Children born with cleft palate are at a high risk for articulation disorders. Electropalato­
go phy (EPG) has emerged as a tool that utilises visual feedback to treat persistent articulation disorders in the cleft palate population. The purpose of this paper is to summarise the current research exploring the use of EPG therapy for children with surgically repaired cleft palate and inform clinicians on the quality of evidence available to guide their clinical practice. A search of the literature identified six articles appropriate for inclusion in the review. The review found that although some evidence exists for the efficacy of EPG therapy, further research should be carried out to form a more robust evidence base prior to initiation of a randomised controlled trial.

Introduction

Children born with a cleft palate are at a higher risk of speech problems than the general population (Hardin-Jones & Chapman, 2008; Peterson-Falzone, Hardin-Jones, & Karneil, 2010). Difficulties with resonance and articulation are the most common areas of speech breakdown within the heterogeneous cleft palate population (Peterson-Falzone et al., 2010). Children with cleft palate are also at an increased risk of developing negative attitudes toward communication. In their study investigating communication attitudes of 10-year-old children with cleft palate, Havstam, Sandberg, and Lohmander (2011) found a statistically significant difference between mean Communication Attitude Test (CAT-S) scores of children with cleft palate compared to their typically developing peers.

Cleft palate

Cleft palate is a craniofacial structural disorder that occurs during the seventh to twelfth week of embryonic development (Lee, Law, & Gibbon, 2006; Siren, 2004). It results from a lack of fusion of the two maxillary processes during the growth period of these structures (Shprintzen, 1995). Clefts may result in a complete cavity of the palate, creating a continuous passage between the oral and nasal cavities (Shprintzen, 1995).

There are many documented variations of cleft palate. The primary distinctions between types of cleft palate relate to unilateral or bilateral clefts with or without cleft lip (Peterson-Falzone et al., 2010; Siren, 2004). Worldwide clefts of the palate and/or lip occur in around 0.13 to 2.53 in 1000 live births every year, with substantial variation across region of birth and gender (Marazita, 2004; Reid, 2004; Wyszynski, 2007). Primary surgical repair of cleft palate is typically carried out between 12 and 18 months of age (Clark, Miesci, Mishra, Ratantje, & Rezk, 2007). Surgical intervention therefore interrupts the typical pattern of speech development at a critical stage. Palatoplasty describes the most common method of surgical repair (Peterson-Falzone et al., 2010) and involves the reconstruction of the palate via plastic surgery, often resulting in altered sensation to the palate.

Speech characteristics associated with cleft palate

A cluster of deviant speech production features are commonly associated with cleft palate (Lohmander, Henriksson, & Havstam, 2010; Michi, Yamashita, Imai, Suzuki, & Yoshida, 1993; Pamplona, Yuszua, & Espinosa, 1999; Peterson-Falzone et al., 2010). Compensatory articulation and disordered resonance are reported as the most prominent findings in the speech of the cleft palate population (Lee et al., 2009; Pamplona et al., 2005; Peterson-Falzone et al., 2010). Various authors (Dalston, 1992; Hardin-Jones & Jones, 2005; Peterson-Falzone, 1990) suggest that prevalence of compensatory articulations in children with repaired cleft palate ranges from 22% to 28% (as cited in Lee, Gibbon, Crampin, Yuen, & McLennan, 2007).

Compensatory articulations are reported to result from altered patterns of speech behaviour due to an inability to obtain adequate intracranial pressure secondary to an irregular oral cavity (Lee et al., 2009). They may also occur as a response to limited sensory feedback from oral structures. Some of the most prevalent misarticulations include posteriorly articulated alveolar stops, palatalised affricates, and palatalised sibilants (Gibbon et al., 2001; Hardin-Jones & Chapman, 2008; Lohmander et al., 2010; Michi et al., 1993; Pamplona et al., 1999; Pamplona et al., 2005). Recent developments in technology have resulted in research exploring the role of visual feedback in treatment of articulation errors (Gibbon, Stewart, Hardcastle, & Crampin, 1999).

Electropalatography

Electropalatography (EPG) is a procedure that uses visual feedback to demonstrate lingual contact on the hard palate....
using a dynamic, direct approach (Michi et al., 1993). The technique of EPG has evolved into a highly established research field in the field of speech intervention (Scobie, Wood, & Wrench, 2004). EPG therapy differs from conventional articulation therapy by providing visual feedback cues to the speaker as well as auditory feedback in the form of voice and kinaesthetic feedback from the articulators (Peterson-Falzone et al., 2010). The real-time nature of EPG permits immediate information about tongue placement and timing of articulatory movements (Gibbon et al., 2001; Gibbon & Hardcastle, 1989; Michi et al., 1993). Through identifying the specific placement of the tongue and its position in reference to the hard palate, EPG allows speakers to alter their linguo-palatal contact in order to produce phonemes with increased accuracy (Gibbon et al., 2001).

Electropalatography has also emerged as a viable tool for the remediation of articulation problems exhibited by the cleft palate population (Fujisawa, 2007; Gibbon & Hardcastle, 1989; Lee et al., 2009). Peterson-Falzone et al. (2010) suggest that the high imageability of the alveolar region of the hard palate facilitates targeting sounds that are incorrectly produced in a more back position. Moreover, its use in populations such as those with repaired cleft palate, who may have decreased oral sensation, is worthy of consideration due to the device's lack of reliance upon kinaesthetic biofeedback (Peterson-Falzone et al., 2010).

Therapy for articulation disorders in children typically involves using the speaker's auditory feedback to guide emergence of an altered pattern of articulation of any one phoneme (Pamplona et al., 1999; Peterson-Falzone et al., 2010). McAuliffe and Cornwell (2008) discussed the need to implement principles of motor learning when altering phoneme production patterns. In their research with a single subject with an articulation disorder not related to cleft palate, the authors found that incorporating EPG with therapy guided by the principles of motor learning and traditional articulation therapy resulted in positive therapy outcomes when treating lateralised /s/ (McAuliffe & Cornwell, 2008).

The limited research that has been conducted in the field of EPG has demonstrated its potential value as a method of treating persistent articulation errors in children with cleft palate when traditional methods fail (Gibbon et al., 2001). However, no large-scale studies have been conducted to support widespread clinical use of EPG with those who have a repaired cleft palate.

This paper provides a narrative review of the evidence to date that explores whether using EPG is an effective method of treatment of persistent articulation errors in children with surgically repaired cleft palate. It aims to: (a) summarise and critique the current research surrounding the most effective approaches to providing EPG therapy for treating articulation disorders in the cleft palate population and (b) inform clinicians on the quality of evidence available to guide their clinical practice.

Method

The electronic databases Medline, Ovid, EMBASE (1998–), CINAHL, SpeechBITE, Cochrane Library, and Psychinfo were searched for relevant articles. The search terms *cleft palate AND electropalatography* OR *EPG OR biofeedback AND articulation* therapy OR *speech intervention OR speech treatment AND articulation* OR *intelligibility OR speech production* produced a final yield of 13 articles after limiting results to English, excluding research on adults, and eliminating duplicates. Six articles were identified that evaluated the use of electropalatography as a speech intervention technique for children with repaired cleft palate and were therefore considered appropriate for inclusion in the review. Table 1 provides further details about the articles selected for review.

Results

**Study design evaluation**

Systematic searching of the literature revealed the majority of studies conducted in the area of electropalatographic treatment for disordered speech in the cleft palate population are classified by the National Health and Medical Research Council (NHMRC) as being low level evidence (NHMRC, 2009). The NHMRC Evidence Hierarchy is a tool used to identify the relative strength of a study according to its design and the type of research question being posed (NHMRC, 2009). The NHMRC Working Party acknowledges

<table>
<thead>
<tr>
<th>Author/s</th>
<th>Sample size</th>
<th>Title</th>
<th>Study design</th>
<th>Level of Evidence</th>
</tr>
</thead>
</table>

Note: * According to NHMRC Evidence Hierarchy. The NHMRC Evidence Hierarchy is a tool used to identify the relative strength of a study according to its design and the type of research question being posed (NHMRC, 2009).
that the hierarchy is "a broad indicator of likely bias and can be used to roughly rank individual studies within a body of evidence" (Merlin, Weston, & Tooher, 2009, p. 6). They contend that ranking individual studies should be undertaken as an initial step in appraising the evidence of any given topic (Merlin et al., 2009).

The research presented in the six studies comprises primarily small case series and single subject experimental designs. Although single subject experimental designs are considered relatively low-level evidence, they have been acknowledged as an appropriate study design when randomised controlled trials (RCT) are not suitable (Rose, 2010). According to Rose (2010), situations deemed inappropriate for the use of a RCT include when research is in the early stages of development, when the target population contains too few individuals to form a robust sample, and when the client group has a high degree of variability. Hegde (1994, as cited in Lohmander et al., 2010) concurs that single subject designs build strength of evidence for treatment strategies when repeated across different individuals. These reasons are likely explanations for such designs that dominate the EPG literature under review. Table 2 further demonstrates the variability across the six studies.

Although the six studies included in this review provide some important insights into the potential benefits of EPG to treat articulation disorders in those with a repaired cleft palate, a number of limitations exist with the nature of the studies. We contend, however, that there are some viable explanations for what appears to be a relatively low level evidence base. Further, it is valuable to examine the available evidence as a means of advancing understanding and progressing this potentially important area of clinical practice.

**Methods of therapy provision**

Stokes, Whitehill, Tsui, and Yuen (1996) based their EPG therapy on a combination of traditional methods for treating sibilants outlined by Blache (1989, as cited in Stokes et al., 1996) and conventional EPG therapy methods when conducting therapy targeting /s/ with two children with repaired cleft palate. Michi et al. (1993) utilised a similar training schedule. Each of the studies found an improvement in production of targeted phonemes using visual comparison of EPG frames. These findings suggest that EPG may play a successful role in treating persistent articulation disorders when coupled with traditional methods.

CleftNET Scotland argued that practical and financial difficulties are one of the primary factors limiting access to EPG treatment (Gibbon et al., 1998), Jones and Hardcastle (1995) developed the EPG-3, a portable training unit (PTU), in order to improve access to EPG therapy. Fujiwara (2007) found marked changes in the EPG patterns of four out of five participants when using the EPG-4. Fujiwara (2007) found delivering therapy through PTU to be especially beneficial for clients residing in remote locations.

Lohmander et al. (2010) also reported improvements in their subject's articulation of /t/ and /s/ in words and sentences following therapy conducted in the home environment using a PTU. Moreover, Lohmander et al. reported improvements to their subject's speech at word level after just 8 hours of therapy, indicating that EPG via PTU has the potential to produce rapid success.

In their randomised controlled trial, Michi et al. (1993) found participants with excessive posterior tongue elevation progressed more rapidly with EPG therapy, whereas participants with less severe misarticulations at the onset of

### Table 2. Study details

<table>
<thead>
<tr>
<th>Author</th>
<th>Cleft type/s</th>
<th>Language</th>
<th>Articulation error/s present</th>
<th>Baseline data collection</th>
<th>Treatment</th>
<th>Primary outcome measure</th>
<th>Follow-up measures</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lohmander et al. (2010)</td>
<td>Isolated soft &amp; hard palate cleft (n = 1)</td>
<td>Swedish</td>
<td>Palatalised /s/ Palatalised /n/</td>
<td>3 pre-treatment measures</td>
<td>Daily, approx. 10min/day, 5 days a week for 5 months via PTU</td>
<td>CoG values</td>
<td>3 times within 3 months</td>
</tr>
<tr>
<td>Fujiwara, Y. (2007)</td>
<td>UCLP (n = 3), BCLP (n = 2)</td>
<td>Japanese</td>
<td>Distorted /s/ Palatalised affricates</td>
<td>Not reported</td>
<td>Daily, approx. 30min/day for 7–9 months via PTU home training</td>
<td>CoG values, qualitative analysis of EPG frames</td>
<td>Not reported</td>
</tr>
<tr>
<td>Scobbie et al. (2004)</td>
<td>Isolated cleft of soft &amp; hard palate (n = 1)</td>
<td>English</td>
<td>Distorted /s/ Distorted /n/</td>
<td>Not reported</td>
<td>Ten 45 min sessions over 4 months</td>
<td>Perceptual analysis of single words or isolated phonemes</td>
<td>Not reported</td>
</tr>
<tr>
<td>Gibbon et al. (2001)</td>
<td>UCLP (n = 7), BCP (n = 2) Soft palate only (n = 3)</td>
<td>English</td>
<td>Palatalised /s/ Palatalised /n/</td>
<td>Not reported</td>
<td>Four 30 to 45 min sessions</td>
<td>CoG values, qualitative analysis of EPG frames</td>
<td>Completed once (6 weeks post-treatment)</td>
</tr>
<tr>
<td>Stokes et al. (1996)</td>
<td>UCLP (n = 2)</td>
<td>Cantonese</td>
<td>Not reported</td>
<td>2 pre-treatment measures</td>
<td>Seven weekly 1 hour sessions</td>
<td>Perceptual analysis &amp; qualitative analysis of EPG frames- construction of tongue/location</td>
<td>4 months post-therapy (1 subject only)</td>
</tr>
<tr>
<td>Michi et al. (1993)</td>
<td>UCLP (n = 3) and BCLP (n = 3)</td>
<td>Japanese</td>
<td>Palatalised /s/</td>
<td>2–4 pre-treatment measures</td>
<td>Eight weekly 1 hour sessions</td>
<td>Visual analysis of EPG frames</td>
<td>Not reported</td>
</tr>
</tbody>
</table>

Note: UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, BCP = bilateral cleft palate, PTU = portable training unit, CoG = centre of gravity.
treatment demonstrated similar progress with EPG therapy and non-EPG therapy. This finding strengthens previous research suggesting EPG therapy is most advantageous when treating articulation disorders that are not responsive to traditional methods (Lohmander et al., 2010; Fujiwara, 2007).

Therapy frequency and intensity for motor-based activities have been shown to impact treatment outcome effects when using EPG to treat articulation disorders not related to cleft palate (McAuliffe & Cornwell, 2008). However, to date, research has not examined ideal dosage of EPG therapy with particular reference to targeting typical cleft palate articulation errors. In their 2001 study, Gibbon et al. reported that when compared with non-EPG therapy, EPG therapy is "more efficient in bringing about positive change in articulation patterns" (p. 57) with only a few therapy sessions. This preliminary evidence suggests that EPG may be an efficient method of delivering articulation therapy to children with repaired cleft palate.

Discussion

This paper summarises the current research exploring the use of EPG therapy for children with surgically repaired cleft palate. Among the six studies reviewed, a significant amount of variability was found. By comparing and contrasting the findings of each study, a limited evidence base can be formed to guide clinical practice in this growing area of speech pathology treatment. The remainder of this section discusses each study's methods, findings, and conclusions in order to provide direction for future research.

Sampling

Notable disparity between cleft types, specific articulatory behaviours, and previous speech pathology intervention were evident across the sample populations of the studies being reviewed. Such variation is likely to be a consequence of subject recruitment difficulties (Lee et al., 2009). Lohmander et al. (2010) contend that the small number of children considered eligible for EPG intervention makes it challenging to obtain a significant sample size in order to conduct a study that would meet the criteria for a higher level of evidence.

Baseline data

The majority of studies did not provide adequate baseline measures of their subjects' speech prior to EPG treatment (Gibbon et al., 2001; Scobie et al., 2004; Stokes et al., 1996). For example, Gibbon et al. (2001) did not report a pre-treatment measure of articulatory accuracy. Baseline data provides stable pre-treatment production patterns in order to provide a valid account of changes produced by the treatment. Without an accurate impression of pre-treatment articulatory performance, the results may have shown fallacious improved outcomes (Portney & Watkins, 2009).

Outcome measures

The primary outcome measure for the majority of studies conducted in this field of research is correct articulation of speech sounds targeted in therapy (Lee et al., 2009). However, differences between how the researchers defined and measured correct articulation render the results somewhat incomparable.

Fujiwara's primary outcome (articulatory accuracy of /t/) was assessed using the centre of gravity (CoG) value. CoG values are obtained by calculating the "relative concentration of electrodes in the anterior-posterior dimension" on the EPG frame (Hardcastle & Gibbon, 1997, as cited in Fujiwara, 2007, p. 67). Lohmander et al. (2010) and Gibbon et al. (2001) also used CoG measures to quantitatively measure change over time.

It has been argued that the type of speech material used in the assessment of speech intelligibility may impact the reliability of results obtained (Klinto, Salameh, Svensson, & Lohmander, 2010). Klinto et al. contended that word naming is the most reliable method of assessing speech intelligibility of children with cleft palate.

A standardised articulation test for Swedish speakers (SVANTE) was implemented by Lohmander et al. (2010) in order to assess articulatory accuracy before and after treatment. Gibbon et al. (2001) also obtained speech intelligibility ratings prior to treatment. The positive relationships shown between listeners' perceptual ratings and standardised articulation test findings added strength to the authors' arguments about the validity of results obtained (Gibbon et al., 2001; Lohmander et al., 2010).

Generalisability

Children with cleft palate are a heterogeneous population. Different types of clefts, types of surgical intervention, age of repair, severity of articulation disorder, and general speech and language development all impact on the resultant speech behaviour of a child with cleft palate (Peterson-Falzone et al., 2010). Additionally, around 50% of those with a cleft palate have co-occurring syndromes. These introduce more complex factors for consideration such as presence of further craniofacial abnormalities and variable cognitive ability (Peterson-Falzone et al., 2010) when reflecting on speech treatment outcomes. It is important to note that the studies reviewed did not include children with cleft palate as part of a syndrome in their samples. As such, the combined results found are not generalisable to the entire cleft palate population (Lee et al., 2009).

Follow-up

The majority of studies investigating the use of EPG as a treatment for cleft palate speech disorders did not provide satisfactory follow-up measures for it to be deemed successful as an enduring method of treatment for articulation disorders. Without adequate follow-up, it is difficult to demonstrate that subjects will continue to show improvements from the treatment or maintain its effects, thus limiting the reliability of the study (Lee et al., 2009). For example, Gibbon et al. (2001) provided only one follow-up measure post-EPG treatment.

Special considerations

Stokes et al. (1996) provided some evidence that patterns of emergence of fricatives and affricates differ across languages. They referred to this as different "cross-linguistic routes of development" (p. 276). For example, in Cantonese there is evidence to demonstrate that children commonly affricate /s/ to /ts/ as their phonetic system develops. This is an uncommon occurrence in developing English, and suggests that phonetic development in disordered speech may be dependent on patterns of typical development in individual languages (Stokes et al., 1996). Such variations must be taken account of when considering the cleft palate population. To demonstrate, retracted articulation of palatal sounds is a universal finding among children with cleft palate (Trost, 1981; Whitehill, Stokes & Yonnie, 1996, as cited in Fujiwara, 2007). However, slight differences in
production of palato-alveolar and alveolar phonemes exist across languages and are important to acknowledge when considering treatment using EPG (McLeod & Roberts, 2005, as cited in Fujiwara, 2007). Lohmander et al. (2010) gathered EPG patterns of typically developing adult Swedish speakers prior to treatment in order to compare outcomes post-treatment to the norm for the Swedish speaking population. Comparing outcomes to native speakers’ norms of production was found to be especially important when quantitative analysis of results was performed, as subtle differences between CoG values and timing of linguo-palatal placement were not always identified by listeners’ perceptual ratings (Lohmander et al., 2010).

Conclusions and future research

Although a limited set of research exists for the potential benefits of EPG to treat articulation disorders in those with a repaired cleft palate, some promising albeit limited preliminary findings have been made about the viability of using EPG to significantly enhance the speech intelligibility of children with cleft palate (Fujiwara, 2007; Lee et al., 2009; Lohmander et al., 2010; Michi et al., 1993; Stokes et al., 1996). In particular, EPG therapy has been found to produce faster improvements to articulation errors that are resistant to conventional articulation therapy in the cleft palate population (Fujiwara, 2007; Gibbon et al., 2001; Lee et al., 2009).

The importance of collecting baseline data prior to the treatment phase in single subject experimental designs has been acknowledged (Rose, 2010). Further research should obtain stabilised pre-treatment production patterns in order to provide a valid account of changes produced by the treatment. Additionally, follow-up measurements should be obtained to ensure the changes are permanent (Lee et al., 2009).

Future research in this area should focus on the factors that may influence therapy outcomes, for example, therapy environment, intensity and duration of sessions and method of therapy provision. Prior to the initiation of a RCT, Gibbon and Paterson (2006) state that controlled group studies should be carried out to ascertain whether EPG therapy is more beneficial than the current methods of treatment for improving longstanding articulation disorders associated with cleft palate. Discovering the ideal conditions for EPG therapy would potentially allow a suitably designed RCT to be carried out in the future (Lee et al., 2009).

As different languages have slightly different norms of production of certain phonemes, all research completed should compare production patterns to that of the typically speaking population. Generalisation to contexts outside the clinic must occur in order for a meaningful improvement in communication to be achieved (Gibbon & Paterson, 2006). Further studies should assess intelligibility both at a spoken word level (Klinto et al., 2010) and in conversational settings to ensure carryover of the change in production pattern (Gibbon & Paterson, 2006).

Current clinical guidelines in the United Kingdom suggest EPG therapy is appropriate for treating articulation errors in children with cleft palate who have had little success when treated previously with conventional articulation therapy methods (National Institute of Clinical Excellence, 2002). This review found there is limited evidence for the widespread use of EPG for treatment of persistent articulation disorders associated with cleft palate at this stage. Future research should aim to increase knowledge about the factors that result in most effective treatment outcomes for the cleft palate population prior to combining these elements and conducting a large-scale randomised controlled trial.

References


Merlin, T., Weston, A., & Tooher, R. (2009). Extending an evidence hierarchy to include topics other than treatment: Revising the Australian "levels of evidence". BMC Medical Research Methodology, 9, 34.


1 Disordered resonance in cleft palate speech is not specifically addressed in this review as it is not amenable to EPG treatment. Please refer to Neumann and Romenath (2011) for a systematic review on current research relating to the use of nasopharyngoscopic biofeedback to treat velopharyngeal insufficiency in cleft palate speech.

Sarah Mainé recently completed her Master of Speech Pathology at La Trobe University, Melbourne. Tanya Serry is a lecturer at La Trobe University in Melbourne. Tanya teaches in the areas of paediatric speech and language in the Department of Human Communication Sciences. She is also engaged in research projects exploring phonological awareness interventions and collaborates on a project exploring school readiness.

Correspondence to:

Tanya Serry, PhD
Department of Human Communication Sciences
Faculty of Health Sciences
La Trobe University
Melbourne Campus, Bunjboola, Australia 3086
phone: +61 (0)3 9479 1814
email: t.serry@latrobe.edu.au

www.speechpathologyaustralia.org.au