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The association of cleft severity and cleft palate repair technique on hearing outcomes in children in northern Finland

Riitta H. Lithovius [a], Ville Lehtonen [a, b], Timo J. Autio [c], Virpi Harila [d, e], Vuokko Anttonen [b, e], George K. Sándor [a, e, *], Leena P. Ylikontiola [a, e]

[a] Department of Oral and Maxillofacial Surgery, Faculty of Medicine, University of Oulu, Oulu University Hospital, Oulu, Finland
[b] Department of Cariology, Paedodontics and Endodontology, University of Oulu, Oulu, Finland
[c] Department of Otolaryngology, Faculty of Medicine, University of Oulu, Oulu University Hospital, Oulu, Finland
[d] Department of Oral Development and Orthodontics, University of Oulu, Oulu, Finland
[e] Medical Research Center Oulu, Oulu University Hospital and University of Oulu, Finland

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ABSTRACT

Background: The consequences of cleft lip and palate include scaring, dental malformations, tooth misalignment, speech problems, and hearing loss. Otitis media with effusion causing hearing loss is a problem for many cleft palate patients.
Methods: This study examines the association among cleft severity, palate repair technique, and hearing outcomes in children from northern Finland with clefts, aged 3–9 years. The study included 90 cleft patients who were treated at the Oulu University Hospital Cleft Lip and Palate Center between 1998 and 2011. The severity of the cleft, the surgical technique used to repair the palate, audiogram configuration data, and the need for ventilation tube placement were determined retrospectively from patient records.
Results: Only 3.3% of cleft patients had an abnormal pure tone average hearing threshold representing abnormal hearing. Neither the surgical technique used to repair the cleft palate nor the severity of the cleft was a significant factor related to hearing loss or to the number of ventilation tubes required. Hearing improved significantly with increasing age over a span of 6 years.
Conclusions: Continuous follow-up with proactive placement of ventilation tubes before or at the time of palatoplasty results in hearing outcomes in cleft children that are similar to those reported in non-cleft children.

1. Introduction

Cleft lip and palate, together with cleft palate, are the most common congenital craniofacial birth defects (Harila et al., 2013; Esmail et al., 2014). The sequelae of orofacial clefts are far reaching (Härtel et al., 1991) and may include facial scarring, nasal deformity, dental malformations, tooth misalignment, speech difficulties, and hearing loss. Practitioners who treat patients with cleft lip and palate should not only be aware of the dental-related difficulties that patients with clefts face, but should also be knowledgeable about the possible hearing loss that these patients may sustain.

Otitis media with effusion (OME) is defined as a collection of fluid in the middle ear space with an intact tympanic membrane that persists for 3 months or longer and can lead to hearing loss (Kuo et al., 2014; Gani et al., 2012). OME is a very common finding in children who are born with cleft lip and palate (CLP) and cleft palate (CP), with studies reporting prevalence rates of at least 90% (Stool and Randall, 1967; Grant et al., 1988). The pathogenesis of OME in patients with CP is multifactorial including displaced palatal musculature (Kriens, 1969, 1970), the curvature of the Eustachian tube lumen and the density of Eustachian tube cartilage (Bluestone, 2009). The palatal muscles, including tensor veli palatini and levator veli palatini, are responsible for the opening of the Eustachian tube, and their displacement by the presence of a palatal cleft results in Eustachian tube dysfunction (Bütow et al., 1991). This dysfunction leads to poor middle ear ventilation and negative pressure, and results in a retracted tympanic membrane and

* Corresponding author. Institute of Dentistry, Aapistie 5, 2126B, 90014 University of Oulu, Finland. Tel.: +358 504 280 906; fax: +358 8 5375560.
E-mail address: george.sandor@oulu.fi (G.K. Sándor).

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mucous secretion (Broen et al., 1996). The consequence of an unusually high incidence of isolated CP patients in northern Finland (Harila et al., 2013) in relation to Eustachian tube dysfunction or hearing outcome is not known.

Several studies have shown the link between hearing loss and its association with OME and CP (Bluestone, 1971; Bennet, 1972; Paradise, 1975). Long-term hearing loss has been shown to have a negative influence on speech development (Schönweiler et al., 1999). Although ventilation tube (VT) insertions have been reported to prevent short-term hearing loss and to improve speech and language development in CP patients (Kuo et al., 2014; Gani et al., 2012; Ponduri et al., 2009; Shaw et al., 2003), the use of prophylactic VT insertions can be associated with complications such as eardrum atrophy, perforations, and otorrhea (Kuo et al., 2014; Ponduri et al., 2009). Other studies have shown an association between multiple VT insertions and an increased risk of conductive hearing loss in the long-term (Sheahan et al., 2002).

Although primary palatoplasty can slightly improve the ventilatory function of the Eustachian tube (Kriens, 1969, 1970; Bütow et al., 1991), the function of the repaired palatal musculature may never normalize, and OME may remain a common problem following palatal repair (Dhillon, 1988; Matsune et al., 1991; Robinson et al., 1992; Nunn et al., 1995). However, it has also been observed that after the adolescent growth phase, Eustachian tube function improves and the incidence of OME greatly decreases (Bluestone et al., 1975; Sheahan et al., 2003).

The audiological assessment of cleft palate patients continues from infancy to adulthood. It begins with the screening of newborns. Regular audiology appointments follow so that any unexpected hearing outcome will be noticed and treated as needed (Shaw et al., 2001; Kuo et al., 2013). The most common type of hearing loss in children with CLP and CP is conductive hearing loss resulting from OME (Kuo et al., 2013).

Although the link between hearing loss and its association with OME and CP has been studied, the effect of cleft severity, its repair, and the associated placement of VT are not known with regard to hearing outcomes. Moreover, since clefts in northern Finland are associated with an unusually high rate of isolated CP (Harila et al., 2013), the effect of such an unusual preponderance on hearing outcome is important. The aim of this study was to examine the associations among cleft severity, palate repair technique, VT placement, and hearing outcomes in children with CLP and CP in a population of children from northern Finland who were between 3 and 9 years of age.

2. Material and methods

This retrospective study included 214 consecutive cleft patients who were registered between 1998 and 2011. The study was approved as a retrospective study by the ethical committee of the Oulu University Hospital (permission number 10/2012), and the data were treated according to the principles of the Declaration of Helsinki. Only patients who had been treated at the Oulu University Hospital Cleft Lip and Palate Center were included in this study. The patients' charts were accessed through the hospital's medical database. The following data were collected: sex, date of birth, type of cleft, surgical technique used for primary repair, date of primary palatoplasty, number of VT placements, hearing test results with pure tone averages (PTA) and the dates of the audiological measures.

Exclusion criteria were as follows: patients who had been treated elsewhere, patients lost to follow-up, patients less than 3 years of age, patients without complete hearing examination records, and mentally disabled patients who were unable to complete the hearing examination. Since the investigators intended to study cleft severity using three-dimensional scanning of dental casts of the affected palates, patients with isolated cleft lip or submucous cleft palate were also excluded from the study, as the nature of these deformities precludes their assignment to a cleft severity scale category. Of the 214 cleft patients who were initially screened for the study, 90 patients who passed the exclusion criteria were enrolled.

Children from 3 to 9 years of age were included in this study and were divided into two groups: those aged 3–5 years and those aged 6–9 years. There were 42 patients who had hearing examinations at both the 3- to 5-year and 6- to 9-year time points. The best hearing result at each time point was recorded for each group and analyzed. The severity of palatal cleft was determined from three-dimensional scanned cleft models using the scale proposed by Jensen et al. (1988) (Table 1).

Patients were grouped as follows: cleft palate (CP), or cleft lip and palate (CLP). In all patients with cleft palate, a one-stage palatal, three-layer closure was performed, in which the hard and soft palates were closed with an intravelar veloplasty (Kriens, 1969, 1970). The ventilation tubes were inserted proactively before or at the time of palatoplasty, as children often develop painful OME following palatoplasty. Patients were followed up by an otolaryngologist one to three times per year, or more frequently if necessary. Tubes were replaced according to the Oulu University Hospital protocol, which included the diagnosis of glue ear or recurrent otitis media with at least three infections over a 6-month period, or more than four infections in a year.

The hearing protocol included audiological examination using standard pure tone audiometry. Ear-specific measurements were performed at 0.5 kHz, 1 kHz, 2 kHz, and 4 kHz and registered in dB hearing levels by a trained audiologist using headphones. The three-frequency PTA was calculated for each ear. Some of the audiograms were not in electronic format, and the PTAs were calculated with the guidance of an otolaryngologist. On the PTA scale, higher numbers represent poorer hearing; a PTA score greater than 20 dB was categorized as the abnormal hearing threshold (Martini, 1996).

2.1. Statistical analysis

Means and standard deviations were used to describe the continuous variables. Frequency was used to describe categorical data such as sex and cleft type. A paired-samples *t*-test and the Wilcoxon signed rank test were used to compare hearing scores between the time windows for the 3- to 5-year-old and the 6- to 9-year-old patients. The Kruskal–Wallis test was used when comparing PTAs to the palatoplasty surgical technique and to the severity of cleft and the cleft type. The Mann–Whitney test was used when determining the association between cleft severity or cleft type and PTAs. Medians and ranges were used with these tests. Analysis of variance (ANOVA) was used to compare the number of tube insertions by surgical technique. The Mann–Whitney test was used when analyzing the association between cleft severity and the number of tube placements. Differences with $p < 0.05$ were considered to be statistically significant. The data were analyzed using IBM SPSS statistics 20.0 software for Windows (IBM SPSS, Chicago, IL, USA).

Table 1
Severity of the cleft palate according to Jensen et al. (1988).

Grade	Severity of cleft palate
1	Soft palate
2	One-third of hard palate
3	More than one-third up to subtotal
4	Total