Speech, language, and reading skills in 10-year-old children with palatal clefts: The impact of additional conditions

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ARTICLE INFO
Article history:
Received 21 January 2016
Received in revised form 19 January 2017
Accepted 6 March 2017
Available online 7 March 2017

Keywords:
Cleft lip and palate
Speech
Language
Reading
Additional condition

ABSTRACT
Background: This study examined speech (hypnasalality and intelligibility), language, and reading skills in children with a cleft palate, specifically investigating additional conditions to the cleft, in order to differentiate challenges related to a cleft only, and challenges associated with an additional condition.
Design: Cross-sectional data collected during routine assessments of speech and language in a centralised treatment setting.
Participants: Children born with cleft with palatal involvement from four birth cohorts (n = 184), aged 10.
Outcome measures: Speech: SVANTE-N; Language: Language 6–16; Reading: Word Chain Test and Reading Comprehension Test.
Results: Descriptive analyses revealed that 123 of the children had a cleft only (66.8%), while 61 children (33.2%) had a cleft that was associated with an additional condition (syndrome, developmental difficulty, attentional difficulties). Due to close associations with the outcome variables, children with specific language impairments and dyslexia were excluded from the sample (n = 14). In the total cleft sample, 33.1% had mild to severe hypernasality, and 27.9% had mild to severe intelligibility deviances. Most children with intelligibility and hypernasality scores within the normal range had a cleft without any other condition. A high number of children with developmental difficulties (63.2%) or AD/HD (45.5%) had problems with intelligibility. Hypernasality scores were also associated with developmental difficulties (58.8%), whereas most children with AD/HD had normal hypernasality scores (83.3%). As could be expected, results demonstrated that children with a cleft and an additional condition had language and reading scores below average. Children with a cleft only had language and reading scores within the normal range. Among the children with scores below average, 33.3%–44.7% had no other conditions explaining difficulties with language and reading.
Conclusions: The findings highlight the need for routine assessments of language and reading skills, in addition to assessments of speech, in children with a cleft, in order to identify potential problems as early as possible. Study designs need to take additional difficulties into account, so that potential problems with language and reading are not ascribed the cleft diagnosis, and can be followed by appropriate treatment and interventions.

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http://dx.doi.org/10.1016/j.jcomdis.2017.03.001
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1. Introduction

A young female, aged 16, was referred to the cleft team by her parents because of suspected dyslexia. The girl had just finished 10 years of elementary school. Language and reading tests had never been undertaken, despite the fact that she had been struggling with reading from the very beginning. The school had always been convinced that the reading problems were explained by the cleft and were associated with the girl’s audible speech problems. Therefore, appropriate testing and interventions had never been facilitated.

Children born with a cleft palate, with or without cleft lip, may be at risk of developing certain deviant speech characteristics, affecting resonance, articulation and intelligibility, directly caused by the original anomaly and/or related to incompetent velopharyngeal function (Klintö et al., 2014). Therefore, several studies have investigated speech characteristics in children born with cleft lip and palate (Brunnegård & Lohmander, 2007; Kuehn & Möller, 2000; Scherer, Williams, & Proctor-Williams, 2008; Stout, Hardin-Jones, & Chapman, 2011). In comparison, research that specifically explores and describes language and reading skills in this group has attracted less attention (Chapman, 2011; Hardin-Jones & Chapman, 2011), and produced conflicting results (Kuehn & Möller, 2000). Given the importance of competency with spoken and written language for a child’s success both in school and beyond (Chapman, 2011), there is a need for a better understanding of language and reading skills in children with a cleft.

It is well known that a significant number of children with a cleft may have additional conditions and/or syndromes (Christensen & Mortensen, 2002; Swansenborg de Veye, Beemer, Mellenbergh, Wolters, & Heineman-de Boer, 2003). More specifically, cognitive dysfunction and learning difficulties ranging from mild to severe impairments have been demonstrated (Milerad, Larson, Hagberg, & Idenberg, 1997; Richman, McCoy, Conrad, & Nopoulos, 2012). If the existence of impairments additional to the cleft are not accurately reported or controlled for within statistical analyses, research in this area is unlikely to provide an accurate picture of language and reading outcomes in children born with CL/P. Consequently, and due to the expected impact of such associated conditions on language and reading skills (Bishop, 2009; Kuehn & Möller, 2000), the presence of other conditions additional to the cleft need to be carefully recorded and taken into account in a study design (Feragen, Stock, & Rumsey, 2014; Maarse et al., 2012). While several studies have described the risk of specific conditions affecting cognitive development, and language and/or reading skills in cleft samples (for a review, see Richman et al., 2012), to our knowledge, few studies have included complete birth cohorts of children born with a cleft, and explored how the presence of all additional conditions affect measures of speech, language and reading. Further, a differentiation between challenges related to the cleft, and those associated with an additional condition that could affect language and reading skills, would have the potential to improve significantly the quality of information that is given to parents regarding their child’s future development.

1.2. Speech, language and reading in children with a cleft

Some research from the general population report that children with persisting speech problems appear to be at higher risk for delays in the acquisition of reading skills (Bishop & Snowling, 2004; Lewis et al., 2015; Peterson, Pennington, & Shriberg, 2009), findings which could put children with a cleft having speech problems, at risk (Hardin-Jones & Chapman, 2011). The impact of delayed or deviant speech on phonological development in children with a cleft, has also been highlighted (Bessell et al., 2013; Willadsen, 2013), in addition to the importance of language skills for the acquisition of early reading skills (Chapman, 2011). In addition, deficits in rapid naming and visual memory, a problem which is known to be a predictor of reading disabilities, have also been demonstrated in cleft samples (Conrad, McCoy, DeVolder, Richman, & Nopoulos, 2014; Richman, Ryan, Wilgenbusch, & Millard, 2004). Evidence suggesting that language-based interventions might improve speech production (Scherer et al., 2008) further illustrates the many associations between speech and language characteristics, and hence the importance of research including these variables. The overall assumption may understandably lead to questions regarding whether children with a cleft are at higher risk for language and reading difficulties (Chapman, 2011; Collett, Leroux, & Speltz, 2010).

Several studies carried out during the 1950s-1970s explored language skills and reported delayed language development in children born with cleft (for a review, see Kuehn & Möller, 2000), a perspective that has gained increased interest and investigation during the last decade. However, the studies report contrasting results. Delays in language development in children with a cleft have been reported (Jocelyn, Penko, & Rode, 1996), while other studies indicate no differences with control groups in terms of their language skills (Chapman, 2011; Collett et al., 2010). Yet another study reported receptive language skills within the normal range, but with varying expressive skills, with some children demonstrating delays and others showing typical language development (Hardin-Jones & Chapman, 2011).

Inconsistent findings may partially be explained by methodological issues, such as a wide range of study designs, difficulties accessing representative and large samples, different approaches to the choice of exclusion criteria, and considerable variation in the choice of measures. Another consideration and possible explanation for conflicting findings could be the presence of mediating factors such as cognitive function, or other potentially influential background variables such as hearing difficulties, cleft type, surgical correction of hypernasal speech (secondary surgery), or gender (Casby, 2001; Kuehn & Möller, 2000; Neumann & Romonath, 2012; Roberts, Mathias, & Wheaton, 2012). Therefore, studies including and
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