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Internationally adopted children with cleft lip and/or cleft palate: Middle ear findings and hearing during childhood☆

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ABSTRACT

Objective: Adopted children with cleft lip and/or cleft palate form a diverse group of patients. Due to increased age at palatal repair, adopted children have a higher risk of velopharyngeal insuffiency and poor speech outcome. Delayed palate repair may also lead to longer lasting Eustachian tube dysfunction. Decreased function of the Eustachian tube causes otitis media with effusion and recurrent acute otitis media, which can lead to other middle ear problems and hearing loss.

Methods: One-hundred-and-thirty-two adopted children treated by the Cleft palate team in Wilhelmina Children's Hospital during January 1994 and December 2014 were included. Retrospectively, middle ear findings, the need for ventilation tube insertion and hearing during childhood were assessed. Findings were compared with 132 locally born children with cleft lip and/or cleft palate.

Results: Adopted children had a mean age of 26.5 months old when they arrived in our country. After the age of two the total number of otitis media with effusion episodes and the need for ventilation tube placement did not significantly differ among adopted and non-adopted children. Adopted children had significantly more tympanic membrane perforations. Hearing threshold levels normalized with increasing age. Although within normal range, adopted children showed significantly higher pure tone averages than locally born children when they were eight to ten years old.

Conclusion: In general, adopted patients with cleft lip and/or cleft palate did not have more middle ear problems or ventilation tubes during childhood. However, theyhave more tympanic membrane perforations.

1. Introduction

Overall adoption numbers in the Netherlands have declined over the past years, but the number of adopted children with 'special needs' has increased [1]. 'Special needs' children are children with congenital malformations, such as cleft lip with or without cleft palate and congenital hand and feet defects [1,2]. Of all 214 internationally adopted children in the Netherlands in 2016 approximately 80% had a medical condition or birth defect. Cleft lip and/or cleft palate (CLP) is common [1]. Consequently our cleft palate team is confronted frequently with the care for these children. Cleft care for internationally adopted children with CLP varies between institutions in the Netherlands due to insufficient evidence to guide treatment. Institutions that described their patient characteristics and initial care and treatment demonstrate a very diverse group of patients who often have velopharyngeal insuffiency and poor speech outcome [2–7]. Different factors are thought to contribute such as a) the new child-family relationship, b) palate and lip repair at an older age with more risks of complications [8,9], c) the variety and quality of former treatments [2–6] with possibly re-do surgery and d) having to learn a new language. Furthermore, children with CLP often suffer from conductive hearing loss due to otitis media with effusion (OME) and recurrent acute otitis media (AOM) [10–13]. Repair of the cleft palate should lead to a more normal Eustachian tube function and thus to a decrease of OME [14–17]. Since palatoplasty in adopted children with CLP commonly takes place at an older age, they have longer exposure to OME.

Current Dutch guidelines recommend insertion of ventilation tubes in children with recurrent acute otitis media, defined as three episodes of AOM per six months or four episodes per year, and in children with persistent otitis media with effusion defined as, OME persisting for more than 3 months with a conductive hearing loss of 25 dB or more [20–25]. Nevertheless, the benefits of ventilation tubes are still subject of debate as in some cases the placement of ventilation tubes itself leads to complications, such as tympanic membrane perforations and

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otorrhea [18,26]. Otorrhea particularly becomes a problem when it is caused by resistant organisms, such as Methicillin-resistant *Staphylococcus aureus* (MRSA). [27] Internationally adopted children are often adopted from countries with a high MRSA carrier rate, they have an increased risk of MRSA-colonization and therefore an increased risk of MRSA otorrhea [2,28–30]. The precise effect of MRSA on the rate of postoperative infections and complications after palate repair and ventilation tube insertion is unknown [2]. However, in our institution otolaryngologists are reluctant to place ventilation tubes in children carrying MRSA because of the fear for chronic otorrhea and the subsequent inability to eradicate MRSA.

Although some studies have been published about speech assessment [2–7] in the adopted cleft palate patients, none have investigated middle ear and hearing problems in this group. Therefore, the aim of this study was to describe characteristics of adopted children with CLP and compare middle ear findings and hearing in adopted children with CLP with locally born children with CLP during childhood in the Netherlands.

2. Patients and methods

2.1. Study population

A retrospective study including all internationally adopted children with CLP presenting to the cleft palate team outpatient clinic in the Wilhelmina Children's Hospital Utrecht between January 1994 and December 2014 was performed. One-hundred-and-two patients were identified.

To make comparison possible, we included a control group consisting of non-adopted CLP children also treated by the cleft palate team in Wilhelmina Children's hospital. To improve the efficiency of our analysis we applied *frequency matching* to select our controls. Locally born controls were matched for gender and cleft type with the adopted group. These two characteristics were chosen for matching because the distribution of both gender and cleft type in our adopted group differs from what is to be expected in the general population. In the general cleft population 50% of the population suffers from cleft lip and palate, 30% has a cleft palate and 20% has cleft lip [31].

As a result of frequency matching 132 non-adopted children born between January 1997 and December 2011 were randomly selected. Medical records of all included patients were reviewed, information concerning surgical and medical history were obtained. Study data were collected and managed in a database.

The cleft palate team encompasses various specialties including pediatrics, plastic surgery, otolaryngology, audiology, speech and language therapy, dentistry and maxillo-facial surgery, leading to multidisciplinary treatment. Although internationally there are still many unresolved questions about timing in cleft palate closure, our cleft palate team did use a standard schedule with minimal consultations for locally born children (see Table A in the appendix). However, many patients consulted specific specialists more frequently. For adopted children this schedule was often not applicable, since they arrived in the Netherlands at an older age. For these children treatment often started with a consultation by a general pediatrician for a thorough medical history and physical examination. If the MRSA status was unknown, tests were done. When a patient tested positive, attempts to eradicate MRSA were often postponed until after the palatal closure was performed, as is advised in our hospital protocol [32]. In our adoption group 36 children (27%) tested MRSA positive when they first arrived. After examination by the general pediatrician, a referral to the cleft team followed. Depending on the treatment the adopted children underwent in their country of origin and the MRSA status the adopted children saw various specialists at their initial presentation to the team. The plastic surgeon was consulted to see whether (re-do) surgery was necessary. An otolaryngologist examined the children to determine if myringotomy or ventilation tube insertion was required.

2.2. Palatoplasty

During study period different plastic surgeons performed palatoplasty. Surgical techniques used in adopted children who had palatoplasty in their native country were unknown. Adopted and non-adopted children that underwent palatoplasty in our hospital before the year of 2007 (30%), were likely to have a palatoplasty according to Furlow's technique with double opposing Z-plasty [62]. Children that underwent surgery after the year of 2007 (70%) underwent an intravelar veloplasty under a microscope following Sommerlad's principles [63].

2.3. Otolaryngology

Every consultation to the otolaryngologist ideally consisted of otoscopic examination, tympanometry and audiological examination. However, due to MRSA in the adopted group and uncooperativeness of patients not all children underwent all these examinations and tests during every visit.

2.3.1. Tympanometry

Tympanometry was performed by an audiologist using an Interacoutics® impedance audiometer AT235h with a 226-Hz probe. Each tympanogram was scored according to the Jerger classification into either subtype A, B, C or E and judged by the otolaryngologist as either normal or abnormal. Subtype A was considered to be normal, whereas subtypes B, C and E were considered abnormal [61].

2.3.2. Audiometry

Pure tone audiometry was performed with Decos audiometer in a soundproof room. Ear specific measurements were obtained at 250 Hz, 500 Hz, 1000 Hz, 2000 Hz and 4000 Hz and registered in dB hearing level by trained audiologists using headphones. In younger children, aged six months to thirty months, behavioral observational audiometry was used to assess hearing. To compare different audiometry results, pure tone averages were calculated. Hearing was considered normal with mean threshold levels of 0–20 dB. Mild hearing loss was defined as mean hearing threshold levels between 21 and 40dB. If thresholds were higher than 40 dB it was considered as severe hearing loss.

2.4. Statistical analyses

For statistical analysis SPSS version 22.0 was used. Demographic and surgical data were analyzed with descriptive statistical methods. Normally distributed data outcomes were compared using chi-square test for categorical and dichotomous data and *t*-test for continuous data. Results were presented as percentages in categorical and dichotomous data and as means with standard deviation in continuous data. To compare outcomes in not normally distributed data a Mann-Whitney test was used. Results were presented as median with a range. Correlations between different variables were calculated using Pearson's correlation test for normally distributed and continuous data and Spearman's correlation test for not normally distributed data and for categorical and dichotomous data. Data concerning OME episodes, ventilation tube insertion and hearing loss were analyzed in total and for different age groups; zero to two years old, two to four years old, four to six years old, six to eight years old, eight to ten years old and older than ten years old. Negative binomial regression analysis was used to find predictors in count data. Linear regression analysis was used to find predictors in continuous data. A *p*-value of less than 0.05 was considered statistically significant.

2.5. Ethics

The medical ethical review board of our institution approved this study (16–656). The requirement for informed consent was waived.