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Main Article

Ear, nose and voice problems in children with orofacial clefts

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Abstract

The purpose of this study was to compare the prevalence of ear, nose and particularly voice problems in groups of children with cleft palate (CP) and with unilateral cleft lip, alveolus and palate (UCLP). On the basis of history, regular otorhinolaryngological examinations and hearing tests, the prevalence of different pathologies was assessed in 80 CP children (35 boys and 45 girls) and 73 UCLP children (47 boys and 26 girls). Ear pathology was reported in 53.8 per cent of CP children and in 58.9 per cent of UCLP children. Nasal breathing was impaired in 14 CP (17.5 per cent) and 36 UCLP (49.3 per cent) children. Dysphonia was detected in 12.5 per cent of CP and 12.3 per cent of UCLP children. In 9.2 per cent of all cleft children, functional voice disorder caused a hoarse voice. Two-thirds of cleft children with functional dysphonia had protracted hearing loss. Therefore, ENT specialists must take an active role early in the treatment of children with clefts.

Key words: Cleft Lip; Cleft Palate; Otitis Media With Effusion; Nasal Obstruction; Voice Disorders

Introduction

Problems with the ears, nose or throat (ENT) appear in almost every child with a cleft of the secondary palate. The majority of children with cleft palate (CP) or cleft lip, alveolus and palate (CLP) have recurrent acute otitis media (ROM) or secretory otitis media (SOM).[1] The incidence of SOM in infants with CLP was reported to range from 40 to 100 per cent.[2–5] Duroux *et al*.[6] initially found SOM in 87 per cent of children with CP; three years after surgical treatment of the CP, SOM persisted in 70 per cent of cases.

Godbersen[7] stated that regular ENT examinations are necessary in children with CLP; otherwise, middle-ear disease is overlooked in almost half of cases. Fluctuating mild-to-moderate conductive hearing loss in early childhood that is associated with SOM can result in impaired speech, language and even cognitive development.[8–11]

Nasal breathing problems are typical in the CLP population; CLP frequently produces nasal deformities that tend to reduce the size of the nasal airway. Approximately 70 per cent of the CLP population has nasal-airway impairment and about 80 per cent breathe orally to some extent. Patients with unilateral CLP (UCLP) have the smallest nasal cross-sectional area.[12]

Cleft palate or CLP can also affect a child's voice and articulation. It has been reported that individuals with CP have a high prevalence of laryngeal voice symptoms such as hoarseness, breathiness, low volume and abnormal pitch. Brooks and Shelton[13] found the incidence of hoarseness to be 10 per cent in a population of 76 CP subjects. McDonald and Baker[14] pointed out that faulty phonation could result in hyperaemia, hyperplasia of the vocal fold covering and hoarseness in adults with CP. Among 154 patients with CLP, Bressman *et al*.[15] found severe voice disorders in 6.5 per cent and moderate voice disorders in 25.3 per cent. D'Antonio *et al*.[16] detected a 41 per cent prevalence of aurally perceived voice symptoms, observable laryngeal abnormalities, or both, in a group of 85 cleft and non-cleft patients with velopharyngeal dysfunction. A report from Denmark showed that more than half of patients who presented with CP and velopharyngeal insufficiency exhibited dysphonia.[17] On the other hand, Takagi *et al*.[18] found an incidence of voice problems other than nasality of only 0.6 per cent.

Investigators have speculated that the prevalence of voice disorders in the cleft population is related to abnormal velopharyngeal valving. McWilliams *et al*.[19] reported on 43 children with CP and hoarseness. Thirty-two children were chronically hoarse. Of these, 84 per cent had pathologic vocal fold findings (vocal fold nodules, oedema or hypertrophy of vocal folds, and functional problems) and 59 per cent had borderline velopharyngeal closure. In a

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follow-up study, McWilliams *et al.*[20] found that altering velopharyngeal competency often improved laryngeal and voice symptoms.

Lohmander-Agerskov *et al.*[21] reported a longitudinal study of 15 CLP children in whom the soft palate repair was performed early and the hard palate repair late. At the age of five years, almost half of the children (47 per cent) were dysphonic. At the age of nine years, one year after the final closure of the hard palate, only four children (27 per cent) were mildly dysphonic.

Godbersen[1] presented a classification system comprising primary, secondary and tertiary speech disorders in CP children. Primary speech disorders are caused by incomplete velopharyngeal closure, resulting in hypernasality, weak plosives, fricatives and affricates. Secondary speech disorders are substitute mechanisms for plosives, fricatives and affricates which are created in the nose, pharynx or larynx. Tertiary speech disorders are functional dysphonias which derive from primary and secondary speech disorders.

Leder and Lerman[22] studied the relationship between hypernasality and laryngeal dysfunction in adults with repaired CP. Acoustic evidence of abnormal laryngeal activity was found only in speakers with significant hypernasality. The authors suggested that the abnormal laryngeal valving detected in the patients with hypernasality may be a compensation for velopharyngeal incompetency. The vocal folds were inappropriately adducted in order to provide a constriction inferior to the inadequately functioning velopharyngeal closure.

D'Antonio *et al.*[16] studied cleft and non-cleft individuals with velopharyngeal insufficiency. They did not find a clear relationship between laryngeal and voice findings and nasoendoscopic or aerodynamic assessments of velopharyngeal dysfunction. However, there was a significant relationship between laryngeal and voice findings and estimated subglottic pressure values. The authors supposed that some speakers compensate for inappropriate velopharyngeal valving with either increased respiratory effort or abnormal laryngeal valving.

Kawano *et al.*[23] combined the use of fibre-optic endoscopy and fluorovideoscopy to investigate the exact site of faulty articulation and the relation between velopharyngeal function and faulty articulation in a cleft population. They showed that faulty articulation in CP speech, such as laryngeal fricatives and affricates, pharyngeal stop, and glottal stop, is secondary to velopharyngeal incompetence. These voices were articulated in the larynx at various sites, such as the epiglottis, arytenoids, aryepiglottic folds and vocal folds – that is, much lower than was supposed on the basis of auditory perception.

In Slovenia, a multidisciplinary team consisting of a maxillofacial surgeon, an ENT specialist, a speech and language pathologist, an orthodontist, and a psychologist treat children who have clefts. The team is organized according to the recommendations of the Eurocran group.[24] This treatment starts at birth, and the patient is monitored until body growth is complete. Therefore, ENT problems can be detected and treated early.

The purpose of this study was to assess the prevalence of ear, nose and voice problems in Slovenian children with CP and UCLP, to make comparisons between both groups (differing with regard to the age when the soft palate became functional), and to determine the risk factors, particularly for voice disorders.

Methods

The research was retrospective. The studied population consisted of all prepubertal CP and UCLP children regularly treated by the Slovenian multidisciplinary team. Eighty CP children (35 boys and 45 girls) and 73 UCLP children (47 boys and 26 girls) were included in the study. They were aged from four to 12 years old at their last visit to the ENT clinic, with a mean age of 6.6 years. In order to exclude mutational voice disorders, children older than 12 years were not included in the study.

In Slovenia, a different surgical approach is used in CP and UCLP children, with regard to the type of cleft. For all the children with UCLP, pre-operative treatment with an intraoral plate and strapping of the upper lip had been performed. The lip repair was performed at six months of age, the soft palate repair at one year and the hard palate repair with mucoperiostal closure of the alveolus at between two and three years of age.[26] In the children with CP, in order not to interfere with maxillary growth, the soft and hard palates were repaired in a single session at the age of 2.5–3.0 years. All surgical procedures were conducted by the same senior maxillofacial surgeon using a standard technique. Children had completed their surgical cleft treatment at least one year before inclusion into the study. The children were followed up after the completed surgical treatment for a period ranging from one and a half to nine years, with a mean period of 4.2 years.

The patients were examined by the team after every surgical procedure. An ENT examination and hearing test was always performed. Since 2002, 21 children with CP or CLP have also received an ENT examination and hearing test performed before soft palate closure. All the cleft children were given a check-up once a year unless findings suggested that more frequent examinations and treatment were necessary. In the case of conductive hearing loss exceeding 30 dB (measured at 0.5, 1, 2 and 4 kHz) and lasting more than six months, grommet insertion was performed. Velopharyngeal competency was assessed during phonation by means of posterior rhinoscopy or nasoendoscopy and during speech by means of a mirror test. The data on ROM and persistent SOM, grommet insertion, hearing test results, deviation of the nasal septum, other nasal breathing disturbances, mobility of the soft palate, velopharyngeal closure, the position of the tongue, laryngoscopic findings, and motor abilities of the articulation organs were obtained from the ENT specialist's medical documentation. The nasal breathing was assessed as

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impaired according to rhinoscopy findings and whether the child breathed orally for more than 50 per cent of the sleeping time. The incorrect position of the tongue was on the bottom of the oral cavity. The characteristics of the groups and the prevalence of these ENT problems were compared for both groups with clefts (UCLP and CP) using the χ^2 test and the analysis of variance test. The risk factors for voice problems were determined using the χ^2 test. The SPSS software package (version 11.0) was used for statistical analysis.

Results

As all prepubertal CP and UCLP children were included in the study, the groups were not matched for sex ratio. There was a significant difference between the CP children (35 boys and 45 girls) and the UCLP children (47 boys and 26 girls) with regard to gender ($p = 0.011$).

The CP children (mean age 6.3 years, standard deviation 2.9 years) and UCLP children (mean age 6.8 years, standard deviation 3.7 years) did not differ with regard to age ($p = 0.396$).

The ENT findings of the regular follow-up examinations are presented in Tables I, II and III.

In the whole group of children with clefts ($n = 153$), there were 86 children (56.2 per cent) with ear pathology resulting in protracted hearing loss lasting more than three months per year and exceeding 30 dB. For 39 cleft children (25.5 per cent), the insertion of grommets was necessary in order to improve hearing ability (Table I).

The ENT examination and hearing test performed before closing the soft palate detected bilateral SOM in all 21 tested children with CP or UCLP.

In the whole group of children with clefts, there were 50 subjects (32.7 per cent) with impaired nasal breathing due to deviation of the nasal septum or/and hyperplastic chronic rhinitis (Table II).

The CP and UCLP groups did not differ with regard to soft palate mobility, motor abilities of the articulation organs, the position of the tongue in the oral cavity or voice disorders. There were more children with velopharyngeal insufficiency in the CP group than in the UCLP group, but the difference was not statistically significant (Table III).

The position of the tongue in the oral cavity was assessed in 140 children. In 28 cases, the tongue was on the bottom of the oral cavity. The children with impaired nasal breathing ($n = 46$) had significantly more instances of incorrect tongue position in the oral cavity (17/46) than did the children with good nasal breathing (11/94) ($p = 0.000$).

In the whole group of children with clefts, there were 19 (12.4 per cent) subjects with voice disorders. Functional dysphonia without vocal fold nodules was the cause of hoarseness in six CP and four UCLP children. Two CP children and two UCLP children had vocal fold nodules. Allergic laryngitis was detected in two CP and three UCLP children.

In five children with functional dysphonia, the hearing ability was normal. In nine children with functional voice disorder, persisting bilateral (eight children) or unilateral (one child) mild-to-moderate hearing impairment was detected on several occasions. The insertion of grommets was necessary in seven children. Two hoarse children were four years of age; the others were of school age.

The other factors possibly influencing the prevalence of functional dysphonia are listed in Table IV.

TABLE I

PREVALENCE OF EAR PATHOLOGY IN CHILDREN WITH CLEFTS*

ENT finding	CP children ($n = 80$)	UCLP children ($n = 73$)	p
ROM	19	20	0.472
SOM	43	43	0.625
SOM + grommets	20	19	0.884

*$n = 153$. CP = cleft palate; UCLP = unilateral cleft lip, alveolus and palate; ROM = recurrent acute otitis media; SOM = secretory otitis media

TABLE II

PREVALENCE OF NOSE PATHOLOGY IN CHILDREN WITH CLEFTS*

ENT finding	CP children ($n = 80$)	UCLP children ($n = 73$)	p
Nasal septum deviation	7	67	0.000
Hyperplastic chronic rhinitis	14	18	0.513
Disturbed nasal breathing	14	36	0.000

*$n = 153$. CP = cleft palate; UCLP = unilateral cleft lip, alveolus and palate

Discussion

The results of the study showed that ENT pathology appeared in almost two-thirds of the children with clefts. Although the soft palate repair was performed at different ages, no differences were found between the CP and UCLP groups with regard to ear pathology. A total of 56.2 per cent of children with clefts had SOM, with half of them (25.5 per cent of all children with clefts) requiring surgical therapy. Duroux *et al.*[6] found SOM in 87 per cent of children with CP;

TABLE III

PREVALENCE OF MECHANICAL AND FUNCTIONAL VOICE PROBLEMS IN CHILDREN WITH CLEFTS*

ENT finding	CP children ($n = 80$)	UCLP children ($n = 73$)	p
Impaired soft palate mobility	6	2	0.186
Impaired motor abilities of articulation organs	8	8	0.853
Incomplete velopharyngeal closure	15	6	0.059
Incorrect tongue position	13	15	0.866
Functional dysphonia	8	6	0.593
Voice disorders	10	9	0.829

*$n = 153$. CP = cleft palate; UCLP = unilateral cleft lip, alveolus and palate

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TABLE IV

FACTORS INFLUENCING PREVALENCE OF FUNCTIONAL DYSPHONIA IN CHILDREN WITH CLEFTS*

Factor	Functional dysphonia ($n = 14$)	p
Disturbed nasal breathing ($n = 50$)	6	0.451
Impaired soft palate mobility ($n = 8$)	0	
Velopharyngeal insufficiency ($n = 23$)	2	0.849
Impaired motor abilities ($n = 16$)	2	0.724

*$n = 153$, $p > 0.05$ means statistical significance.

three years after surgical treatment of the CP, SOM persisted in 70 per cent of cases. Gordon *et al.*[26] studied late sequelae of SOM in CP patients; among 50 adolescent patients, 81 per cent had normal hearing but half of these patients had a history of grommet insertion. The ENT care of cleft children in Slovenia starts very early, and this might be the reason why the number of children with ear problems in our study was lower than in some other studies.

According to our preliminary results for hearing tests performed before surgical closure of the soft palate, all the cleft children had SOM. It is expected that after the closure of the cleft palate the function of the eustachian tube will be enabled. Closure of the soft palate was performed at the age of one year in the UCLP children and between two and three years of age in the CP children. We supposed that there would be less ear pathology in the UCLP group as a consequence of this early repair of the soft palate. However, the results of our study did not show any differences between the groups, suggesting that the time of soft palate closure is not crucial to reducing ear pathology. All children were followed up for a period ranging from one and a half to nine years, with a mean follow-up period of 4.2 years.

Others have noted that UCLP frequently produces nasal deformities that tend to impair nasal breathing;[12] this was also confirmed in the present study. Possible causes for the reduced size of the nasal airway can include a deviated nasal septum or mucosal hypertophy (chronic hyperplastic rhinitis). Although nasal septum deviation was detected in 48.4 per cent and hypertrophic rhinitis in 20.9 per cent of cleft children, nasal breathing was impaired in only 32.7 per cent of the studied cleft population. The cleft children were regularly examined and treated by an ENT specialist since the age of two and a half years. Early, effective treatment of hyperplastic rhinitis could be the cause of the better nasal patency in our series. We suppose that the use of presurgical orthopaedics could contribute to a diminution of nasal septum deviation and so improve patency of the nasal cavity.

The incorrect position of the tongue in the oral cavity can be a consequence of permanently impaired nasal breathing, possibly resulting in abnormal maxilla growth. In 28 children with CP or UCLP, improper position of the tongue in the bottom of the oral cavity was detected. In 60.7 per cent of these children, nasal breathing was impaired as a result of a marked septum deviation. In other children, hyperplastic nasal mucosa reduced the airway and disturbed normal nasal breathing. We believe that, in the very few cases in which treatment with medication is not successful and the growth of the maxilla is jeopardized, surgical treatment of the deviated nasal septum or mucosal hypertrophy is necessary.

Some authors report that CP children require a velopharyngeal flap significantly more often than UCLP children in order to improve velopharyngeal valving.[27] The results of the present study showed incomplete velopharyngeal closure in 18.8 per cent of CP children and in 8.2 per cent of UCLP children. This difference was close to being statistically significant. The mobility of the soft palate was impaired more often in CP children than in UCLP children. No differences were detected in the motor abilities of the articulation organs in both groups of cleft children. We suppose that it was not the surgical procedure that influenced the soft palate mobility or velopharyngeal competency because all the cleft children were operated on by the same senior surgeon using the same surgical technique. In almost two-thirds of cleft children with velopharyngeal insufficiency, a disproportion between soft palate length and pharyngeal depth was noticed.

The possible consequences of incomplete velopharyngeal closure in CP or CLP children include tertiary speech disorder-type voice problems.[1] These appear as a result of hyperactivity of the muscles involved in phonation and articulation. The results of our study showed a smaller prevalence of voice disorders in cleft children than that found in other studies.[15–17,19–21] However, voice disorders in the cleft children were no more common than in the non-cleft population. The prevalence of voice disorders in pre-school and school children varies between 6 per cent,[28] 10 per cent[29] and 23 per cent.[30] There were 8% out of 202 children having functional dyshonia. There were 14.9% dysphonic children having functional dysphonia or laryngitis.[31] The reason for the relative rarity of voice disorders among the cleft children in Slovenia may be the early introduction of speech therapy, which is commenced immediately after completion of surgical treatment. As a result, the development of faulty articulation patterns and consequent voice disorders is prevented.

We also attempted to identify the factors influencing the prevalence of voice disorders. From the prevalence of velopharyngeal insufficiency in CP children, voice disorders would be expected to be more common in the CP group. The results of the present study did not show any differences in the prevalence of voice disorders, and particularly functional dysphonia, in CP and UCLP children. One of the possible reasons for this could be the early speech therapy, which begins at the age of 2.5–3 years in both groups.

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Long-lasting hearing loss was found to be the only factor possibly influencing the prevalence of dysphonia among cleft children. Any hearing loss impairs auditory control of the child's voice. Hearing-impaired children often speak loudly and abuse their voices. The results of the study did not detect any important differences in the prevalence of ear pathology in the groups of CP and UCLP children. This could be another reason why the prevalence of voice disorders in both groups of cleft children is almost the same.

Conclusion

We found that various ENT problems appeared in almost two-thirds of children with CP or UCLP in our series. In about 25 per cent of our children, a surgical procedure was necessary to normalize their hearing ability. Nasal septum deviation or chronic hyperplastic rhinitis may have caused impaired nasal breathing in the great majority of the UCLP children. Voice disorders were no more common than in the population of non-cleft children and were frequently related to hearing loss.

Therefore, an ENT specialist should take an active role early in the treatment of children with clefts in order to improve the children's hearing ability and nasal breathing and to prevent functional voice disorders. Good cooperation with the maxillofacial surgeon and the whole multidisciplinary team is also essential.

- **This study reports otolaryngological pathology in 153 children with facial and palatal clefts**
- **Fifty-six per cent of children with clefts had otitis media with effusion, half of them requiring surgical therapy**
- **Nasal breathing was impaired in 32 per cent of patients. Deviation of the nasal septum with airway impairment was significantly more common in children with unilateral cleft lip, alveolus and palate compared with children with cleft palate**
- **Dysphonia was present in 12 per cent of cleft children, a lower rate than that reported in previous studies**
- **The importance of early multidisciplinary treatment is emphasized**

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