Treatment outcome after neonatal cleft lip repair in 5-year-old children with unilateral cleft lip and palate

Olga Košková a, b, d, *, Jitka Vokurková a, b, d, Jan Vokurka c, d, Alena Bryšová c, d, Pavel Šenovský c, d, Julie Čefelinová a, Darina Lukášová a, Petra Dorociaková e, Juraj Abelovský d

a Department of Pediatric Plastic Surgery - Department of Pediatric Surgery, Orthopedics and Traumatology, University Hospital Brno, Černopolní 9, 613 00 Brno, Czech Republic
b Department of Burns and Reconstructive Surgery, University Hospital Brno, Jihlavská 20, 625 00 Brno, Czech Republic
c Clinic of Dentistry, St. Anne’s Faculty Hospital, Fekářská 53, 656 91 Brno, Czech Republic
d Faculty of Medicine, Masaryk University, Brno, Czech Republic
e Receto, Faculty of Science, Masaryk University, Kamenice 753/5, 625 00, Brno, Czech Republic

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ABSTRACT

Introduction: The aim of this study was to assess speech outcomes and dental arch relationship of 5-year-old Czech patients with unilateral cleft lip and palate (UCLP) who have undergone neonatal cleft lip repair and one-stage palatal closure.

Methods and materials: Twenty-three patients with UCLP, born between 2009 and 2010, were included in the study. Three universal speech parameters (hypernasality, articulation and speech intelligibility) have been devised for speech recordings evaluation. Outcomes of dental arch relationship were evaluated by applying the GOSLON Yardstick and subsequently compared with the GOSLON outcome of other cleft centers.

Results: Moderate hypernasality was present in most cases, the mean value for articulation and speech intelligibility was 2.07 and 1.93, respectively. The Kappa values for inter-examiner agreement for all the three speech outcomes ranged from 0.786 to 0.808. Sixty-three percent of patients were scored GOSLON 1 and 2, 26% GOSLON 3, and 10% GOSLON 4. GOSLON mean score was 2.35. Interrater agreement was very good, represented by kappa value of 0.867.

Conclusion: The treatment protocol, involving neonatal cleft lip repair and one-stage palatal repair performed up to the first year of UCLP patient’s life, has shown good speech outcomes and produced very good treatment results in regard to maxillary growth, comparable with other cleft centers.

1. Introduction

The incidence of cleft lip and palate (CLP), one of the most common craniofacial malformations, is 11.13 for 10 000 live births in the Czech Republic [1]. Treatment protocol of CLP is different in each cleft center. In the Czech Republic there is an early surgery trend being observed [2,3]. In our department cleft lip repair is performed in neonatal period and cleft palate repair begins at the age of 6 months and later respectively.

Final treatment outcomes of CLP patients cannot be fully assessed until adulthood; however, there are some predetermined periodic follow-up visits during the CLP patient’s maturing. According to Eurocleft study [4] patients with CLP should undergo follow-up assessment sessions at 5, 10 and 18 years of age which include standardized speech audio recordings, photos and dental models. Inter-center comparison is necessary for treatment success rating.

The purpose of the study was to analyze speech outcomes and dental arch relationship in the same sample group of 5-year-old children with unilateral cleft lip and palate (UCLP), treated with neonatal cleft lip repair and one-stage palatal reconstruction.

There is no standard speech-sampling protocol or guidelines for capturing cleft-palate speech errors in Czech language which can
be used for inter-language comparison. Speech-sampling protocols created by Kerekretiova [5] or Brohm [6] are mostly used for Czech CLP patients but, in our view, they are either too extensive or too short and thus insufficient. Therefore the study was focused on creating a contemporary speech protocol, specifically designed for Czech language, and its assessment in accordance with the recognized speech therapy schemes currently available.

There are more options for assessment of maxilla’s growth and resulting analysis of the occlusal effects of primary surgery [7]. The Great Ormond Street, London and Oslo, Norway Yardstick, more commonly known as the GOSLON Yardstick [8], was applied in the study. The outcomes evaluated by GOSLON Yardstick were compared with other cleft center results.

2. Material and methods

The study was based on the approval of the ethics committee of St. Anne’s University Hospital in Brno (reference number 25V/2015).

2.1. Subjects

In the Department of Pediatric Plastic Surgery, the University Hospital Brno, 25 patients, born between January 2009 and December 2010, were treated. Two patients were excluded because of diagnosis of syndrome associated with cleft; all the remaining children proceeded in the study. All patients were operated on by an experienced surgeon (Vokurkova, MD). Speech audio recordings, photo documentation and dental models were being completed in all the children with UCLP. Children’s age at the time of lip and palate closure, gender and presence of oro-nasal communication were also observed. The mean age of the 23 patients was 5.3 years (4.5–6.2). Unilateral cleft lip and palate affected the left side in 70% of the children in the study. Seventeen patients (77%) were boys. The mean age at cleft lip surgery was 6.2 days (2–18 days), the mean age at palate repair was 6.9 months (6–12months). Four out of the 23 children had an oro-nasal fistula after primary palatal surgery.

2.2. Surgical procedures and treatment protocol

The optimal surgery timing is determined by each cleft center [4,9,10]. In our department, cleft lip repair is performed in neonatal period (0–28 days of age). A modified Fisher’s method, combining a correction of the lip and nose [11], was used in each case in the study. Early lip closure, as a natural nasoalveolar molding, helps bring both alveolar segments closer [2]. All patients underwent palate closure between six and twelve months of age. One stage palate repair using intravelar veloplasty is preferred. The emphasis is on the most proper restoring of all abnormal muscle insertions [12], minimal lateral incisions, and no fracture of hamulus. A vomer flap was not applied as a standard; it was used only in a few unique cases of particularly serious clefts. In case of velopharyngeal insufficiency presence at pre-school age, the Furlow palatoplasty or the veloplasty with “butterfly-suture”, based on principles by Haase’s method, is performed [13].

Every patient comes for follow-ups according to Eurocleft study [4]. Speech development is observed after palate repair and early speech therapy is indicated. We consider the follow up at the age of five as one of the most crucial ones. Speech progress assessment is determined by standardized speech audio recordings. Dental models are made for exploration of dental arch relationship for the purpose of planning the future orthodontics therapy before attending school.

2.3. Assessment

2.3.1. Speech recordings

A new system for evaluating speech outcomes in CLP patients was created for Czech language. At first, it was necessary to build a set of speech tests. Its scheme was based on principles developed by Kerekretiova [5], then it was revised and edited in accordance with Lohmander et al. [14]. The final speech protocol consists of articulation test (based on description of sample of pictures), test specialized on repetition of sentences containing high-pressure sensitive consonants and nasal consonants, counting from 1 to 10, connected speech – re-telling of a previously told story. Hypernasality was analyzed also by Gutzmann test and Czermak’s Mirror test. All children speech samples were recorded in the same room using the same recorder (Olympus linear PCM recorder LS-11EU). Subsequently, all audio recordings were taken for perceptual analysis. The speech outcomes were evaluated in three categories: hypernasality, articulation and speech intelligibility. All categories were rated on the 5-point scale (Table 1). The evaluation of recordings was performed independently by two speech therapists experienced in treating cleft patients. Speech samples were blinded before perceptual evaluation. The following rules had been respected; speech therapists were allowed to listen to all recordings repeatedly before making a final assessment, but the actual rating had to be done with no interruption. Two patients had to be excluded from the study because of diagnosis of selective mutism and dysphasia; twenty-one recordings were assessed.

2.3.2. The GOSLON Yardstick

The Goslon Yardstick is a 5-point scoring system for evaluation maxilla’s growth and dental arch constriction which categorizes all patient outcomes into 5 groups: from 1- excellent to 5 – very poor outcome (Table 2). All the 23 patients included in the study were rated. None of the patients had undergone active orthodontic treatment prior to this. Two experienced orthodontists (A.B. and P.S.), who rated the models, adhered to the following rules. Prior to the actual evaluation, each examiner was thoroughly familiarized with applying the GOSLON Yardstick in order to prevent systematic bias. Therefore, as the first step, testing assessment and subsequent standardization of evaluation were performed. The final evaluation process, determined after a certain period, was performed by each examiner separately and with no interruption during the evaluation. The results were compared with outcomes of other cleft

<table>
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<tr>
<th>Score</th>
<th>Hypernasality</th>
<th>Articulation</th>
<th>Speech intelligibility</th>
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<tbody>
<tr>
<td>1</td>
<td>Absent</td>
<td>Normal articulation or other common dysarticulation</td>
<td>Normal (100%)</td>
</tr>
<tr>
<td>2</td>
<td>Minimal</td>
<td>Weak pressure consonants, presence of nasality</td>
<td>Good (90–99%)</td>
</tr>
<tr>
<td>3</td>
<td>Mild</td>
<td>Mild cleft type compensatory misarticulation</td>
<td>Lower (75–89%)</td>
</tr>
<tr>
<td>4</td>
<td>Moderate</td>
<td>Moderate cleft type compensatory misarticulation</td>
<td>Very low (50–74%)</td>
</tr>
<tr>
<td>5</td>
<td>Severe</td>
<td>Severe cleft type compensatory misarticulation, consonantless speech</td>
<td>Unintelligible (49% and less)</td>
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centers worldwide. Only the studies concerning 4–7 year-old patients were selected following the age categories by Nollet [15].

2.4. Statistical analysis and inter-examiner agreement

The kappa statistic was used to evaluate inter-rater reliability for both, the speech therapists and the orthodontists. Significance levels were set at \( p \leq 0.05 \). According to Landis and Koch (1977) [16], kappa value > 0.6 represents “good” strength of agreement, and kappa >0.8 indicates “very good” strength of agreement. The analysis was carried out using the statistical package SPSS Ver. 22.

3. Results

Speech recordings were evaluated in three categories — hypernasality, articulation and speech intelligibility - each rated on the 5-point scale scoring system. Mean value of hypernasality was 3 (average 2.79), 38.2% patients were rated hypernasality of 3 by both speech therapists. Mean value of other categories was 2 (average 2.07 for articulation and 1.93 for speech intelligibility). More than 90% achieved rating 1 to 3 for articulation as well as speech intelligibility. All values are shown in Table 3. The Kappa values for inter-examiner agreement for all the three speech outcomes ranged from 0.786 to 0.808, indicating good/very good agreement.

Out of the 23 patients, 13% were scored GOSLON 1 (Figs. 1–4), GOSLON 2 represented approximately 50% of children and the remaining ones were classified into GOSLON score of 3 and 4 (Figs. 5–8). No dental study model was found to be in group 5. GOSLON mean score was 2.35. Inter-rater agreement was very good, represented by kappa value of 0.867 (standard error 0.088) (Table 4).

4. Discussion

The success of the surgery depends on quality of soft tissues, severity of the cleft [17], surgeon’s experience, smooth post-operative course, and highly specialized multidisciplinary treatment from birth to early adulthood [18]. Treatment protocol and particularly timing of surgeries in cleft patients are still a highly debated topic in academic circles. Timing of cleft lip and palate surgical repairs is determined by each institution and country
In the Czech Republic there is an early surgery trend being observed, cleft lip repair is performed in neonatal period and cleft palate repair follows at about 6 months of age respectively [2,3]. Early cleft lip closure is considered controversial but sometimes criticized unsubstantially. It is not uncommon to find both, opponents as well as supporters of this method. The first mention of neonatal cleft lip surgery is dated to the sixties of the last century [20]. In the other decades NS Desai was an important personality in promoting this method in England [21,22]. Early cleft lip repair is now performed in France, the Czech Republic and Slovakia [2,3,23,24]. This method’s expansion has always been limited by the need for special equipment of pediatric recovery department, and particularly an experienced team of pediatric anesthesiologists. Lately, the negative effect of anesthesia in children is being increasingly discussed. It’s a question for anesthesiology societies whose conclusion will affect the future of this method. Currently we are awaiting the results of large multicenter studies [25]. Recently published works both confirm and refute the negative effect of anesthesia. According to Wilder et al. [26], children receiving multiple anesthesia demonstrate increasing incidence of

![Fig. 2. 5-year-old girl with UCLP (side face)- GOSLON score 1.](image1)

![Fig. 3. 5-year-old girl with UCLP (occlusion-frontal view) – GOSLON score 1.](image2)

![Fig. 4. 5-year-old girl with UCLP (occlusion-side view)- GOSLON score 1.](image3)

![Fig. 5. 5-year-old boy with UCLP (front view)- GOSLON score 4.](image4)
learning disabilities. Conversely, the Czech study by Petrakova et al. [27] observed that no negative impact of earlier anesthesia on intelligence quotient in 3–7 year old children with cleft lip was recorded compared to the group of patients on which later anesthesia was performed.

In our institution we prefer the following treatment protocol: primary cleft lip repair is performed from the second day of life, primary cleft palate repair starting at 6 months of age in patients with both unilateral and bilateral complete cleft lip and palate. This protocol has been used in our department since 2005, when the first cleft lip repair was performed in neonatal period by J. Vokurkova, MD, in the Czech Republic. On parents’ request cleft lip repair might be postponed to a later age. Early cleft lip surgery enables cleft palate repair soon to be followed in the first year of life. Early cleft closure has a beneficial effect on infant feeding. The neonates accept first nourishment per os mostly in four hours after a lip surgery. In our department the need for nasogastric tube was in less than 5% cases in 2014 [28].

Plastic surgeons, dealing with cleft palate issues, are used to finding the delicate balance between the pressure to perform an earlier cleft palate repair because of the right development of speech, and a postponed surgery in order not to affect maxilla’s growth [29]. Cleft palate repair is essential for creating correct speech stereotypes and it is difficult to correct once acquired compensatory speech patterns [30]. In addition to poor speech development, feeding problems are observed in cases of late cleft palate closure. Especially the transition from liquid to solid food can cause many difficulties for patients as well as for parents. Early palate surgery enables timely introduction of non-milk-based foods into the infant’s diet. However, in some studies authors advocate maxilla’s growth over speech development [31]. As a result some surgeons choose the middle path and prefer two stage palate closure; at first, closure of soft palate before the twelfth month of life, and hard palate closure following later on [32].

There are many rating systems for evaluation maxilla’s growth and dental arch constriction available [33]. The GOSLON Yardstick [8], the 5 Year Olds’ Index [34] or modified Huddart/Bodenham scoring system [35,36] are most commonly utilized. The GOSLON Yardstick was designed by Mars et al. in 1987 and until present day

<table>
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<th>Score</th>
<th>GOSLON Yardstick</th>
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<tr>
<td></td>
<td>Examiner 1</td>
</tr>
<tr>
<td>1</td>
<td>3 (13.04%)</td>
</tr>
<tr>
<td>2</td>
<td>11 (47.83%)</td>
</tr>
<tr>
<td>3</td>
<td>7 (30.43%)</td>
</tr>
<tr>
<td>4</td>
<td>2 (8.70%)</td>
</tr>
<tr>
<td>5</td>
<td>0 (0.00%)</td>
</tr>
<tr>
<td>Mean</td>
<td>2.35</td>
</tr>
<tr>
<td>Average</td>
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<table>
<thead>
<tr>
<th>Kappa value</th>
<th>Standard error</th>
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<tr>
<td>0.867</td>
<td>0.088</td>
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it is considered to be one of the best available and the most widely used system for assessing surgical outcomes [37,38]. Initially the Goslon Yardstick was created for evaluating 10 year-old UCLP patients’ dental models. However, applying this method for rating in younger children was justified by Mars et al. in 2006 [39]. The Goslon Yardstick has been used in many studies, including the Eurocleft comparative studies of patients with UCLP [15,40]. The Goslon Yardstick assesses malocclusions in patients with UCLP. Its reliability is comparable to the 5-year-old index by Attack [39]. The Goslon reliability by comparison with another frequently used scoring system, modified Huddart/Bodenham (MHB) [37], is lower. The major advantage of Goslon Yardstick is also its most common use in published works in the last twenty years [41] and as a result, there is a great level of comparison with other cleft centers available [15,42]. Our treatment protocol, neonatal cleft lip repair and one-stage palate repair in the first year of life, was found to produce the Goslon score that approximated the results of other cleft centers worldwide. A comparison with other cleft centers is shown in Table 5 and Figs. 9 and 10.

A new testing sample for evaluation of speech outcomes in Czech language was created. In the study most of patients were rated hypernasality of 3 in 5-point scale, which represents an average outcome. Articulation (mean value 2.07) and speech intelligibility (mean value 1.93) showed good speech outcomes. The disadvantage of our study is a small number of participants. The evaluation of UCLP patients proceeds and more data will be presented in the future.

5. Conclusion

A new speech testing sample for Czech language was created and assessed as sufficient for purpose of speech outcome rating. Hypernasality of mean value 2.79, articulation of 2.07 and speech intelligibility (mean value 1.93) on the 5-point scale show good speech outcomes. The mean Goslon score of 2.35 is comparable with other cleft centers worldwide.
centers. In summary, this study shows that our concept seems to be a proper approach to treating cleft patients based on the results achieved in terms of occlusion and speech outcomes of 5-year-old UCLP patients.

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None.

**Conflict of interest**

None.

**References**