achieve this compared to the SLT delivered therapy. The study suggests that the CBW model could serve as a viable model of intervention in places where the services of SLT is minimal.

There is great potential for health care professionals and parents to contribute to the promotion of psychosocial resilience in children affected by cleft. Ungar (2015) uses an ecological model in which resilience is determined both by the individual's capacity and by the capacity of their social ecology (home, school, social context) to promote and support effective coping. Ridley's studies with adults affected by cleft and with health care professionals (2020) both highlight the particular influence of effective support from families as a key resource in the development of resilience and the broader research literature also indicates that parents play a critical role in their child's health and wellbeing.

In Ridley's study of the views of cleft psychosocial specialists (2020), participants highlighted the important influence of the family in levels of adjustment in their children through the modelling of their own beliefs and attitudes, as well as their own behaviour (for example, in relation to teasing and 'appearance-talk' within the family; their views on the desirability of surgery to alter appearance and improve the functionality underpinning speech; in the extent to which they foster friendships for the child). Parents have the dual challenge of achieving positive adjustment for themselves as well as in their children. This is no easy task. In addition to living through the challenges experienced by their children, researchers and clinicians alike have highlighted that the impacts for parents of supporting a child with a condition such as cleft can be considerable. Parents are often plagued by the question 'Why has this happened to us' and may struggle to see the positives in having a child with a cleft. In a systematic review of the potential of interventions to improve mental health in parents of children affected by craniofacial conditions, Costa & colleagues (in preparation, 2020) highlight that additional impacts can include stress, anxiety, depression, social isolation, problems with family relationships, sleep disturbance and reduced quality of life.

Parental distress is a key risk factor for poor medical and psychosocial outcomes in children and young people and in a survey of psychosocial specialists in the UK specialising in cleft, a key recommendation is that attention should be paid to enhancing family functioning (Stock et al, 2020). Parents may be prone to a pessimistic outlook on life. Most parents worry about their child's future and want to optimise the chances of positive outcomes for their offspring. But how should they do this? Not only may they feel unsure about how to promote resilience in their children, they may lack the attributes and resources associated with resilience themselves. Access to appropriate authoritative support and intervention is limited, but it is clear that efforts to optimise mental health to promote effective coping in parents and their children is likely to pay dividends.

Research is in its infancy, but Costa et al (in preparation, 2020) have summarised the potential for interventions, focussing on how parents can deal with the management of the physical aspects of their child's condition to improve the mental health of parents. Clearly



Speech Communication, Resilience – supporting children and parents with clefts

there could be beneficial effects in extending these interventions to the acquisition of other key skills (of communication skills and resilience) with potential benefits for both parents and children. This project focuses on realising this potential. Although little work yet exists specifically relating to the skills parents of children affected by cleft may usefully acquire to promote the development of key attributes, including communication skills and resilience, in themselves and also in their children. Relevant research in related fields, including ways of promoting a positive outlook on life, self-compassion, a positive body image and speech and language development is available (see for example, Diedrichs & Halliwell, 2012; Rosenberg & Yi-Frazier, 2016; Crerand et al, 2017; Scherer et al 2008; Scherer and Kaiser 2010; Sweeney et al, in press) and will be used in this project.

6) Summary and 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.

References

Allard, E. R., & Williams, D. F. (2008). Listeners' perceptions of speech and language disorders. *Journal of communication disorders*, 41(2), 108-123.



Speech Communication, Resilience – supporting children and parents with clefts

- Balasubramaniyan, S., Raghunathan, V., Rajashekhar, B., Sathiyasekaran, B. W. C., & Nagarajan, R. (2017). Planning community-based intervention for speech for children with cleft lip and palate from rural South India: A needs assessment. *Indian Journal of Plastic Surgery*, 50(03), 295-301.
- Ball, M. J., Muller, N., & Rutter, B. (2014). *Phonology for communication disorders*. Psychology Press.
- Bauman-Waengler, J. (2004). *Articulatory and phonological impairments: A clinical focus*. Boston, USA. Pearson
- Barr, J., McLeod, S., & Daniel, G. (2008). Siblings of children with speech impairment: Cavalry on the hill. *Language, Speech, and Hearing Services in Schools*.
- Barton EE, Fetting A: Parent-implemented interventions for young children with disabilities: a review of fidelity features. *J Early Interv*. 2013; 35(2): 194–210.
- Bercow report– ten years on. (2018) <https://www.bercow10yearson.com>
- Bowen, C. (1998). Typical speech and language acquisition in infants and young children. Retrieved from <http://www.speech-language-therapy.com/> on [17.02.20]).
- Cavalli, L. (2011). Voice assessment and intervention. In S. Howard and A. Lohmander (Eds.), *Cleft Palate Speech: Assessment and intervention* (pp. 181-198). West Sussex: John Wiley & Sons Ltd.
- Chapman, K. L. (1993). Phonologic processes in children with cleft palate. *Cleft Palate-Craniofacial Journal*, 30(1), 64–72. doi: 10.1597/1545-1569_1992_029_0435_papsot_2.3.co_2.
- Chapman K. (2011). The relationship between early reading skills and speech and language performance in young children with cleft lip and palate. *The Cleft Palate–Craniofacial Journal* ;48,:301–11.
- Berger, Z. E., & Dalton, L. J. (2009). Coping with a cleft: Psychosocial adjustment of adolescents with a cleft lip and palate and their parents. *The Cleft Palate–Craniofacial Journal*, 46, 435–443. <http://dx.doi.org/10.1597/08-093.1>
- Berger, Z. E., & Dalton, L. J. (2011). Coping with a cleft II: Factors associated with psychosocial adjustment of adolescents with a cleft lip and palate and their parents. *The Cleft Palate–Craniofacial Journal*, 48, 82–90. <http://dx.doi.org/10.1597/08-094>
- Bettens K, Alighieri C, Bruneel L, De Meulemeester L, Van Lierde K (2020). Peer attitudes toward children with cleft (lip and) palate related to speech intelligibility, hypernasality and articulation, *Journal of Communication Disorders* (2020), doi: <https://doi.org/10.1016/j.jcomdis.2020.105991>



Boyce, J. O., Kilpatrick, N., Reilly, S., Da Costa, A., & Morgan, A. T. (2018). Receptive and expressive language characteristics of school-aged children with non-syndromic cleft lip and/or palate. *International Journal of Language & Communication Disorders*, 53(5), 959-968. doi:10.1111/1460-6984.12406.

Bowen, C. (1998). Typical speech and language acquisition in infants and young children. Retrieved from <http://www.speech-language-therapy.com/> on [17.02.20].

Britton, L., Albery, L., Bowden, M., Harding-Bell, A., Phippen, G., & Sell, D. (2014) A cross-sectional cohort study of speech in five-year-olds with cleft palate ± lip to support development of national audit standards: benchmarking speech standards in the United Kingdom. *Cleft Palate- Craniofacial Journal*, 51(4), 431-451.

Chapman, K.L. (2004). Is presurgery and early postsurgery performance related to speech and language outcomes at 3 years of age for children with cleft palate? *Clinical Linguistics and Phonetics*, 18(4-5), 235-57. doi: 10.1080/02699200410001693486.

Chapman, K.L., Hardin-Jones, M., & Halter, K.A. (2001). Vocal development of 9-month old babies with cleft palate. *Journal of Speech Language Hearing Research*, 44(6), 1268-83. doi: 10.1044/1092-4388(2001/099).

Chapman K. (2011). The relationship between early reading skills and speech and language performance in young children with cleft lip and palate. *The Cleft Palate–Craniofacial Journal* 48, 301–11.

Chapman, K. L., & Willadsen, E. (2011). The development of speech in children with cleft palate. *Cleft palate speech: Assessment and intervention*, 23-40.

Clarke A, Thompson A, Jenkinson E, Rumsey N & Newall R (eds), (2014) *CBT for Appearance Anxiety: Psychosocial Interventions for Anxiety due to Visible Differences*. Wiley-Blackwell

Clinical Reference Group for Cleft Lip and Palate (2016). Variation in speech and language therapy provision for children born with a Cleft Lip and Palate in England and Wales. Written and agreed by Lead SLT group and Chair of Cleft Clinical Reference Group. Retrieved from <http://www.cleftsig.co.uk/wp-content/uploads/2012/02/Report-to-NHS-England-on-SLT-Provision-in-Cleft-Care-V13-Final-Anonymised-1-3-17-june.pdf>

Collett, B. R., Leroux, B., & Speltz, M. L. (2010). Language and early reading among children with orofacial clefts. *The Cleft Palate-Craniofacial Journal*, 47(3), 284-292.

Collins-Donnelly K (2014) *Banish your Body Image Thief: A Cognitive-Behavioural Workbook on Building Positive Body Image for Young People*. Jessica Kingsley Publishers: London & Philadelphia



Costa, B. (in preparation 2020) The effectiveness of psychosocial interventions to improve psychosocial outcomes in parents of children with an appearance-affecting health condition: A systemic review.

Crerand, C. E., Sarwer, D. B., Kazak, A. E., Clarke, A., & Rumsey, N. (2017). Body Image and quality of life in adolescents with craniofacial conditions. *The Cleft Palate–Craniofacial Journal*, 54, 2–12. <http://dx.doi.org/10.1597/15-167>

Crerand C, Rumsey N, Kazak A, Clarke A, Rausch J & Sarwer D (2020) Sex differences in perceived stigmatization, body image disturbance and satisfaction with facial appearance and speech among adolescents with craniofacial conditions. *Body Image*, 32, 124-136.

Dobbelsteyn, C., Bird, E. K. R., Parker, J., Griffiths, C., Budden, A., Flood, K., Stilson, A., 2014, Effectiveness of the corrective babbling speech treatment program for children with a history of cleft palate or velopharyngeal dysfunction. *Cleft Palate-Craniofacial Journal*, 51(2), 129-144.

D’Antonio, L. L., & Nagarajan, R. (2003). Use of a consensus building approach to plan speech services for children with cleft palate in India. *Folia phoniatrica et logopaedica*, 55(6), 306-313.

de Boysson-Bardies, B., & Vihman, M. M. (1991). Adaptation to language: Evidence from babbling and first words in four languages. *Language*, 67(2), 297-319.

Derakhshandeh, F., Nikmaram, M., Hosseinabad, H. H., Memarzadeh, M., Taheri, M., Omrani, M., ... & Sell, D. (2016). Speech characteristics after articulation therapy in children with cleft palate and velopharyngeal dysfunction—A single case experimental design. *International journal of pediatric otorhinolaryngology*, 86, 104-113.

Dodd, B., & Bradford, A. (2000). A comparison of three therapy methods for children with different types of developmental phonological disorder. *International Journal of Language and Communication Disorders*, 35(2), 189-209.

Feragen, K. B., Borge, A. I. H., & Rumsey, N. (2009). Social experience in 10-year-old children born with a cleft: Exploring psychosocial resilience. *The Cleft Palate–Craniofacial Journal*, 46, 65–74. <http://dx.doi.org/10.1597/07-124.1>

Feragen, K. B., Kvaalem, I. L., Rumsey, N., & Borge, A. I. H. (2010). Adolescents with and without a facial difference: The role of friendships and social acceptance in perceptions of appearance and emotional resilience. *Body Image*, 7, 271–279. <http://dx.doi.org/10.1016/j.bodyim.2010.05.002>

Feragen K B & Stock N M (2014) When there is more than a cleft: Psychological adjustment when a cleft is associated with an additional condition. *Cleft Palate-Craniofacial Journal*, 51 (1), 5-14.



- Feragen, K. B., Stock, N. M., & Kvalem, I. L. (2015). Risk and protective factors at age 16: Psychological adjustment in children with a cleft lip and/or palate. *The Cleft Palate-Craniofacial Journal*, 52(5), 555-573.
- Feragen, K. B., & Stock, N. M. (2016). A longitudinal study of 340 young people with or without a visible difference: The impact of teasing on self-perceptions of appearance and depressive symptoms. *Body Image*, 16, 133–142. <http://dx.doi.org/10.1016/j.bodyim.2016.01.003>
- Feragen, K. B., Aukner, R., Saervold, T. K., & Hide, O. (2017). Speech, language, and reading skills in 10-year-old children with palatal clefts: The impact of additional conditions. *Journal of Communication Disorders*, 66, 1-12. doi: 10.1016/j.jcomdis.2017.03.001.
- Feragen K B & Stock N M (2018) Factors affecting subjective appearance evaluations among patients with congenital craniofacial conditions: An application of Cash’s cognitive-behavioral model of body image development. *Body Image*, 24, 124-136.
- Gibbard, D. (1994). Parental-based intervention with pre-school language-delayed children. *European Journal of Disorders of Communication*, 29(2), 131-150.
- Frey, J. R., Kaiser, A. P., & Scherer, N. J. (2018). The influences of child intelligibility and rate on caregiver responses to toddlers with and without cleft palate. *The Cleft Palate-Craniofacial Journal*, 55(2), 276-286.
- Glogowska, M., & Campbell, R. (2004). Parental views of surveillance for early speech and language difficulties. *Children & society*, 18(4), 266-277.
- Ha, S. (2015). Effectiveness of a parent-implemented intervention program for young children with cleft palate. *International Journal Of Pediatric Otorhinolaryngology*, 79(5), 707-715.
- Hanchanlert, Y, Pramakhatay, W, Pradubwong, S, Prathanee B, Pradubwong, S. (2015). Speech correction for children with cleft lip and palate by networking of community-based care. *J Med Assoc Thai*, 98(7), S132-S139.
- Hancock TB, Kaiser AP: Enhanced milieu teaching. In: McCauley RJ, Fey ME editor(s). *Treatment of Language Disorders in Children*. Baltimore (MD): Paul H Brooks, 2007; 203–36.
- Harcourt, D, Hamlet C, Feragen K, Garcia-Lopez L, Masnari O, Mendes J, Nobile F, Okkerse J, Pittermann A, Spillekom-van Koulil S, Stock N & Williamson H (2018). The provision of specialist psychosocial support for people with visible differences: A European survey. *Body Image*, 25, 35-39.



Speech Communication, Resilience – supporting children and parents with clefts

Hardin-Jones, M., Chapman, K. L., & Schulte, J. (2003). The impact of cleft type on early vocal development in babies with cleft palate. *The Cleft Palate-Craniofacial Journal*, 40(5), 453-459.

Hardin-Jones, M. A., And Jones, D. L. (2005). Speech production of pre- schoolers with cleft palate. *Cleft Palate-Craniofacial Journal*, 42(1), 7-13.

Hardin-Jones and Chapman, 2019 Early intervention for infants and young children with cleft palate. In Harding-Bell, A. (Ed.) *Case studies in cleft palate speech: Data analysis and principled intervention*. p. 91-110. UK: J&R Press.

Harding, A., & Grunwell, P. (1998). Active versus passive cleft-type speech characteristics. *International Journal of Language & Communication Disorders*, 33(3), 329-352. doi: <https://doi.org/10.1111/j.1460-6984.1995.tb01679.x>.

Harding-Bell, A. & Howard, S. (2011). Phonological approaches to speech difficulties associated with cleft palate. In S. Howard and A. Lohmander (Eds.). *Cleft palate speech: Assessment and intervention* (pp. 275-291). UK: John Wiley & Sons Ltd.

Harding-Bell, A. (Ed.). (2019). *Case studies in cleft palate speech: Data analysis and principled intervention*. UK: J&R Press.

Henningsson, G., Kuehn, D., Sell, D., Sweeney, T., Trost-Cardamone, J., & Whitehill, T. (2008). Universal parameters for reporting speech outcomes in individuals with cleft palate. *Cleft Palate-Craniofacial Journal*, 45, 1-17. doi: 10.1597/06-086.1.

Hodson, B. W., Chin, L., Redmond, B., & Simpson, R. (1983). Phonological evaluation and remediation of speech deviations of a child with a repaired cleft palate: A case study. *Journal of Speech and Hearing Disorders*, 48(1), 93-98.

Howard, S. (2010). Orofacial anomalies. In J.S. Damico, N. Muller & M.J. Ball (Eds.), *The Handbook of language and speech disorders*. Blackwell Publishing Ltd. doi: 10.1002/9781444318975

Howard, S., Heselwood B., & Harding-Bell A. (2019). The nature of speech associated with cleft palate. In A. Harding-Bell (Ed.), *Case studies in cleft palate speech: Data analysis and principled intervention* (pp.23-50). Chichester: J&R Press.

James, S. L. (1990) *Normal language acquisition*. Boston, MA: Allyn & Bacon.

John, A., Sell, D., Sweeney, T., Harding-Bell, A., & Williams, A. (2006). The Cleft audit protocol for speech-augmented: A validated and reliable measure for auditing cleft speech. *Cleft Palate-Craniofacial Journal*, 43(3), 272-288. doi: 10.1597/04-141.1.

Kaiser, A. P., Scherer, N. J., Frey, J. R., & Roberts, M. Y. (2017). The effects of enhanced milieu teaching with phonological emphasis on the speech and language skills of young children with cleft palate: a pilot study. *American journal of speech-language pathology*, 26(3), 806-818.



Kent, R. D., Miolo, G., & Bloedel, S. (1994). The intelligibility of children's speech: A review of evaluation procedures. *American Journal of Speech-Language Pathology*, 3(2), 81-95.

Klintö, K., Salameh, E.K., Svensson, H., & Lohmander, A. (2011). The impact of speech material on speech judgement in children with and without cleft palate. *International Journal of Language & Communication Disorders*, 46(3), 348-360. doi: 10.3109/13682822.2010.507615.

Klintö, K., Olsson, M., Flynn, T., Svensson, H., & Lohmander, A. (2014). Phonology in Swedish-speaking 3-year-olds born with cleft lip and palate and the relationship with consonant production at 18 months. *International Journal of Language & Communication Disorders*, 49(2), 240-254. doi: 10.1111/1460-6984.12068.

Kummer, A. (2011). Speech therapy for errors secondary to cleft palate and velopharyngeal dysfunction. *Seminars in Speech and Language*. 32(2), 191-8. doi: 10.1055/s-0031-1277721.

Lambadusuriya, S. P., Mars, M., & Ward, C. M. (1988). Sri Lankan cleft lip and palate project: a preliminary report. *Journal of the Royal Society of Medicine*, 81(12), 705-709.

Lancaster, H. S., Lien, K. M., Chow, J. C., Frey, J. R., Scherer, N. J., & Kaiser, A. P. (2019). Early speech and language development in children with nonsyndromic cleft lip and/or palate: a meta-analysis. *Journal of Speech, Language, and Hearing Research* 63(1), 14-31. doi:10.1044/2019_JSLHR-19-00162.

Lee, A., Gibbon, F. E., & Spivey, K. (2017). Children's attitudes toward peers with unintelligible speech associated with cleft lip and/or palate. *The Cleft Palate-Craniofacial Journal*, 54(3), 262-268.

Lyons R & Roustone S (2018) Well-being and resilience in children with speech and language disorders. *Journal of Speech, Language and Hearing Research*, 61, 324-344.

McCormack, J., McLeod, S., McAllister, L., & Harrison, L. J. (2009). A systematic review of the association between childhood speech impairment and participation across the lifespan. *International Journal of Speech-Language Pathology*, 11(2), 155-170.

McCormack, J., McLeod, S., McAllister, L., & Harrison, L. J. (2010). My speech problem, your listening problem, and my frustration: The experience of living with childhood speech impairment. *Language, Speech, and Hearing Services in Schools*.

McCormack, J., McAllister, L., McLeod, S., & Harrison, L. (2012). Knowing, having, doing: The battles of childhood speech impairment. *Child Language Teaching and Therapy*, 28(2), 141-157.

Makarabhirom K, Prathanee B, Suphawattariyakul R, Yoodee P (2015). Speech therapy for children with cleft lip and palate using a community-based speech therapy model with speech assistants. *J Med Assoc Thai*, 98(7), S140-S150.



Speech Communication, Resilience – supporting children and parents with clefts

Morgan S. (2018). Is the communication pyramid a useful model of language development? p26-28. May edition, Bulletin of the Royal College of Speech and Language Therapists.

Muir C, O'Callaghan MJ, Bor W, Najman JM, Williams GM. Speech concerns at 5 years and adult educational and mental health outcomes. (2011) *J Paediatr Child Health*, 47(7), 423-428.

Murray L., Bozicevic L., Ferrari P., Vaillancourt K., Dalton L., Goodacre T., Chakrabarti B., Bickness S., Cooper P, Stein A & de Pascalis L (2018) The effects of maternal mirroring on the development of infant social expressiveness: The case of infant cleft lip. *Neural Plasticity*, 2018, 1-10.

Nathan, L., Stackhouse, J., Goulandris, N., & Snowling, M. J. (2004). The development of early literacy skills among children with speech difficulties. *Journal of Speech, Language, and Hearing Research*.

Nelson P, Kirk S, Caress A & Glenny A-M (2012) Parents emotional and social experiences of caring for a child through cleft treatment. *Qualitative Health Research*, 22(3) 346-359.

Nyberg, J., & Havstam, C. (2016). Speech in 10-year-olds born with cleft lip and palate: what do peers say? *Cleft Palate-Craniofacial Journal*, 53(5), 516-526.

Nyberg, J., Hagberg, E., & Havstam, C. (2020). “She sounds like a small child or perhaps she has problems”—peers' descriptions of speech in 7-year-olds born with cleft palate. *The Cleft Palate-Craniofacial Journal*, 57(6), 707-714.

O'Toole, C., Lyons, R., Ó'Doibhlín, D., O'Farrell, F., & Houghton, C. (2020). Stage 1 Registered Report: The experiences and perceptions of parent-child interaction therapy for parents of young children with communication difficulties: A qualitative evidence synthesis protocol. *HRB Open Research*, 2(36), 36.

Pamplona, M. C., Ysunza, A., & Espinosa, J. (1999). A comparative trial of two modalities of speech intervention for compensatory articulation in cleft palate children, phonologic approach versus articulatory approach. *International journal of pediatric otorhinolaryngology*, 49(1), 21-26.

Pamplona, M. C., Ysunza, A., & Ramírez, P. (2004). Naturalistic intervention in cleft palate children. *International journal of pediatric otorhinolaryngology*, 68(1), 75-81.

Pamplona, C., Ysunza, A., Patiño, C., Ramírez, E., Drucker, M., & Mazón, J. J. (2005). Speech summer camp for treating articulation disorders in cleft palate patients. *International Journal of Pediatric Otorhinolaryngology*, 69(3), 351-359.

Peterson-Falzone, S.J., Trost-Cardamone, J.E., Karnell, M.P., & Hardin-Jones, M.A. (2006). *The Clinician's Guide to Treating Cleft Palate Speech*. US: Mosby Elsevier.

Prathanee, B., Makarabhirom, K., Pummnum, T., Seepuham, C., Jaiyong, P., & Pradubwong, S. (2014). *Khon Kaen: a community-based speech therapy model for an area*



Speech Communication, Resilience – supporting children and parents with clefts lacking in speech services for clefts. *Southeast Asian Journal of Tropical Medicine and Public Health*, 45(5), 1182.

Richman LC. (1978). Parents and teachers: differing views of behavior of cleft palate children. *The Cleft palate journal*. 1978;15(4):360-364

Richman LC, McCoy TE, Conrad AL, Nopoulos PC. (2012) Neuropsychological, behavioural and academic sequelae of cleft: early developmental, school age and adolescent/young adult outcomes. *The Cleft Palate Craniofacial Journal*, 49, 387-396

Ridley M (2020) Factors Contributing Towards Psychological Resilience in Individuals with and without Cleft Lip and/or Palate. Unpublished PhD thesis, UWE Bristol UK.

Rosenberg, A & Yi-Frazier J (2016) Commentary: Resilience defined: An alternative perspective. *Journal of Pediatric Psychology*, 41(5) 506-509

Rumsey N & Stock N (2013) Living with a Cleft: Psychological Challenges, Support & Intervention. Chapter 45 in S. Berkowitz (ed) *Cleft Lip & Palate: Diagnosis & Management*. 3rd ed. Springer-Verlag

Russell, J. (2010). Orofacial anomalies. In J.S. Damico, N. Muller & M.J. Ball (Eds.), *The Handbook of language and speech disorders* (pp. 474-496). Blackwell Publishing Ltd. doi: 10.1002/9781444318975

Scherer, N. J. (1999). The speech and language status of toddlers with cleft lip and/or palate following early vocabulary intervention. *American Journal of Speech-Language Pathology*, 8(1), 81-93.

Scherer, N. J., Williams, A. L., & Proctor-Williams, K. (2008a). Early and later vocalization skills in children with and without cleft palate. *International Journal of Pediatric Otorhinolaryngology*, 72(6), 827-840.

Scherer, N. J., D'antonio, L. L., and McGahey, H. (2008b). Early intervention for speech impairment in children with cleft palate. *Cleft Palate- Craniofacial Journal*, 45(1), 18-31.

Scherer, N. J., & Kaiser, A. (2010). Enhanced milieu teaching/Phonological emphasis: Application for children with cleft lip and palate. *Speech sound disorders in children*, 427-452.

Scherer, N. J., & Louw, B. (2011). Early communication assessment and intervention. *Cleft Palate Speech: Assessment and Intervention*. West Sussex, UK: Wiley-Blackwell, 259-274.

Scherer, N. J., Oravkinova, Z., & McBee, M. T. (2013). Longitudinal comparison of early speech and language milestones in children with cleft palate: a comparison of US and Slovak children. *Clinical Linguistics & Phonetics*, 27(6-7), 404-418.



Speech Communication, Resilience – supporting children and parents with clefts

Sell, D., Harding, A., & Grunwell, P. (1999). GOS. SP. ASS.'98: an assessment for speech disorders associated with cleft palate and/or velopharyngeal dysfunction (revised). *International Journal of Language & Communication Disorders*, 34(1), 17-33. doi: 10.1080/136828299247595

Sell, D., Grunwell, P., Mildinhal, S., Murphy, T., Cornish, T. A., Bearn, D., ... & Sandy, J. R. (2001). Cleft lip and palate care in the United Kingdom—the Clinical Standards Advisory Group (CSAG) Study. Part 3: speech outcomes. *The Cleft palate-craniofacial journal*, 38(1), 30-37.

Sell, D., John, A., Harding-Bell, A., Sweeney, T., Hegarty, F., & Freeman, J. (2009). Cleft Audit Protocol for Speech (CAPS-A): A comprehensive training package for speech analysis. *International Journal of Language & Communication Disorders*, 44(4), 529-548. doi: 10.1080/13682820802196815.

Sell D, Nagarajan R, Wickenden M. Cleft Palate speech in the majority world: models of intervention and speech outcomes in diverse cultural and language contexts. *Cleft Palate Speech: Assessment and Intervention* eds. Howard and Lohmander. p. 105-121. pub. Wiley

Sell D, Mildinhal S, Albery L, Wills Ak, Sandy Jr, Ness Ar. (2015). Perceptual speech outcomes in the Cleft Care UK study. *Orthod Craniofac Res* 2015 18 Suppl 2(S2): 36-46.

Stock N M, Feragen K B & Rumsey N (2016) Adult narratives of growing up with a cleft lip and/or palate: Factors associated with psychological adjustment. *The Cleft Palate-Craniofacial Journal*, 53(2), 222-239

Stock N M, Feragen K B, Moss T & Rumsey N (2018) Toward a conceptual and methodological shift in craniofacial research. *The Cleft Palate-Craniofacial Journal*, 55(1) 105-111

Stock N, Costa, B, Williams J, Martindale, A & the VTCT Foundation Research Team at the Centre for Appearance Research (2019) Breaking the news: Parents experiences of receiving an antenatal diagnosis of cleft lip. *Cleft Palate-Craniofacial Journal*, 56(9) 1149-1156 DOI: [10.1177/1055665619830884](https://doi.org/10.1177/1055665619830884)

Stock N M & Costa B (2020) Provision of care for families affected by craniofacial conditions: The views of non-specialist health professionals. *The Cleft Palate-Craniofacial Journal*, 57(4) 470-476.

Subramanian, B. (2019). Effectiveness of Community Based Worker Implemented Speech Correction Program for Children with Repaired Cleft Lip and Palate in Rural Districts of Tamil Nadu. (Unpublished doctoral dissertation) submitted to SRIHER (DU), Chennai, India Sugden, E., Munro, N., Trivett, Cm., Baker, E., Lynn Williams, A., 2019 Parent experiences of completing home practice for speech sound disorders. *Journal of Early Intervention*, 1-23.

Sugden, E., Baker, E., Lynn Williams, A., Munro, N., Trivett, Cm., 2019a. Evaluation of parent- and speech-language pathologist-delivered multiple oppositions intervention for



Speech Communication, Resilience – supporting children and parents with clefts
children with phonological impairment: a multiple baseline design approach. [online].
Available: https://doi.org/10.1044/2019_AJSLP-18-0248 (Accessed 23 January 2020)

Sweeney, T. (2011). Nasality assessment and intervention. In S. J. Howard & A. Lohmander (Eds.) *Cleft palate speech: Assessment and intervention* (pp.199-220). Oxford: John Wiley & Sons Ltd.

Sweeney T, Sell D, Hegarty F., 2016-2017, Parent Led Articulation Therapy in cleft palate speech: A feasibility study. *Journal of Clinical Speech & Language Studies*, 23, 21-41.

Sweeney T, Hegarty F, Powell K, Deasy L, O'Regan M, Sell D. (in press). A Randomised Controlled Trial Comparing Parent Led Therapist Supervised Articulation Therapy (PLAT) with Routine Intervention for Children with speech disorders associated with cleft palate. *International Journal for Language and Communication Disorders*.

Vallino-Napoli, L. (2011). *Evaluation & evidence based practice. Cleft Palate Speech*. Chichester: John Wiley and Sons, 317-54.

Whitehill, T. L., & Chau, C. H. F. (2004). Single-word intelligibility in speakers with repaired cleft palate. *Clinical Linguistics & Phonetics*, 18(4-5), 341-355.

Whitehill, T.L., Gitzke C.L., & Hodge M. (2011) *Speech intelligibility. Cleft Palate Speech Assessment and intervention*. Eds Howard S. & Lohmander A. Wiley-Blackwell, 293-301.

Willadsen, E. (2007). *From babbling to meaningful speech in Danish children born with and without cleft lip and palate. Unpublished doctoral dissertation*. University of Aarhus, Aarhus, Denmark.

Willadsen, E., Lohmander, A., Persson, C., Lundeborg, I., Alaluusua, S., Aukner, R., ... & Emborg, B. (2017). Scandleft randomised trials of primary surgery for unilateral cleft lip and palate: Speech outcomes in 5-year-olds-consonant proficiency and errors. *Journal of Plastic Surgery and Hand Surgery*, 51(1), 38-51. doi: 10.1080/2000656X.2016.1254647

Young, A. R., Beitchman, J.H., Johnson, C., Douglas, L., Atkinson, L., Escobar, M., & Wilson, B. (2002). Young adult academic outcomes in a longitudinal sample of early identified language impaired and control children. *Journal of Child Psychology and Psychiatry*, 43, 635–645. doi: 10.1111/1469- 7610.0005

Young, S. E., Purcell, A. A., & Ballard, K. J. (2010). Expressive language skills in Chinese Singaporean preschoolers with nonsyndromic cleft lip and/or palate. *International Journal of Pediatric Otorhinolaryngology*, 74(5), 456-464.



Appendix

Speech and Language Therapy Services. Survey of participating partner countries (Bulgaria, Estonia, Italy, Malta, Romania and Serbia).

- 1. Are there any recommended standards of care in your country in relation Speech and Language for children with cleft palate e.g., in Ire/UK our standard is for the child to have near normal speech by school entry. Do you have any equivalent standards in your country?**

Bulgaria: For the last 10-15 years, the only organization that takes care of speech therapy for children with clefts is ALA. The association develops and applies standards for speech therapy diagnostics, therapy and prevention of cleft-related problems. We are open to train anyone who is interested and wants to work through annual meetings of speech therapists, where we exchange practices, discuss cases and introduce as a standard the work with the Electronic Medical Record for Facial Anomaly EMRFA. The system is as follows - the diagnostic criteria are included in the posts of the EMRFA, the speech therapist must describe each case according to the parameters set in the EMRFA. The minimum therapeutic procedures are also described in the EMRFA, and each speech therapist can add therapeutic procedures according to his school. An important part of the team's work is the system of Global assessments in several age diapasons, which is the only standard for case tracking in Bulgaria in general. Thus, the speech therapist is involved in multidisciplinary treatment. For several months now, we have been developing a relevant post for early assessment of development and early intervention in children with cleft, as the goal is for every child to have normal or close to normal speech by school age. We believe that children with cleft should be consulted and work with them and families should begin as early as the pre-speech period. So in Bulgaria the standards for speech therapy for clefts are developed and applied by specialists related to the ALA Association. Against the background of the overall picture in the country only ALA



Speech Communication, Resilience – supporting children and parents with clefts

boasts the existence of rules and standard procedures for practicing, documentation, financing and monitoring the development of children with clefts.

Estonia: The recommended standard is not only for cleft palate children but in general. We aim that the children have a normal speech by the age of six or by school entry. Estonian children go to school at the age of 7.

Italy: No, in Italy we don't have a recommended standard, but of course the speech therapist tries to work in order to make sure that the child goes to school with a good speech (inventory of sounds and quality of voice) and if needed usually we ask to control the requirements for learning abilities.

Malta: As far as I know we follow same standards of care as per UK....targeting near normal speech by school entry.

Romania: There are no recommended standards regarding speech and language in Romania, but the purpose of the speech therapy is to obtain a quality of speech as close as possible to the normal, preferably as the child starts school.

2. Can you briefly summarise the current provision of SLT services for children with cleft palate in your country?

Bulgaria: Children with cleft in Bulgaria can receive speech therapy in the following places:

In the Department of Plastic Surgery in Plovdiv - counseling and therapy. From a Trusted Speech Therapist of ALA - after a recommendation and referral from a speech therapist in Plovdiv - at the place of residence or close to home. There are 26 registered colleagues in the Trusted Speech Therapists network. In kindergarten (3 to 7 years), if a speech therapist is appointed. Recently, the policies are such that the kindergarten often offers such a specialist. At school (7 to 18 years), if a speech therapist is appointed and at the request of the family. Larger schools usually have a speech therapist or a district speech therapist who serves several schools.

In the Resource Center (3 to 18 years) - every major city has a Center that serves children with special educational needs and has a speech therapist, resource teacher, psychologist but the therapy offered there is too general From a private speech therapist (there are many in big cities and in the capital)

Estonia: All CLP children receive SLP services. It is covered by health insurance. Some children may not receive proper speech therapy because of language barriers (e.g Russian children) or because they live in rural areas. SLP work in kindergartens, hospitals, clinics - the amount of speech therapy is not regulated. If needed, the child gets therapy as long as needed.

Italy: Speech therapy is usually provided by the public service; we don't have speech therapist in schools (few exceptions in private schools). The main problems are due to timing: services are often full and in order to start therapy there may be a long



Speech Communication, Resilience – supporting children and parents with clefts

waiting list. Private practices are present in all the country, and the waiting lists for receiving therapies there are usually better.

In Italy there are few centers that offer specific speech therapy for cleft patients, thus the main centers may give advices to the units all around the country. Right now, I'm trying to collect the names of the different specialists that work in the main cleft centers.

In Milan, we offer speech therapy either at the hospital or with telerehabilitation platforms, while no assessment is done online.

Malta: SLT service is offered neonatally in the acute general hospital and in primary health centres once the infant is discharged from hospital. Older children are also followed up in schools in some instances.

Romania: There are no standardized tests and we have no protocols regarding speech therapy for children with cleft. The purpose of the therapy is for the child to manage to emit sounds in the correct manner and to integrate the emitted sounds into words, being able to speak normally and to make himself understood by those around him.

In the cleft teams around the country we have speech therapists who evaluate the patient and start treatment. The majority of them are working in private centers. Afterwards the therapy is ensured by the local School Speech Centers or by another speech therapist in a local private center. In our School Speech Centers are included in therapy programs children above 5 years of age. Unfortunately the speech therapists dealing with cleft are not coordinated in a national, standardized manner.

Serbia: In our country we have speech therapists, but only few of them work with children with cleft palate. More of them work better in private centers.

3. **Are there any problems accessing regular speech and language therapy (SLT)?**

Bulgaria: As far as the multidisciplinary cleft treatment is not provided by the National health care system, the access to a regular cleft specific SLT is not granted. Thanks to ALA's supported network of professionals, there is no problem for the families to access SLT in the biggest cities of Bulgaria. At the same time in many small cities there are problems accessing regular speech therapy. The parents are referred to the closets professionals and sometimes it is very distant location.

Estonia: Yes, we do not have enough SLT's in Estonia. Especially in rural areas.

Italy: The main problem in accessing regular speech and language therapy in Italy – whether in a public structure or in a National Health System affiliated private clinic or practice – is the waiting lists; it's often quicker to have access in a private practice (there is a large number of them in every big city in Italy), but in this case the family would have to cope with the cost of a periodic speech and therapy treatment. For this reason, accessing speech and language therapy can be difficult for socially disadvantaged people.



Malta: No, we fall under the quality charter to have the first appointment following a referral within 15 days. Then, depending on the waiting list and client's needs a child may be seen once a week/fortnight or monthly. S/he may also be followed at school; at the moment SLPs are trying to engage in telepractice.

Serbia: There are some problems accessing regular speech and language therapy (SLT).

4. Have you any figures regarding the numbers on waiting lists or the time children have to wait to get therapy?

Bulgaria: As it is not nationally provided and organized multidisciplinary cleft treatment there is no official statistics and we cannot give numbers on waiting list.

Estonia: Waiting list is about 2months

Italy: It's basically impossible to know a precise waiting time or the number of people waiting (we tried to retrieve some information and figures from our region online portal but there's no such information accessible to the public) but given our experience in dealing with kids who cannot access public services for regular speech therapy, we can assess an average waiting time which can go from a year (and this is our best case scenario) up until two years, two years and a half.

Malta: No

Serbia: No, we haven't. In our country speech therapists aren't electronically connected yet.

5. Is there any information about when, how often and how much therapy children with cleft palate receive, are they getting enough?

Bulgaria: We do not have such information. ALA is providing 20 sessions of speech therapy to each child with cleft palate and these sessions are delivered in 8 Bulgarian cities.

Estonia: It depends on clinics: some clinics offer unlimited services for CLP patients, other clinics offer 10x per year. Most kindergartens and schools have SLT's. Some children receive their therapy at school.

Italy: No, the only information we receive about this matter is given to us during the parent interview, which happens before every meeting with a child: from the subsequent evaluation we try to understand if the numbers of sessions which that particular kid is receiving are enough or if it could be useful to add more speech



Speech Communication, Resilience – supporting children and parents with clefts

therapy stimulation; we also try to determine if the activities proposed by the speech therapist are suited for the kid pathology.

When necessary and according to the parents, we are available to talk directly to the colleagues that follow their child, in order to share observations and advices or updates on the kid articulatory/phonatory status.

Malta: Parents often claim that they are not seen often enough. There are waiting lists and clients are prioritised once they are assessed depending on specific needs.

Serbia: We haven't any information in our country about when how often and how much therapy children with cleft palate receive, and are getting enough.

6. **Are there other health care professionals delivering SLT services to children with cleft palate?**

Bulgaria: As far as we know there are no other specialists providing SLT.

Estonia: No

Italy: No, the only other figure involved in our same area of influence is the IBCLC (lactation consultant) who gives assistance to the mother during the first phases of feeding in newborns, in case of difficulties during initial latchment, nutrients shortage or lack of growth.

Malta: Sure, they can be referred to the Child Development Assessment Unit, where a team of allied health professionals assess the children, including clinical psychologist, OT, PT and paediatrician, issue reports and refer for respective services where necessary. The orthodontist and ENT specialist often sees these children also.

Serbia: No, there aren't. There are only speech therapists.

7. **Do parents ever have to pay for regular speech and language therapy?**

Bulgaria: Once they have used the funding provided by ALA the parents have to pay the SLT of their children to the end of the treatment.

Estonia: No

Italy: In private practices you always have to pay for every session attended, and the cost may vary from a professional to another professional (in Milan 45 minutes of speech and language can vary from a minimum of 40-45€ up to 70-75€).

In public structure the situation depends on your region of residence; in some cases you have to pay a ticket in order to get a first visit from a child psychiatrist or a phoniatician and then proceed with a cycle of speech therapy, in other cases (for



Speech Communication, Resilience – supporting children and parents with clefts
example in our region, Lombardy) the costs are completely covered by the health sanitary system.

Malta: Not through the state service. However, many parents take their kids to private clinics also. Sometimes, they refer them to the University clinic as well.

Serbia: The majority of speech therapists are working in private centers.

Compiled June 2020