Frequently Asked Questions by Speech-Language Therapists on Cleft Palate +/- Lip Evidence-Based Answers

The Cleft Palate and Craniofacial Committee

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Introduction

A craniofacial disorder is an umbrella term that refers to a structural abnormality of the skull and/or the face. Most craniofacial disorders are present at birth, and the type and severity of the abnormality can vary widely. The most common craniofacial disorder is Cleft Palate ± Lip (see below). Less common and rare craniofacial disorders include craniofacial dysostoses syndromes such as Apert syndrome and Crouzon syndrome, which are characterised by premature closure of fibrous joints (cranial sutures) between the skull bones (craniosynostosis), as well as distinctive facial abnormalities (e.g. reduction in growth of the upper jaw, cheekbones and eye sockets). In craniosynostosis, the abnormally shaped skull can restrict growth of the brain that can lead to increased pressure in the brain requiring medical intervention. Other types of craniofacial disorders include malformation of the outer ear (microtia) and underdevelopment of one or both sides of the face (hemifacial microsomia).

The rest of this document focuses on Cleft Palate ± Lip, within a context where a Cleft Lip and Palate Team and/or speech and language therapy (SLT) services are available. Where there is limited or no access to SLT services, there have been initiatives to use other health care staff to provide advice and intervention (Sell et al., 2008). These include training other health care professionals to deliver speech and language therapy (D'Antonio & Nagarajan, 2003), speech camps where intensive therapy is delivered by a trained SLT while simultaneously training other health care professionals (Pamplona et al., 2004, 2005; Wirt,

Wyatt, Sell, Grunwell, et al., 1990; Wirt, Wyatt, Sell, Mars, et al., 1990) and training community based workers (Balasubramaniyan et al., 2018).

What is Cleft Palate +/- Lip?

Cleft Palate ± Lip (CP±L) is the most common congenital craniofacial disorder. It can manifest in various expressions, ranging from a small split of the upper lip (i.e. incomplete cleft lip) or the soft palate (i.e. bifid uvula) to a cleft of the upper lip, alveolus, and the soft and hard palate (i.e. complete cleft lip and palate). Moreover, the cleft can occur on one side of the face (i.e. unilateral) or both sides of the face (i.e. bilateral).

Cleft Palate ± Lip is the result of disrupted embryological development of the primary palate (upper lip and alveolus) and secondary palate (hard palate and velum) during the first trimester of pregnancy. The disruption to the formation of the lip and palate is caused by genetic or a chromosomal condition, or environmental factors (e.g. cigarette smoke, alcohol, maternal use of several medicinal drugs etc.). Although the exact causal factors remain unknown, consensus exists about the multifactorial nature of the causes (Kummer, 2020).

Orofacial clefts arise in 1.7 per 1000 live born babies (Mossey & Castilla, 2003); however, the prevalence varies when variables such as cleft type, gender and ethnicity are considered. Regarding cleft type, cleft lip with or without cleft palate is more prevalent compared to cleft palate only (0.91 vs. 0.62 per 1000) and cleft lip and palate arises more often in comparison with cleft lip only (Calzolari et al., 2004, 2007). A predominance in females is observed for cleft palate (male/female-ratio 0.83 to 0.93), whereas cleft lip with or without cleft palate most typically occurs in males (male/female-ratio 1.70 to 1.81) (Calzolari et al., 2004, 2007; Mossey & Castilla, 2003; Mossey et al., 2009). Studies of ethnic and geographical differences in prevalence of orofacial clefts showed overall higher prevalence of cleft lip and/or palate in Asians, a lower prevalence for Africans and an intermediate prevalence for Caucasians. This racial and ethnical

variation might be explained by a mixture of differences in environmental risk factors and genetic predisposition.

What are the potential consequences of Cleft Palate ± Lip?

Feeding: In the first year of life, feeding may be a challenge if there is a palate involvement. Feeding difficulties in CP±L centre on the infant's inability to suck, which causes a disruption of the oral phase of swallowing (Miller, 2011). The infant is unable to create negative intraoral pressure due to the unrepaired cleft palate which prevents a separation of the oral from the nasal cavities. This can result in increased feeding times, nasal regurgitation, inadequate volume of oral intake, excessive intake of air, feeding fatigue and the possible consequential ulceration of the nasal mucosa (Bannister, 2001; Miller, 2011), all of which may impact negatively on weight gain. See guidelines below on feeding.

Middle ear diseases: Middle ear diseases such as otitis media with effusion and acute otitis media frequently occur in young children with CP±L up to 5-6 years of age (Goudy et al., 2006). The high incidence of middle ear problems in CP±L is often due to eustachian tube dysfunction. The functions of the eustachian tube are to provide aeration of the middle ear, equalization of pressure between the middle ear and atmospheric pressure, and drainage of any middle ear fluid and secretions into the nasopharynx (Peterson-Falzone, Hardin-Jones & Karnell, 2001). It has been suggestd that the eustachian tube dysfunction is a result of aberrations of the tensor veli palatini muscle, but the exact relationship between the tensor veli palatini muscle and Eustachian tube function in CP±L is still not well-defined (Heidsieck et al., 2016). It has been reported that deviations in the anatomy of the eustachian tube cartilages as well as abnormal insertion of the tensor veli palatini and levator veli palatini muscles often lead to malfunctioning

of the eustachian tube, resulting in accumulation of fluid in the middle ear which may cause a conductive hearing loss varying from 10 to 40 dB (Lennox, 2001). The hearing loss can have a significant negative effect on the development of speech and language (Willging & Kummer, 2008).

Dental abnormalities: The early stages of dental development are characterised by delayed eruption of primary teeth on the cleft side, congenital absence of primary teeth, abnormal morphology of teeth, extra teeth (supernumerary) around the cleft site, resulting in an increased risk of dental carries and hence, a focus on oral hygiene is needed (Huebener, 2016). Dental development continues to be characterised by dental anomalies and many dental interventions are required at the mixed dentition stage between the ages of 6 to 12 years (Huebener, 2016). Repair of the lip and palate can also result in narrowing of the (palate) arch form resulting in anterior and posterior crossbites (Daskalogiannakis & Antonarakis, 2016), potentially affecting oral hygiene and speech articulation.

Facial growth: Abnormal or deficient facial growth is also a potential consequence in CP±L, attributable to a combination of intrinsic developmental deficiencies, iatrogenic condition (as a result of treatment), and functional or adaptive factors (Kreiborg, Hermann, & Darvann, 2013). Abnormal facial growth becomes increasingly evident during the pubertal growth spurt resulting in a concave facial profile and a class III jaw relationship (Ross, 1987; Semb, 1991). This corresponds with the time when appearance becomes an important element for self-esteem and sexual identity (Bradbury & Bannister, 2005). A reported 10-50% of individuals with CP±L will require orthognathic surgery to correct this facial deformity (e.g. Friede, Liljia & Lohmander, 2011; Good, Mulliken & Padwa, 2007) The orthognathic surgery is typically carried out around 17-18 years of age, when active facial growth is complete. The surgery can have a negative

impact on speech and velopharyngeal function, necessitating pre- and post-operative assessments (e.g. Pereira, Sell & Tuomainen, 2013).

Speech & language: Speech difficulties in CP±L range from early speech difficulties to speech difficulties related to velopharyngeal insufficiency and/or speech difficulties related to dental anomalies and class III occlusal status. There is increasing evidence of language difficulties particularly in toddlers with CLP and poorer academic achievement in the later school-age years. These are described in detail in a later section.

Psychosocial aspects and (health-related) quality of life: Potential risks associated with CP±L are medical risks associated with having a birth defect (e.g. feeding in infancy), functional and/or cognitive problems (e.g. speech and language, academic achievement) and psychological stress (e.g. stresses on the family around treatment options and medical interventions) (Kapp-Simon & Gaither, 2016). These potential risks can impact on the individual's social and emotional adjustment and well-being, self-concept, self-esteem, satisfaction with appearance, body image and quality of life (Hunt et al., 2005; Kapp-Simon & Gaither, 2016; Stock & Feragen, 2016). In spite of these many challenges faced by the individual with CP±L across the lifespan, reviews of studies have concluded that the impact of having a CP±L on psychological aspects and quality of life is low (Hunt et al., 2005; Stock & Feragen, 2016) and that many individuals lead "happy and productive lives" (Kapp-Simon & Gaither, 2016, p. 397). For example, in the review by Stock and Feragen (2016) they concluded that although dissatisfaction with appearance may be a specific concern in CP±L, this is comparable to or better than levels reported in the general population, and that there is no strong evidence that anxiety and depression levels are raised in CP±L. As psychological adjustment can fluctuate across the lifespan, formal psychological support should be given even in childhood and continued on into adulthood (Kappen et al., 2019). Resistance factors such as family strengths and adaptation (e.g. family cohesion and emotional well-being,

social/financial support) and the child's resilience (e.g. hardiness, spirit) are necessary elements and early identification and management of potential problems are crucial (Kapp-Simon & Gaither, 2016).

What are typical speech characteristics in patients with Cleft Palate ± Lip?

Normal velopharyngeal closure is the coordinated action of the velum (soft palate), the lateral pharyngeal walls and the posterior pharyngeal walls (Kummer, 2008). Velopharyngeal dysfunction (VPD) in speech is a generic term used to describe impairment of function due to structural deficits (velopharyngeal insufficiency), neurological disorders (velopharyngeal incompetency) and faulty learning (velopharyngeal mislearning) (D'Antonio & Scherer, 1995). Atypical articulatory errors often referred to as cleft speech characteristics (CSCs) are associated with cleft palate and/or velopharyngeal dysfunction (VPD) (Peterson-Falzone et al., 2006; Sell, Harding, & Grunwell, 1999). Cleft speech consists not only of consonant sound errors but abnormal resonance (hypernasality, hyponasality) and nasal airflow errors (audible nasal emission, nasal turbulence) may also be present.

Articulatory errors can be due to structural aetiologies such as a fistula or velopharyngeal insufficiency (described as passive characteristics) or due to active mislearning sometimes associated with a history of VPD, and frequently these co-occur. Cleft speech chracteristics (CSCs) tend to be errors in place of articulation, where the sound is produced further back in the oral cavity e.g. alveolar targets are produced as palatals or velars/uvulars or further back in the larynx, pharynx, or velopharynx as glottal stops, pharyngeal fricatives or active nasal fricatives. Passive errors may affect manner of articulation resulting in weak, nasalised or absent oral

pressure consonants. Children with cleft palate may also have phonological delay and/or disorder.

Articulatory errors may also be attributable to a class III jaw relationship which tends to affect production of speech sounds such as labiodental fricatives /f/ (which may be produced as reverse labiodentals) and alveolars /t/ and /s/ (which may be dentalised or interdentalized) (Pereira, Sell & Tuomainen, 2013; Vallino, 1990). These articulation errors are not responsive to speech therapy and tend to improve spontaneously following orthognathic surgery (Lee et al., 2002; Pereira, 2012).

The most frequently occurring resonance disorder is hypernasality. Hypernasality refers to excess nasal resonance perceived during speech production due to a coupling of the oral and nasal resonating cavities (Sweeney, 2011). Nasal emission and nasal turbulence comprise the escape of audible air through the nasal cavity during the production of oral pressure consonants (plosives, fricatives and affricates). In the case of nasal emission, the sound quality is frictional but not turbulent or snorting (Sweeney, 2011). It can be due to incomplete closure of the velopharyngeal sphincter or a palatal fistula. Nasal turbulence is distinguished from nasal emission as the sound quality is turbulent or snorting and it is often due to a small gap in the velopharyngeal sphincter. Both nasal emission and nasal turbulence accompany the oral consonant, unlike the CSC, nasal fricative, which replaces the consonant.

Early speech and language development may be compromised in children with CP±L. Young children may experience delay relative to their non-affected peers across multiple speech and language constructs, performing significantly below their peers on measures of consonant inventory, speech accuracy, expressive language and receptive language (Lancaster et al., 2019). Infants may have a later onset of babbling, which can be characterised by fewer canonical syllables and fewer oral consonants compared with non-cleft infants. Instead, they produce

more nasal consonants, more posterior sounds than anterior sounds e.g. pharyngeal and glottal realizations both in babble and early word productions across languages (Chapman, 2011; Chapman & Willadsen, 2011; Willadsen, 2007). They have smaller consonant inventories, both in terms of the types of sounds (plosives, fricatives), individual sounds and less complex syllable and word structures (Chapman & Willadsen, 2007).

How are speech disorders in patients with Cleft Palate ± Lip assessed and diagnosed?

Speech and language disorders in patients with CP±L will be assessed and diagnosed by the speech and language therapist (SLT). In some areas, two SLTs may be involved, one in the community and one specialist SLT working with the cleft and craniofacial team. During the first two years, assessment often includes checklists such as the Receptive-Expressive Emergent Language Scale (REEL-3) (Bzoch, League, & Brown, 2003) that uses reports by parents/carers to identify receptive and expressive language problems. The SLT may listen to and analyse a sample of babble or early vocalisations either in the clinic or recorded at home by parents.

Articulation can be assessed at word level using a standardized picture-naming test and detailed phonetic transcription. Pictures can be used to elicit words which contain all single speech sounds in different word positions and various consonant clusters. At sentence level, patients can be asked to read or repeat short sentences loaded with high pressure consonants (plosives, fricatives and affricates). The target sounds should be phonetically transcribed. Following this, errors should be categorized according to their Cleft Speech Characteristics. Stimulability is an important diagnostic aspect of assessment. Stimulability refers to the elicitation of a target sound or an approximate of the target sounds using auditory, visual and tactile cues. This

indicates the child's potential for speech improvement with therapy and also indicates which sounds should be targeted in therapy. Speech intelligibility is usually assessed by the SLT using perceptual ratings or by parent's perception of their child's understandability using a scale such as the Intelligibility in Context Scale (McLeod, Harrison, & McCormack, 2012).

Assessment of resonance and nasal airflow errors is usually performed by perceptual evaluation of connected speech, automatic speech (i.e. counting, days of the week) and repetition of sentences using a 4- or 5-point rating scale (e.g. 1 = normal, 2 = borderline, 3 = mild, 4 = moderate, 5 = severe). When assessing nasal airflow errors, it is important to distinguish between nasal emission/turbulence which accompanies sound production and nasal fricatives, which are articulatory errors that replace oral consonants with a devoiced nasal fricative sound.

Instrumental measurements can be used to confirm the presence of hypernasality. This can be performed using the Nasometer (Kay Elemetrics Corporation, 2010). The Nasometer is a device that contains a sound separator plate, which is placed between the nose and the upper lip. The microphones in front of the nose and the mouth collects the nasal and oral acoustic energy, respectively. These signals are sent to a computer that calculates the nasalance score. Nasalance values can vary considerably across different languages and regional dialects and hence the assessor should be aware of normative nasalance values for the child's language/dialect when interpreting results. Instrumental assessment should always be used in conjunction with perceptual evaluation.

If perceptual assessments indicate possible velopharyngeal dysfunction, a differential diagnosis can be made using direct visualization of the velopharyngeal sphincter. The two most common methods are videofluoroscopy and nasendoscopy which provide information about the structure, movement and closure/non closure of the velopharyngeal sphincter which can be video recorded (Sell & Pereira, 2011). Multiview videofluoroscopy involves low dose continuous

x-ray of the velopharyngeal sphincter during speech using different views of the sphincter. Nasendoscopy involves passing a flexible fibre-optic scope through the nose as far as the nasopharynx in order to observe the soft palate, the lateral and posterior pharyngeal walls and surrounding structures (Sell & Pereira, 2011). Results of these assessments should be reviewed by the cleft team and secondary speech surgery may be recommended for hypernasality, nasal airflow errors and passive cleft speech characteristics such as weak intraoral pressure and nasal realisations of consonants.

How can speech disorders in patients with Cleft Palate ± Lip be treated?

All children with CP±L should be seen by an SLT of the cleft team within the first year. They may be referred to local SLT services for assessment and management of communication development around 9-12 month or older depending on the service. Children can be regularly monitored and if a problem with communication (speech or language) is identified, parents should be given advice on how to help stimulate normal speech and language development. Early intervention for speech and language problems is advised in infants known to be at risk, as is the case with cleft palate. When children with cleft palate present with any of the following – delay in receptive language, delay in use of words and gestures, reduced vocalisations, a lack of plosive sounds and canonical babble by 11 months – early intervention is recommended (Hardin-Jones & Chapman, 2014). Early intervention approaches for children with CP±L can be delivered by the SLT or often it can focus on parent training/education, with the emphasis on speech sound development, phonological interventions and increase in vocabulary, using programmes that simultaneously intervene in speech and language areas (Scherer & Louw, 2011). Naturalistic approaches such as Enhanced Milieu Teaching and focused stimulation have

been found to improve sound production and vocabulary in children with cleft palate (Kaiser et al., 2017; Scherer, D'antonio, & McGahey, 2008). In young children, speech may be targeted using indirect therapy such as Multi Sensory Input Modelling (MSIM) (Harding & Bryan, 2000) and MSIM with Output (MSIM+O) (Calladine, 2009), where a high dose of models of target speech sounds are provided to the child with the aim of creating new speech motor programmes (Calladine & Vance, 2019).

As the child gets older, around 2½ or 3 years of age, they may be able to cooperate with direct therapy. If they present with language problems, these should be addressed in therapy along with speech errors. For speech difficulties, therapy should focus on errors of articulation, as hypernasality and accompanying nasal emission/turbulence are usually associated with a structural problem. Direct therapy is usually initiated when the child can cooperate for a short session and is stimulable on sound production, however, it may take several sessions to elicit a sound. Two main approaches to therapy include the traditional articulation intervention approach (Van Riper & Erickson, 1996) and a phonological approach, such as minimal pair therapy (Barlow & Geirut, 2002) or multiple opposition intervention (Boers et al., 2015; WIlliams, 2000). In the articulation approach, speech therapy will primarily focus on the correction of articulation errors by teaching the correct place and manner of articulation for the affected consonants. In order to achieve these objectives, different specific therapy techniques can be used (Kummer, 2011; Peterson-Falzone et al., 2006). Using a phonological approach, therapy targets phonological patterns rather than individual sounds (Harding-Bell & Howard, 2011). Often a combined phonological and articulatory approach to therapy is successful (Pamplona, Ysunza, & Espinosa, 1999; Sweeney et al., 2020).

When velopharyngeal insufficiency has been diagnosed by the cleft team, secondary surgical management of the palate and/or velopharyngeal sphincter is required. Such secondary speech-

improving surgery can only resolve speech disorders that are directly caused by the abnormal structure or function of the cleft (e.g. hypernasality, nasal emission, weak production of plosives or fricatives) , but active errors or disorders caused by mislearning will not spontaneously improve (Kummer, 2011).

Finally, a prosthetic approach can sometimes be considered as well; for example, in the case of absence of the soft palate or a large anatomic defect and failed primary surgical treatment (Sell, Mars, & Worrell, 2006). Furthermore, prosthetic treatment can be used to evaluate the potential advantages of surgical treatment or as a temporary solution to facilitate the treatment of compensatory articulation errors by the speech-language therapist.

What about language development and academic achievement?

A delay in measures of expressive language may be evident in young children with non-syndromic CP±L (Lancaster et al., 2019). For example, they may have a lower mean length utterance (Scherer et al., 2013) and/or smaller number of total words (Frey, Kaiser, & Scherer., 2018). This difference when compared to typically developing children appears to decrease with age (Lancaster et al., 2019). Studies looking at older children with CP+L have found language test scores within the normal range, although the child may still be facing specific language difficulties, performing below the mean or at the lower end of the normal range (e.g. Boyce et al., 2018; Klintö, Salameh, & Lohmander, 2015). These persistent language difficulties may be an underlying cause of difficulties with academic achievement (Paul, Norbury & Gosse, 2017) and there is evidence to show that children with CP±L may have difficulties with reading (e.g. Conrad, Richman, & Nopoulus, 2015), and mathematics, attributable to deficits in rapid naming and visual-spatial skills (Goodwin et al., 2017). Large-scale population studies have shown evidence

of poorer academic achievement resulting in non-attainment of school leaving certificate (Persson et al., 2012) and an increased rate of requiring a statement of educational needs, or extra help in school (Fitzsimons et al., 2018). The clinical implication of this is the need for the SLT to continue to monitor these children and in collaboration with the child's school and school teachers (Sell et al., 2021).

How can babies with unrepaired Cleft Palate ± Lip be supported during feeding?

In order to facilitate the feeding process, oral feeding facilitation techniques as well as specialized feeding equipment are often recommended (Cleft Palate Foundation, 2009; Cleft Lip & Palate Association, 2021; Miller, 2011). Below are some common methods that can help with oral feeding:

a. Positioning the infant in an upright position of at least 60 degrees. This will allow gravity to assist with posterior transfer of fluid and with swallowing. In addition, it will help prevent nasal regurgitation (i.e. liquids or food passing through the nose during drinking or eating).

b. Oral-motor techniques such as stabilization of the jaw or closing the lip with the thumb. These techniques can be used to facilitate the nutritive intake.

c. Assisted feeding. This can be used to help the child compensate for the inability to create a negative pressure in the mouth. As such, the feeder will squeeze the nipple in synchrony with the infant's sucking pattern.

d. Specialised teats and bottles. Specialized feeding equipment can be used to assist feeding. A wide variety of specialized nipples, bottles and cups is available. Overall, a nipple with a broad base is preferred in patients with a cleft lip as this may assist closure of the lip during feeding. Moreover, the size of the nipple hole needs to be adjusted depending on the infant's drinking speed. In addition, some nipples are designed to release milk by the infant's lip pressure instead of by sucking or the nipples can be squeezed by the feeder. Similarly, special bottles and cups are designed to avoid the need for active infant sucking during feeding.

Oral feeding facilitation techniques or feeding equipment are determined for each individual child by the nurse in consultation with the parents. Once the lip and palate are closed, the feeding problems will generally disappear.

Where can patients with Cleft Palate ± Lip find treatment?

Given the various interrelated problems associated with CP±L, a multidisciplinary team care approach is recommended, which typically includes a plastic and reconstructive surgeon, an orthodontist, an otorhinolaryngologist, a speech-language therapist, a maxillofacial surgeon, a social worker, a geneticist, a dentist, a psychologist, an audiologist and a paediatrician. Some cleft teams may also include a geneticist. Each specialist makes evaluations within their own discipline as well as interact closely with the other disciplines within the cleft team, in order to determine the appropriate timing and sequence of treatment, to optimize the functional, aesthetical and psychosocial outcomes, and to improve the patient's quality of life.

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