International Journal of Pediatric Otorhinolaryngology 94 (2017) 76–81

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: http://www.ijporlonline.com/

Hearing sensitivity in adults with a unilateral cleft lip and palate after two-stage palatoplasty

I.F.P.M. Kappen [a, *], J.B.S. Schreinemakers [b], K.P.Q. Oomen [b], D. Bittermann [c], M. Kon [a], C.C. Breugem [a], A.B. Mink van der Molen [a]

[a] Department of Plastic Surgery, University Medical Centre Utrecht, P.O. Box 85090, 3508 AB, Utrecht, Utrecht, The Netherlands
[b] Department of ENT Surgery, University Medical Centre Utrecht, P.O. Box 85090, 3508 AB, Utrecht, Utrecht, The Netherlands
[c] Department of Maxillofacial Surgery, University Medical Centre Utrecht, P.O. Box 85090, 3508 AB, Utrecht, Utrecht, The Netherlands

ARTICLE INFO

Article history:
Received 14 October 2016
Received in revised form
24 December 2016
Accepted 24 December 2016
Available online 26 December 2016

Keywords:
Adult
Cleft palate
Cleft lip
Hearing loss
Audiometry
Tympanometry

ABSTRACT

Objective: To evaluate long-term hearing and middle ear status in patients treated for a unilateral complete cleft lip and palate (UCLP) by two-stage palatoplasty.
Methods: Forty-nine UCLP patients aged 17 years and older were included in this retrospective study. Patients were invited for a multidisciplinary long-term follow-up of their treatment at a tertiary center for craniofacial surgery in the Netherlands. ENT assessment included tympanometry and pure-tone audiometry. Medical files were searched for medical and surgical history.
Results: In total, 19.4% of the patients had significant long-term hearing loss (PTA > 20 dB), comprising conductive hearing loss in 21.5% of the patients. In the majority (70%), this hearing loss was more pronounced at higher frequencies. In 25% the high fletcher index showed hearing thresholds above 20 dB. Ventilation tubes were placed at least once in 78.7% of the patients. The frequency of tube insertion was positively correlated with the incidence of reduced tympanic compliance (tympanogram type B) and the need for a pharyngoplasty.
Conclusion: The present study reports long-term hearing outcomes in UCLP patients with hard palate closure at 3 years of age. Persistent hearing loss was observed in 19.4% of our patients (PTA > 20 dB), mainly at the higher frequencies. Ventilation tube placement was associated with reduced tympanic compliance and higher risk on hearing loss (PTA air conduction). Our high incidence of velopharyngeal incompetence during early childhood, which is likely associated with Eustachian tube dysfunction, might have contributed to these results.

1. Introduction

Although cleft lip and palate is one of the most common encountered congenital craniofacial anomalies worldwide, surgical treatment varies greatly amongst centers. The main objectives of cleft lip/palate treatment are to achieve unrestrained maxillofacial growth, normal speech and hearing in combination with a good aesthetic outcome. The timing of cleft palate closure is however challenging, as it is often a compromise between adequate speech development, mid-facial growth and it possibly influences middle ear status and hearing outcome. Children with a unilateral cleft lip and palate (UCLP) commonly suffer conductive hearing loss (HL) due to a high occurrence of otitis media with effusion (OME) [1–6]. This HL may in turn affect speech and language learning [7].

The presence of chronic OME in patients with a cleft is thought to be caused by malfunction of the Eustachian tube [2,8–11]. The cause for this malfunction is multifactorial, as both the aberrant morphology [11,12] and the detrimental effects of palatal surgery [9,10,13] likely contribute. The Eustachian tube in cleft palate patients shows anatomical [9,14–16], morphological [15] and histological [11,13,17] differences compared to the general population. Furthermore, an abnormal insertion of the levator veli palatini muscle into the edge of the hard palate [9,15,16] in combination with an abnormal origin of the tensor veli palatini muscle at the Eustachian tube [14] changes the vector at which the tube is pulled open. All these factors contribute to decreased and often inadequate middle ear ventilation.

* Corresponding author. Dijk 30, 5521AZ, Eersel, The Netherlands.
E-mail address: isabellekappen@me.com (I.F.P.M. Kappen).

http://dx.doi.org/10.1016/j.ijporl.2016.12.030
0165-5876/

The peak incidence of middle ear problems in cleft patients appears to occur around 4–6 years, with a decrease after the age of 12 years [18]. Although the incidence of HL seems to decrease accordingly [19], in a significant portion of the UCLP patients the hearing loss persists into adulthood [2,19]. In the general population, OME episodes physiologically decrease [20] due to regression of the adenoids and normal craniofacial growth resulting in elongation of the Eustachian tube, change of the muscle vectors and an increase in the tensor veli palatine surface area [21]. In cleft patients, the surgical repair of the cleft and anatomic restoration of the velopharyngeal apparatus might therefore contribute to the improvement of middle ear ventilation. Timing of cleft surgery is therefore possibly of influence on middle ear complications and long-term hearing outcomes.

In contrast to mid-facial growth and speech outcome, hearing sensitivity remains an underreported outcome measure in the literature. Studies assessing long-term hearing and middle ear status in UCLP adults are scarce [22,23]. Furthermore, the current literature investigating the influence of the timing of palatal closure on middle ear status is inconsistent. In the Wilhelmina Children's Hospital (WCH), two-stage palate closure has been performed for nearly thirty years. This study aims to evaluate long-term hearing sensitivity and middle ear status in UCLP adults treated according to the Utrecht protocol, which includes a two-stage palatoplasty with hard palate closure at 3 years of age.

2. Methods

2.1. Patients

Patients were recruited from the cleft lip and palate database at the Utrecht University's Wilhelmina Children's Hospital (WCH). All cleft patients of 17 years and older were invited for a long-term follow-up. Medical records of all invited patients were retrospectively reviewed for their surgical and medical history. Only the patients with an isolated UCLP treated by two-stage palatoplasty performed by the plastic surgeons at our center were selected for the present study. Patients with syndromal clefts, additional anomalies or incomplete surgical data regarding the timing of soft and/or hard palate closure were excluded (n = 10).

From the 78 patients that met our inclusion criteria eventually 52 attended follow-up (67%). On the day of follow-up, patients were seen by each specialist from the cleft team, including an audiologist and an otolaryngologist. For three patients, no audiograms were obtained during follow-up (4%). Of the patients that did not attend follow-up (n = 26), nine were not interested in follow-up or could not attend the appointment. The remaining patients were lost to follow-up due to incorrect contact details or non-response (n = 17, 22%).

Patient characteristics are described in Table 1. The surgical protocol of the patients that attended follow-up was compared with patients that did not attend (Table 1).

2.2. Surgical protocol

The treatment protocol is described in Table 2. Patients received ventilation tubes in case of persistent OME with an inadequate improvement after three months. Ventilation tubes are preferably placed after the age of nine months. In 76.6% of our patients, ventilation tubes had been inserted during childhood (n = 72, 94 ears). The number of tube insertions per ear varied from 0 to 5 times, with a median of 2 times (IQR 1 - 2). Out of all the ventilation tubes placed, 32% were T-drains.

Patients requiring additional surgical interventions represented 16% (n = 9) including a mastoidectomy (n = 1), BAHA insertion (left ear) (n = 1), myringoplasty (n = 7), or attico-antrostomy (n = 1) due to a cholesteatoma (2%). One patient showed a notch at 2000 Hz suspect for otosclerosis (2%). A pharyngoplasty was performed in 43% of the patients (n = 21).

2.3. Hearing sensitivity

Hearing levels were assessed by pure tone audiometry during long-term follow-up. Tests were performed using a Decos audiometer in a soundproof room. For each ear an audiogram was obtained which described the air and bone conduction thresholds at 500, 1000, 2000, 4000 and 8000 Hz. The fletcher index was given for both bone and air conduction at higher (1000–4000 Hz, high fletcher index) and lower frequencies (500–2000 Hz, pure tone average (PTA)). The mean hearing threshold was defined as the mean of the 500, 1000, 2000 and 4000 Hz thresholds, to allow for comparison with previous reports. Mean hearing threshold levels (HTL) of 0–20 dB were considered as normal. A mean HTL of between 21 and 40 dB were defined as mild hearing loss, higher than 40 dB as severe hearing loss [4,23,24].

2.4. Tympanometry

Tympanometry was performed by an audiologist using an Interacoutics® impedance audiometer AT235h with a 226-Hz probe. The compliance of the eardrum, the peak pressure and the ear canal volume was measured. Each diagram was scored by two independent examiners on subtype (A, Ad, As, B, C). In the case of disagreement, consensus was reached by discussion. Subtype A was regarded as normal, while subtype Ad, As, B and C were regarded as abnormal.

2.5. Statistical analysis

The data was analysed using IBM SPSS Statistics 20 (Statistical Package for the Social Sciences, SPSS Inc., Chicago, IL, USA). Mean values were calculated where relevant. The chi-square test or the

Table 1
Baseline table and comparison of groups.

Baseline and treatment characteristics of patient groups				
Baseline and treatment characteristics		Included for analysis n = 49	Did not attend follow-up[a] n = 26	p-value[b]
Gender	Male	35 (73.5%)	18 (69.2%)	0.722
Lip closure	Mean age in mo.	5.6	6.6	0.108
Soft palate closure	Mean age in mo.	7.9	6.9	0.462
Hard palate closure	Mean age in mo.	39	47	0.258
Pharyngoplasty	Total performed (%)	21 (43%)	6 (23%)	0.051
Fistulas		13 (27%)	5 (19%)	0.452

mo. = months, yrs. = years.
[a] 7 patients were excluded from analysis as surgical data could not be retrieved.
[b] p-value <.05 was regarded as significant.