



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



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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.

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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

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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 1
Speech outcomes evaluation – hypernasality, articulation and speech intelligibility.

Score	Hypernasality	Articulation	Speech intelligibility
1	Absent	Normal articulation or other common dysarticulation	Normal (100%)
2	Minimal	Weak pressure consonants, presence of nasality	Good (90–99%)
3	Mild	Mild cleft type compensatory misarticulation	Lower (75–89%)
4	Moderate	Moderate cleft type compensatory misarticulation	Very low (50–74%)
5	Severe	Severe cleft type compensatory misarticulation, consonantless speech	Unintelligible (49% and less)

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