NASAL FUNCTION AND AESTHETICS IN PATIENTS WITH COMPLETE UNILATERAL CLEFT LIP, ALVEOLUS AND PALATE

Summary of the Doctoral Thesis for obtaining the degree of a Doctor of Medicine

Speciality – Oral and Maxillofacial Surgery

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The study was carried out: at Rīga Stradiņš University Department of Oral and Maxillofacial Surgery and P. Stradins CUH, 45th Department, Centre of Dentistry and Facial Surgery.

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The Doctoral Thesis is available in the RSU library and at RSU webpage: www.rsu.lv

Secretary of Doctoral Council:
*Dr. med.*, Professor **Ilga Urtāne**
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ABBREVIATIONS

2D  2 dimensional
3D  3 dimensional
al(L)  Alare (left)
al(R)  Alare (right)
alL-N  Nasal length on left side
alL-PRN  Alar wing length on left side
alL-Sn  Width of the left nostril at the base
alR-alL  Nose width
alR-N  Nasal length on right side, nose lateral length
alR-PRN  Alar wing length on right side
alR-Sn  Width of the right nostril at the base
ANOVA  Analysis of variance
ch(L)  Cheilion (left)
ch(M)  Cheilion (middle) upper
ch(M)lower  Cheilion (middle) lower
ch(R)  Cheilion (right)
chL-cphL  Length of red lip to the crista philtrum left
chR-cphR  Length of red lip to the crista philtrum right
CLCSI  Cleft Lip Component Symmetry Index
cm³/s  Cubic centimetre per second
Cph(L)  Crista philtrum left
Cph(R)  Crista philtrum right
cphL-chM  Length of red lip from crista philtrum to midline on left side
cphR-chM  Length of red lip from crista philtrum to midline on right side
Dx  Right side (in Latin dextra)
e.g.  for example
en(L)  Endocanthion (left)
en(M)  Endocanthion (middle)
en(R)  Endocanthion (right)
ex(L)  Exocanthion (left)
ex(R)  Exocanthion (right)
G  Glabella
i.e.  that is
IS  Institute of Stomatology
mm  millimetre
N  Nasion
NA  Not applicable
nd(L)  Nostril dome left
nd(R)  Nostril dome right
nl(L)  Nostril lower border left
nl(R)  Nostril lower border right
nlL-ndL Nostril height left
nlR-ndR Nostril height right
p  probability value (level of statistical significance)
P. Stradins CUH Pauls Stradins Clinical University Hospital
Pa Pascal
PRN  Pronasale
PRN-N Nose dorsum length
PRN-Sn Nasal height
RSU Rīga Stradiņš University
Sbal(L) Subalare left
Sbal(R) Subalare right
SbalL-cphL Length of white lip on left side
SbalR-cphR Length of white lip on right side
SbalR-SbalL Alar base width
Sin Left side (in Latin sinistra)
Sn Subnasale
Sn0(L) Edge of the columellar base left
Sn0(R) Edge of the columellar base right
VAS Visual analogue scale
INTRODUCTION

Cleft lip and palate is the most frequent congenital craniofacial deformity, an aetiology that is complex, involving both genetic and environmental factors (Krasone et al., 2014). Nose deformation is remarkable in patients with complete unilateral cleft lip, alveolus and palate (Kim et al., 2004), causing aesthetic and functional disturbances in early childhood, and negatively affecting physical and emotional development in childhood (Barkane, 1997). The mean prevalence of cleft lip and palate in Latvia is 1.2-1.4 per 1000 live births (Akota et al., 2001). The mean prevalence of cleft lip and palate in Europe is 1.4 to 2 per 1000 live births (Peterka et al., 2000).

Traditionally correction of cleft nasal deformity was postpone till nasal growth and development had been completed (Torre et al., 2000). A slow change in the attitude to cleft nasal deformity management and timing has occurred over the last 20 years. At present, it is becoming an increasingly popular opinion for correction of cleft nasal deformity during primary cleft lip repair (Byrd & Salomon, 2000). Although this operation reduces some of the psychological problems, it does not exclude subsequent deformation of the nose, with indications for additional correction of the nose (Guyuron, 2008).

As the central feature of the face, the nose has a profound effect on general facial aesthetics (He et al., 2009). Distortions of the nose range from almost invisible to catastrophic are dependent on the severity and type of cleft (van Loon et al., 2010). Remarkably, nasal deformity can cause psychological problems even in childhood. Therefore, Gosain and Fathi (2009) consider that secondary nasal deformity of unilateral cleft lip, alveolus and palate frequently is indicated for children as early as 6-8 years.

The outcome of the nose surgery is difficult to evaluate, due to the three-dimensional (3D) complexity of its shape (Nolst Trenité et al., 1997). There is
no single reliable method of aesthetically evaluating the nose for patients with clefts (Russell et al., 2001). Discussions of aesthetic evaluation of the nose in patients with cleft lip and palate remain problematical, many different methods having been described in the literature. In the review by Al-Omari et al. (2005), no single reliable method for aesthetic evaluation of the nose was noted. However, 3D images had already become most highly rated and remain so (Al-Omari et al., 2005). Though the most popular method for evaluating aesthetic of the nose is by 2D photography because it is simple, widely available, has a large database of information and is convenient. There is neither consensus about measurements to determine nasal aesthetic.

Barkane in Latvia evaluated cleft lip and nose surgical results of patients with different types of clefts in 1968. One of this study aims was to evaluate late deformations of cleft lip and nose of patients with different types of clefts who had been treated between 1900 and 1964 (Баркане, 1968).

Studies on congenital cleft lip and palate remain relevant today; there is no single option for the cleft nasal correction timing and methods to achieve both a good functional and aesthetic results. To evaluate nasolabial appearance and nasal function in patients with complete unilateral cleft lip, alveolus and palate objective, convenient methods are required so that the results of treatment can be assessed qualitatively and quantitatively.

**Aim of the study**

To evaluate nasolabial aesthetic and nasal function in patients with complete unilateral cleft lip, alveolus and palate, treated in cleft lip and palate centre at the RSU Institute of Stomatology (IS). To prepare clinical recommendations in practical oral and maxillofacial surgery, based on the results.
**Objectives of the study**

2. To evaluate nasal aesthetic in patients with complete unilateral cleft lip, alveolus and palate after cleft lip and nasal repair in two-dimensional (2D) photos on the visual analogue scale (VAS).
3. To assess nasal function in patients with complete unilateral cleft lip, alveolus and palate (study group) after cleft lip and nasal repair, and compare it with the control group.
4. To measure nasolabial symmetry in three-dimensional (3D) photos in patients with complete unilateral cleft lip, alveolus and palate (study group) after cleft lip and nasal repair, and to compare it with the control group.
5. To prepare clinical recommendations for children with unilateral cleft lip, alveolus and palate care.

**Hypotheses of the study**

1. Evaluation of nasal aesthetic in 2D photos is variable in small groups, but is sufficiently objective in 3D photos.
2. Nasal repair in children with complete unilateral cleft lip, alveolus and palate in preschool age, provides acceptable nasal function and symmetry.
Scientific novelty and practical value

In Latvia, nasal function and aesthetics in patients with complete unilateral cleft lip, alveolus and palate after surgical treatment has not been properly evaluated. In the first part of study, a modified Anastassov and Chipkov (2003) clinical scheme was adapted to evaluate nasal aesthetic in 2D photos. The second part was a complex evaluation of nasolabial symmetry and nasal function, applying the innovative equipment and method for nasolabial symmetry assessment in 3D photos, as well as measuring nasal function with active anterior rhinomanometry. This is the first time in Latvia that treatment results of patients with complete unilateral cleft lip, alveolus and palate have been compared with healthy individuals as the control group.

Objective and convenient methods are necessary in clinical practice to evaluate treatment results of nasolabial appearance and nasal function in patients with complete unilateral cleft lip, alveolus and palate regularly at a certain age.
1. MATERIAL AND METHODS

1.1. Design of the study

The study was conducted in RSU SI cleft lip and palate centre and the P. Stradins Clinical University Hospital (CUH), 45th department, Centre of Dentistry and Facial Surgery. Evaluation of nasal aesthetic in 2D photos was made retrospectively. Assessment of nasal function with active anterior rhinomanometry and measurement of nasolabial symmetry in 3D photos was a cross-sectional study.

The study had the permission of RSU Ethics Committee and informed consent of the patients and the parents of minors.

1.2. Study sample

The target population was children with congenital non-syndromic unilateral cleft lip, alveolus and palate.

For evaluation of nasal aesthetic in 2D photos, a systematic selection was made of all consecutive patients with unilateral cleft lip, alveolus and palate with primary cleft lip repair and rhinoplasty in RSU SI cleft lip and palate centre between 1 January 1995 and 31 December 2008, and who were >3 years old in 2011. There was a total of 59 children according to the RSU SI cleft lip and palate centre database.

For evaluation of nasal function and nasolabial symmetry in 3D photos, a systematic selection was made of all consecutive patients with congenital non-syndromic unilateral cleft lip, alveolus and palate who were born between 1 January 1994 and 31 December 2004 (aged 10-18 years), and were registered
in RSU SI cleft lip and palate centre. There was a total of 62 children according to the centre’s database.

Inclusion criteria:

Children with congenital non-syndromic complete unilateral cleft lip, alveolus and palate who:
1. Were born in Latvia and registered in RSU SI cleft lip and palate centre.
2. Were treated with appropriate surgical methods in RSU SI cleft lip and palate centre.
3. Had not been diagnosed with genetic syndromes.
4. Have available 2D photos in frontal and columellar projection at 3 or 5 years (or both ages), only for nasal aesthetic evaluation in these photos.

Exclusion criteria:
1. Children with congenital incomplete unilateral cleft lip, alveolus and palate or with Simonart’s band.
2. Children with complete unilateral cleft lip, alveolus and palate who were dead.
3. Children with complete unilateral cleft lip, alveolus and palate who were adopted abroad or had emigrated.
4. Children with complete unilateral cleft lip, alveolus and palate who were born in Latvia, but operated upon abroad.
5. Children who did not have qualitative 2D photos neither at 3 or 5 years of age (only for nasal aesthetic evaluation in 2D photos).

Diagnosis was checked by photos before primary cleft lip and nose repair. For nasal aesthetic evaluation in 2D photos after exclusion criteria, 27 children (11 girls and 16 boys) were included with complete unilateral cleft
lip, alveolus and palate who had qualitative 2D photos of frontal and columellar projections at 3 or 5 years, or both ages. The cleft was positioned on the left side in 20 patients, and on the right side in 7 patients. From the study, 32 children were excluded, 14 due to incomplete cleft lip, alveolus and palate, 4 due to Simonart’s band, 1 that had died before 3 years of age, 3 that had been adopted or emigrated abroad before 3 years of age, and 10 due to poor quality of their 2D photos or they were unavailable at 3 and 5 years of age.

For evaluation of nasal function and nasolabial symmetry in 3D photos according to exclusion criteria, 35 children with complete unilateral cleft lip, alveolus and palate were included. Of these patients, 30 (15 male, 15 female) responded for recall and were involved in the study. Nasal function was assessed and nasolabial symmetry in 3D photos was measured (the study group). The cleft was positioned on the left side in 21 cases and in 9 cases on the right side. The mean age was 14 years (range 10-18 years). Of the 27 children excluded from the study, 13 had incomplete cleft lip, alveolus and palate, 4 had Simonart’s band, 5 had died, 3 had been adopted or had emigrated abroad, and 2 had been operated on abroad.

The control group consisted of the first consecutive 35 (18 male, 17 female) volunteers who were healthy children without congenital deformities at 10 years of age from another randomised study at the Institute of Stomatology in Latvia.

1.3. Nasal aesthetic evaluation in 2D photos

After 2D photo analysis of the 27 children involved, and preparation for evaluation of their nasal aesthetic, 23 pairs of the photos in frontal and columellar projections were set at 3 years of age and 20 pairs of photos at 5 years of age. Photos were prepared by a unified system – on a grey scale, in
cropped triangles where only the nose and lips were visible (Fig. 1.1.). In all photos, the cleft was positioned on one side of lip, first on the left side, and then on the right side. Photo pairs, which consisted of frontal and columellar projection of 2D photos, anonymous, encoded and randomised, were inserted in Power-Point presentation.

![2D photo pair prepared for nasal aesthetic evaluation at 3 years of age, with cleft positioned on the left side](Image)

For nasal aesthetic evaluation in 2D photos, a modified Anastassov and Chipkov (2003) scheme was adapted. Visual analogue scale (VAS) was also used to compare nasal appearances. Before evaluation, experts were introduced to the photos and trained in assessing. Every pair of photos was evaluated one minute after the modified Anastassov and Chipkov (2003) clinical scheme, which was specially adapted for nasal aesthetic evaluation in 2D photos of patients with complete unilateral cleft lip, alveolus and palate (Table 1.1.). This nasal aesthetic evaluation scheme consisted of negative points from 0.5 to 3, which reflects severity of nasal deformation; the more points given, the more severe the nasal deformity. In total, 2 evaluators provided 4 evaluations. Evaluation was made twice, once with the cleft positioned on the left side, and the second when the cleft was on the right side. Evaluations were made at an interval of one week.
For evaluation of nasal aesthetic in 2D photos with VAS, vertical, without sections, 100 mm long posts were used, where the top is indicated by “a symmetric nose, without deformation”, with the bottom by “a greater deformation cannot be imagined”. Four observers made the evaluations, which was done once with all the clefts being positioned on the left side, and once with all the clefts being positioned on the right side, performed at one-week intervals.
1.4. Nasal function assessment

To assess nasal respiration for both groups, active anterior rhinomanometry involved the Otopront combined rhinomanometry and acoustic rhinometry tool RHINO-SYS (Happersberg otopront GmbH, Germany). Rhinomanometry of children with complete unilateral cleft lip, alveolus and palate was carried out at least 6 months after last nasal correction (without and with nasal decongestion; nasal drops of 0.1% Xymelin, according to international standards). The results were compared with normal values given in Bachmann (1983) (Table 1.2).

### Table 1.2.

<table>
<thead>
<tr>
<th>Degree of nasal obstruction</th>
<th>Nasal flow [cm3/s] at 150 Pa</th>
</tr>
</thead>
<tbody>
<tr>
<td>No obstruction</td>
<td>&gt; 500</td>
</tr>
<tr>
<td>Slight obstruction</td>
<td>300-500</td>
</tr>
<tr>
<td>Moderate obstruction</td>
<td>180-300</td>
</tr>
<tr>
<td>Severe obstruction</td>
<td>&lt; 180</td>
</tr>
</tbody>
</table>

1.5. 3D stereo-photogrammetry and measurements

Three-dimensional stereo-photogrammetry was performed with a 3D stereo-photogrammetric camera set-up 3dMD face System (3dMDfaceSystem; 3dMD LLC, Atlanta, GA, USA). The subject was seated on a chair at a fixed distance of 90 cm from the camera in a natural head position and habitual occlusion. For children with complete unilateral cleft lip, alveolus and palate, all 3D photographs of the face were taken at least 6 months after the last nasal correction. Photographs with right-sided clefts were reflected so that every cleft appeared on the left side, after which 25 anthropometric landmarks
and 18 distances were selected, based mainly on Farkas et al. (1993), with the modifications by Hood et al. (2004) and van Loon et al. (2010) (Fig. 1.2., Table 1.3. and Table 1.4.).

Fig. 1.2. 3D photo with selected anthropometric landmarks

Anthropometric landmarks were set manually on a facial scan using the 3dMD Vultus software landmarking tool (3dMD Vultus Software Platform, 3dMD LLC, Atlanta, USA). The reference vertical plane (Y) was positioned through glabella and nasion, and the horizontal plane (X) was positioned through right and left endocanthison soft tissue landmarks (Verzé et al., 2014). Landmarks were repeatedly set with a one-week interval, and intra-observer reliability was calculated.
### Table 1.3.

**Anthropometric landmarks**

<table>
<thead>
<tr>
<th>No.</th>
<th>Anthropometric landmarks</th>
<th>Abbreviation</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Endocanthion (left)</td>
<td>en(L)</td>
</tr>
<tr>
<td>2</td>
<td>Endocanthion (right)</td>
<td>en(R)</td>
</tr>
<tr>
<td>3</td>
<td>Endocanthion (middle)</td>
<td>en(M)</td>
</tr>
<tr>
<td>4</td>
<td>Exocanthion (left)</td>
<td>ex(L)</td>
</tr>
<tr>
<td>5</td>
<td>Exocanthion (right)</td>
<td>ex(R)</td>
</tr>
<tr>
<td>6</td>
<td>Nasion</td>
<td>N</td>
</tr>
<tr>
<td>7</td>
<td>Glabella</td>
<td>G</td>
</tr>
<tr>
<td>8</td>
<td>Pronasale</td>
<td>PRN</td>
</tr>
<tr>
<td>9</td>
<td>Subnasale</td>
<td>Sn</td>
</tr>
<tr>
<td>10</td>
<td>Cheilion (middle) upper</td>
<td>ch(M)</td>
</tr>
<tr>
<td>11</td>
<td>Cheilion (middle) lower</td>
<td>ch(M)lower</td>
</tr>
<tr>
<td>12</td>
<td>Cheilion (left)</td>
<td>ch(L)</td>
</tr>
<tr>
<td>13</td>
<td>Cheilion (right)</td>
<td>ch(R)</td>
</tr>
<tr>
<td>14</td>
<td>Crista philtrum left</td>
<td>Cph(L)</td>
</tr>
<tr>
<td>15</td>
<td>Crista philtrum right</td>
<td>Cph(R)</td>
</tr>
<tr>
<td>16</td>
<td>Alare (left)</td>
<td>al(L)</td>
</tr>
<tr>
<td>17</td>
<td>Alare (right)</td>
<td>al(R)</td>
</tr>
<tr>
<td>18</td>
<td>Subalare left</td>
<td>Sbal(L)</td>
</tr>
<tr>
<td>19</td>
<td>Subalare right</td>
<td>Sbal(R)</td>
</tr>
<tr>
<td>20</td>
<td>Edge of the columellar base right</td>
<td>Sn0(R)</td>
</tr>
<tr>
<td>21</td>
<td>Edge of the columellar base left</td>
<td>Sn0(L)</td>
</tr>
<tr>
<td>22</td>
<td>Nostril dome left</td>
<td>nd(L)</td>
</tr>
<tr>
<td>23</td>
<td>Nostril dome right</td>
<td>nd(R)</td>
</tr>
<tr>
<td>24</td>
<td>Nostril lower border left</td>
<td>nl(L)</td>
</tr>
<tr>
<td>25</td>
<td>Nostril lower border right</td>
<td>nl(R)</td>
</tr>
</tbody>
</table>
For nasolabial measurements in 3D photos, the cleft lip component symmetry index (CLCSI) of Amaratunga (1988) was adapted, further in the study named the Symmetry Index. Symmetry index was calculated by formula:

\[ \text{distance in cleft side versus distance in non-cleft side} \times 100. \]

Perfect symmetry was given a value of 100, but values less or more than 100 characterised the degree of nasolabial asymmetry. If the nasolabial aesthetic index was in the range 90-110%, the surgical result was considered successful according to Li et al. (2010).

**Anthropometric distances**

<table>
<thead>
<tr>
<th>No.</th>
<th>Abbreviation</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>cphR-chM</td>
<td>Length of red lip from crista philtrum to midline on right side</td>
</tr>
<tr>
<td>2</td>
<td>cphL-chM</td>
<td>Length of red lip from crista philtrum to midline on left side</td>
</tr>
<tr>
<td>3</td>
<td>chR-cphR</td>
<td>Length of red lip to the crista philtrum right</td>
</tr>
<tr>
<td>4</td>
<td>chL-cphL</td>
<td>Length of red lip to the crista philtrum left</td>
</tr>
<tr>
<td>5</td>
<td>SbalR-cphR</td>
<td>Length of white lip on right side</td>
</tr>
<tr>
<td>6</td>
<td>SbalL-cphL</td>
<td>Length of white lip on left side</td>
</tr>
<tr>
<td>7</td>
<td>nlR-ndR</td>
<td>Nostril height right</td>
</tr>
<tr>
<td>8</td>
<td>nL-ndL</td>
<td>Nostril height left</td>
</tr>
<tr>
<td>9</td>
<td>alR-Sn</td>
<td>Width of the right nostril at the base</td>
</tr>
<tr>
<td>10</td>
<td>alL-Sn</td>
<td>Width of the left nostril at the base</td>
</tr>
<tr>
<td>11</td>
<td>alR-N</td>
<td>Nasal length on right side, nose lateral length</td>
</tr>
<tr>
<td>12</td>
<td>alL-N</td>
<td>Nasal length on left side</td>
</tr>
<tr>
<td>13</td>
<td>alR-PRN</td>
<td>Alar wing length on right side</td>
</tr>
<tr>
<td>14</td>
<td>alL-PRN</td>
<td>Alar wing length on left side</td>
</tr>
<tr>
<td>15</td>
<td>alR-alL</td>
<td>Nose width</td>
</tr>
<tr>
<td>16</td>
<td>SbalR-SbalL</td>
<td>Alar base width</td>
</tr>
<tr>
<td>17</td>
<td>PRN-N</td>
<td>Nose dorsum length</td>
</tr>
<tr>
<td>18</td>
<td>PRN-Sn</td>
<td>Nasal height</td>
</tr>
</tbody>
</table>
1.7. Statistical analysis

For statistical analysis of nasal aesthetic evaluation in 2D photos with modified and adapted Anastassov and Chipkov (2003) scheme, Cohen's Kappa coefficient and agreement were calculated. The Kappa values were 0.8-1.0 = excellent, 0.6-0.8 = good, 0.4-0.6 = moderate, 0.2-0.4 = fair, < 0.2 poor agreement (Landis & Koch, 1977). The ANOVA test was used for statistical analysis of results of VAS, with statistical significance set at p < 0.05.

For statistical analysis of nasal function descriptive methods were used. To determine the difference of nasal flow between the cleft nostril and non-cleft nostril, and between left and right nostril in control group, the paired t-test was used (significance p < 0.05). To determine the difference of nasal flow between study group and control group, the unpaired t-test was used (significance p < 0.05).

For statistical analysis of 3D measurements, the Dahlberg formula (Dahlberg, 1940) was used to check measurement error, considered acceptable if it was < 3% of the total variance. The Wilcoxon signed rank test was used to determine difference between the cleft and non-cleft sides in the study group, and the Mann-Whitney test was used to determine difference between the study and control group (significance p < 0.05). To determine the difference of symmetry index between the cleft nostril and the non-cleft nostril, and between the left and the right nostril in the control group, the paired t-test was used (significance p < 0.05). To determine the difference of symmetry index between study group and control group, the unpaired t-test was used (significance p < 0.05). ANOVA test was used to determine the correlation between nasal function and nasal symmetry (significance p < 0.05).
1.8. Surgical procedures

Two maxillofacial surgeons operated on all the patients with complete unilateral cleft lip, alveolus and palate. Lip closure was performed with the Millard rotation-advancement repair (Millard Jr, 1976). The design of this flap was also used for the concomitant primary nasal repair. The alar base was completely mobilized from the piriform area. Nasal septum reposition without fixation has been used since 1 January 2001, in all patients with complete unilateral cleft lip, alveolus and palate during primary lip repair. Secondary rhinoplasty was performed with an open approach, and included reposition and fixation of the nasal septum and lower lateral cartilages.

In the first part of the study, where nasal aesthetic was evaluated in 2D photos, primary lip repair was performed at a mean age of 6.3 months (range 2-24 months). Nasal septum reposition during primary lip repair without fixation was performed in 15 children. From the included patients at ages 3 and 5 years, no secondary nasal correction was performed before nasal aesthetic evaluation in 2D photos.

In the second part of the study, where nasal function and nasolabial symmetry was evaluated in 3D photos, primary lip repair was performed at a mean age of 6.6 months (range 3-24 months). 26 patients had secondary rhinoplasty at a mean age of 7.8 years (range 6-10 years). Five patients at early school age during secondary rhinoplasty also had additional correction of the upper lip. Fourteen patients had more than one secondary nose correction, unless the result was accepted from patients and surrounding point of view. Four patients had only primary lip and nose correction, and no secondary rhinoplasty.
2. RESULTS

2.1. Nasal aesthetic evaluation in 2D photos

The sum of the results of nasal aesthetic evaluation with the adapted and modified Anastassov and Chipkov (2003) evaluation scheme were between 0 and 3 points. None of patients had deformations referred to as “narrow sill”, “columellar base too narrow”, or “flat and hypoplastic ala”; there were mostly noted positions, such as “deformation of upper part of nostril rim” and “long sill”, but the opinions of the evaluators differed considerably about the positions (high variability) (Tables 2.1. and 2.2.). It was not possible to calculate the Kappa coefficient in some cases because the numbers were too small, in which case the percentage agreement was taken into account. An intra-rater variability was calculated for evaluators A and B for measurements when cleft was on the left or the right side (Table 2.1.). In some positions, the Kappa coefficient was 0.4, i.e. low agreement, but in some positions Kappa was 1, i.e. excellent agreement. Intra-rater variability between A and B evaluations was similar. In most positions, the Kappa coefficient was between 0.5 and 1.0, which indicates moderate to very good reliability (Table 2.1.).

To determine the reliability of the method, Kappa coefficient was calculated between repeated measurements made by evaluators A and B as to whether the cleft was on the left side or the right side of the lip (inter-rater reliability; Table 2.2.). Measurements were performed twice with a one-week interval. The results were similar to intra-rater reliability, with excellent agreement Kappa values were in positions “symmetric nose”, “narrow sill”, “columellar base too narrow”, “flat and hypoplastic ala”. Kappa coefficient was
the lowest in positions which were noted mostly as “deformation of upper part of nostril rim” and “long sill”, indicating poor agreement between evaluators.

Table 2.1.

<table>
<thead>
<tr>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>%</td>
<td>Kappa</td>
<td>%</td>
<td>Kappa</td>
</tr>
<tr>
<td>1</td>
<td>Symmetric nose</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>2</td>
<td>Columella too short</td>
<td>90.7</td>
<td>0.8</td>
<td>90.7</td>
<td>0.8</td>
</tr>
<tr>
<td>3</td>
<td>Visible subluxation or deviation of the septum</td>
<td>97.7</td>
<td>0.9</td>
<td>86.1</td>
<td>0.6</td>
</tr>
<tr>
<td>4</td>
<td>Long sill</td>
<td>74.4</td>
<td>0.5</td>
<td>79.1</td>
<td>0.6</td>
</tr>
<tr>
<td>5</td>
<td>Narrow sill</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>6</td>
<td>Columellar base too wide</td>
<td>100</td>
<td>1.0</td>
<td>93.0</td>
<td>1.0</td>
</tr>
<tr>
<td>7</td>
<td>Columellar base too narrow</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>8</td>
<td>Insufficient wrapping of ala</td>
<td>90.7</td>
<td>0.8</td>
<td>88.4</td>
<td>0.8</td>
</tr>
<tr>
<td>9</td>
<td>Excessive wrapping of ala</td>
<td>97.7</td>
<td>0.9</td>
<td>76.7</td>
<td>0.5</td>
</tr>
<tr>
<td>10</td>
<td>Deformation of upper part of nostril rim</td>
<td>93.0</td>
<td>0.5</td>
<td>88.4</td>
<td>0.7</td>
</tr>
<tr>
<td>11</td>
<td>Poor position of alar cartilage</td>
<td>72.1</td>
<td>0.4</td>
<td>83.7</td>
<td>0.5</td>
</tr>
<tr>
<td>12</td>
<td>High position of ala</td>
<td>100</td>
<td>1.0</td>
<td>93.0</td>
<td>0.6</td>
</tr>
<tr>
<td>13</td>
<td>Low position of ala</td>
<td>86.1</td>
<td>0.2</td>
<td>97.7</td>
<td>0.8</td>
</tr>
<tr>
<td>14</td>
<td>Flat and hypoplastic ala</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>15</td>
<td>Sum</td>
<td>46.5</td>
<td>0.3</td>
<td>30.2</td>
<td>0.1</td>
</tr>
</tbody>
</table>

*NA – Kappa not applicable. Kappa 0.8-1.0 = excellent, 0.6-0.8 = good, 0.4-0.6 = medium, 0.2-0.4 = poor, < 0.2 = very poor agreement
Table 2.2.

Inter-rater variability between A and B evaluators after repeated measurements when cleft was positioned on left (sin) or right (dx) side

<table>
<thead>
<tr>
<th>No.</th>
<th>Position of modified Anastassov &amp; Chipkov (2003) scheme</th>
<th>sin 1</th>
<th>sin 2</th>
<th>dx 1</th>
<th>dx2</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td>%</td>
<td>%</td>
<td>%</td>
<td>%</td>
</tr>
<tr>
<td>1</td>
<td>Symmetric nose</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>2</td>
<td>Columella too short</td>
<td>74.4</td>
<td>0.2</td>
<td>79.1</td>
<td>NA</td>
</tr>
<tr>
<td>3</td>
<td>Visible subluxation or deviation of the septum</td>
<td>74.4</td>
<td>0.2</td>
<td>81.4</td>
<td>0.4</td>
</tr>
<tr>
<td>4</td>
<td>Long sill</td>
<td>76.7</td>
<td>0.5</td>
<td>86.1</td>
<td>0.7</td>
</tr>
<tr>
<td>5</td>
<td>Narrow sill</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>6</td>
<td>Columellar base too wide</td>
<td>93.0</td>
<td>NA</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>7</td>
<td>Columellar base too narrow</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>8</td>
<td>Insufficient wrapping of ala</td>
<td>65.1</td>
<td>0.2</td>
<td>67.4</td>
<td>NA</td>
</tr>
<tr>
<td>9</td>
<td>Excessive wrapping of ala</td>
<td>74.4</td>
<td>0.1</td>
<td>83.7</td>
<td>0.1</td>
</tr>
<tr>
<td>10</td>
<td>Deformation of upper part of nostril rim</td>
<td>69.8</td>
<td>0.4</td>
<td>65.1</td>
<td>0.3</td>
</tr>
<tr>
<td>11</td>
<td>Poor position of alar cartilage</td>
<td>79.1</td>
<td>0.4</td>
<td>74.4</td>
<td>0.3</td>
</tr>
<tr>
<td>12</td>
<td>High position of ala</td>
<td>86.1</td>
<td>NA</td>
<td>93.0</td>
<td>NA</td>
</tr>
<tr>
<td>13</td>
<td>Low position of ala</td>
<td>90.7</td>
<td>0.6</td>
<td>97.7</td>
<td>0.7</td>
</tr>
<tr>
<td>14</td>
<td>Flat and hypoplastic ala</td>
<td>100</td>
<td>1.0</td>
<td>100</td>
<td>1.0</td>
</tr>
<tr>
<td>15</td>
<td>Sum</td>
<td>32.6</td>
<td>0.2</td>
<td>41.9</td>
<td>0.2</td>
</tr>
</tbody>
</table>

*NA – Kappa not applicable. Kappa 0.8-1.0 = excellent, 0.6-0.8 = good, 0.4-0.6 = medium, 0.2-0.4 = poor, < 0.2 = very poor agreement

Nasal aesthetic evaluation with VAS was a subjective and variable method, both in the assessment of the left and the right side by one evaluator, as also between evaluators. Intra-rater variability in all measurements by VAS were statistically significant at p < 0.05, which means that each time the same evaluator assessed nasal aesthetic differently on the same case (Fig. 2.1.).
Intra-rater variability for measurements with VAS when the cleft was positioned on the left (sin) or the right (dx) side

* statistically significant difference ($p < 0.05$)

2.2. Nasal function

The difference of nasal flow values before and after decongestant was not significant in the study group; for further comparison, therefore, only values after decongestant were used. The mean value of nasal flow after decongestant in the non-cleft side was 330.6 cm$^3$/s and in the cleft side 169.1 cm$^3$/s at 150Pa in patients with complete unilateral cleft lip and palate. The difference between cleft side and non-cleft side was statistically significant ($p < 0.05$). The mean value of nasal flow was lower in children who had cleft on the right side compared with the cleft being positioned on the left side, but the difference was not statistically significant. In the study group, the mean value of nasal flow was in the range from no obstruction to slight obstruction according to Bachmann (1983) through the non-cleft side in 18 cases, but through the cleft side in 7 cases. Moderate obstruction as per Bachmann was seen in 6 cases.
through the non-cleft side and in 9 cases through the cleft side. Severe obstruction was observed in 6 cases through the non-cleft side, and in 14 cases through the cleft side (Fig. 2.2). In 4 cases mean nasal flow was worse through the non-cleft side, worse through the cleft side in 18 cases, and through both nostrils was similar in 8 cases. The severity of nasal flow was not associated with secondary nasal correction timing or additional nasal corrections, and their number in both cases means that nasal flow was in the range of no obstruction to severe obstruction on the basis of Bachmann (Fig. 2.2).

Fig. 2.2. **Degree of nasal obstruction for one nostril according to Bachmann (1983)**

1 – no obstruction, 2 – slight obstruction, 3 – moderate obstruction, 4 – severe obstruction

In the control group, the mean nasal flow through the right side was 308.4 cm$^3$/s, and through the left side 330.4 cm$^3$/s at 150 Pa after decongestant,
a difference that was not statistically significant \((p > 0.05)\). The difference of the mean nasal flow before and after decongestant in both the left and the right side in the control group was statistically significant \((p < 0.05)\).

The difference between nasal flow through the cleft side in patients with complete unilateral cleft lip, alveolus and palate and either the left or the right side in the control group was statistically significant \((p < 0.05)\) after decongestant (Fig. 2.3.), but the difference between the non-cleft side in study group and either the left or the right side in control group was not significant. Data of acoustic rhinomanometry showed statistically significant difference between the cleft side and the non-cleft side when compared to the control group.

![Graph](image)

**Fig. 2.3.** Mean nasal flow value at 150 Pa in control group and study group

\(d\) – decongestant (0.1% Xymelin)
2.3. Nasolabial symmetry in 3D photos

Method error between the first and second measurements in 3D photos for all variables did not exceed 3% of the total variance. No statistically significant difference was found between the first and second measurements (intra-rater variability), neither in study group and control group; therefore, the mean value of first and second measurement was used for further comparison of the measurements. In the study group, only alar wing length (al-PRN) was significantly different between the cleft and the non-cleft side, and it was shorter and flatter on the cleft side (p < 0.05; Fig. 2.4.)

![Anthropometric distances in study group](image)

Fig. 2.4. **Anthropometric distances in study group**  
* Statistically significant difference (p < 0.05)

The difference of nasolabial anthropometric landmarks, and distances in control group between the left and the right side, was not statistically significant. The difference between the cleft group and the control group was
statistically significant at all anthropometric distances, except for the lateral lip length to cupid’s bow on both sides (chR-cphR, chL-cphL; Fig. 2.5.). The nasolabial appearance of acceptable symmetry after cleft lip and nose reconstructive surgery was achieved in all patients.

Fig. 2.5. Anthropometric distances in mm between study and control group
* not a significant difference (p > 0.05)

In the study group the symmetry index between the cleft and the non-cleft side in relation to 100 (perfect) symmetry, a statistically significant difference was found between length of the red lip from crista philtrum to midline (cph-cM), the nose lateral length (al-N) and the alar wing length (al-PRN). The difference between other distances was not statistically significant (Table 2.3.).
The postoperative results were satisfactory in patients with complete unilateral cleft lip, alveolus and palate according to Li et al. (2010), because the mean symmetry index was in the range of 95 to 110%. They were considered satisfactory if nasolabial symmetry index was in this range 90-110% (Li et al., 2010). In the control group, the difference of symmetry index between the left and the right side compared to 100 was statistically significant in nostril height (nl-nd) (Table 2.3.).

<table>
<thead>
<tr>
<th>No.</th>
<th>Symmetry index</th>
<th>Description</th>
<th>The mean value of symmetry index</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td>Study group</td>
</tr>
<tr>
<td>1</td>
<td>cphL−chM ×100</td>
<td>Length of red lip from crista philtrum to midline</td>
<td>110*</td>
</tr>
<tr>
<td>2</td>
<td>chL−cphL ×100</td>
<td>Length of red lip to the crista philtrum</td>
<td>100</td>
</tr>
<tr>
<td>3</td>
<td>SbalL−cphL ×100</td>
<td>Length of white lip</td>
<td>97</td>
</tr>
<tr>
<td>4</td>
<td>nlL−ndL ×100</td>
<td>Nostril height</td>
<td>98</td>
</tr>
<tr>
<td>5</td>
<td>alL−Sn ×100</td>
<td>Width of the nostril at the base</td>
<td>100</td>
</tr>
<tr>
<td>6</td>
<td>alL−N ×100</td>
<td>Nose lateral length</td>
<td>98*</td>
</tr>
<tr>
<td>7</td>
<td>alL−PRN ×100</td>
<td>Alar wing length</td>
<td>95*</td>
</tr>
</tbody>
</table>

*Statistically significant difference (p < 0.05)

Comparing symmetry index between the study and the control group, statistically significant difference was found in the length of white lip (Sbal-cph), nose lateral length (al-N) and alar wing length (al-PRN) (Table. 2.4.).
Table 2.4.

<table>
<thead>
<tr>
<th>No.</th>
<th>Symmetry index</th>
<th>Description</th>
<th>Unpaired t test (p &lt; 0.05)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>cphL − chM/cphR − chM × 100</td>
<td>Length of red lip from crista philtrum to midline</td>
<td>p = 0.15</td>
</tr>
<tr>
<td>2</td>
<td>chL − cphL/chR − cphR × 100</td>
<td>Length of red lip to the crista philtrum</td>
<td>p = 0.22</td>
</tr>
<tr>
<td>3</td>
<td>SbalL − cphL/SbalR − cphR × 100</td>
<td>Length of white lip</td>
<td>p = 0.03</td>
</tr>
<tr>
<td>4</td>
<td>nlL − ndL/nlR − ndR × 100</td>
<td>Nostril height</td>
<td>p = 0.29</td>
</tr>
<tr>
<td>5</td>
<td>alL − Sn/alR − Sn × 100</td>
<td>Width of the nostril at the base</td>
<td>p = 0.48</td>
</tr>
<tr>
<td>6</td>
<td>alL − N/alR − N × 100</td>
<td>Nose lateral length</td>
<td>p = 0.003</td>
</tr>
<tr>
<td>7</td>
<td>alL − PRN/alR − PRN × 100</td>
<td>Alar wing length</td>
<td>p = 0.002</td>
</tr>
</tbody>
</table>

Correlation between nasal function and nasolabial symmetry was set in patients with complete unilateral cleft lip, alveolus and palate. The relationship between postoperative nasal symmetry and nasal function was not proven. However, nasal flow through the non-cleft side was dependent on total nose width (alR-alL) (p = 0.04), the wider nose, the worse nasal flow; but nasal flow through the cleft side nose width was unaffected. Nasal flow through the cleft side was statistically significant in correlation with nose dorsum length (PRN-N) (p = 0.04). The longer nose dorsum, the better nasal flow through the cleft side, but nasal flow through the non-cleft side nose dorsum length was unaffected.
3. DISCUSSION

3.1. Study Sample

Cleft lip and palate is the most frequent congenital craniofacial deformity. Cleft lip and palate irregularities vary greatly in terms of cleft width and other characteristics. Patients with complete unilateral cleft lip and palate are mostly used in studies due to the uniform deformation at the correct diagnosis; however, cleft width can be different. Therefore, to evaluate postoperative results as objectively as possible, it was decided in both parts of the study to include only patients with complete unilateral cleft lip, alveolus and palate, who are always accompanied with typical nasal deformity. Rhinoplasty for the patients with clefts is very challenging, making it difficult to achieve good functional and aesthetic results. Nasolabial asymmetry is remarkable in patients with complete unilateral cleft lip, alveolus and palate.

In the first part of the study, 27 children with complete unilateral cleft lip, alveolus and palate were included for nasal aesthetic evaluation in 2D photos. However, several 2D photos were of poor quality, of inaccurate position at the children’s ages of 3 or 5 years, and were not valid for nasal aesthetic evaluation. When data were processed statistically, we were faced with the difficulties of analysing them due to their being small groups. Initially the aim of the first study was to compare nasal aesthetic results in children who had only primary cleft lip, and nose repair with children who had repositioning of the nasal septum during primary cleft lip and nose repair. Unfortunately, nasal aesthetic evaluation in 2D photos showed high variability of agreement; therefore, it was not possible to compare the groups due to unreliable results.

In the second part of the study, 35 children aged 10-18 years with complete unilateral cleft lip, alveolus and palate were included,
of which 30 (86% responsiveness) responded to recall. In Latvia, the mean prevalence of facial clefts is 1.2-1.4 to 1000 live birth every year, of which only 36% are cleft lip and palate (Akota et al., 2001). The incidence of complete unilateral cleft lip, alveolus and palate is even lower. Therefore, to provide a sufficiently large study group for statistically reliable results, the patient age range was selected from 10 to 18 years. Bell et al. (2014) point out that 10 years in patient age is a milestone in the management of clefts, when it is then the time to assess disparity of the child’s face, as well as the time to consider the need for further aesthetic and functional surgical corrections.

With 10 year-old children it is possible to get good communication and collaboration for different investigations, such as 3D stereo-photogrammetry and active anterior rhinomanometry. In this study, we encountered no difficulty in carrying out qualitative examinations with 3D stereo-photogrammetry or active anterior rhinomanometry. The upper limit of patient age was selected at 18 years due to the annual control in RSU IS cleft lip and palate centre and completion of the residual cleft deformation corrections.

Patients included in nasal aesthetic evaluation in 2D photos and nasolabial symmetry measurement in 3D photos were mostly the same. Children born after 2004 that were under 10 years old were not included in the 3D study; however, to increase the size of the study group, children born in 1994 were included. Over the 10 years (1994-2004), 35 children with complete unilateral cleft lip and palate were born in Latvia, of which 10 children were born in the year 1994, but only 1 child was born in the year 2002 and 2004. Mercado et al. (2011) points out that optimal sample size for a multicentre study should be 30-40 participants per centre. Even in a study in Great Britain had to increase the range of included patient age to 5 years to get a sufficiently large study group, which is common phenomenon in studies of patients with different cleft types (Bugaighis et al., 2014a,b).
In the control group, 35 healthy children of 10 years of age were included. An age limit of 10 years was selected because it was the lowest border of included patient age in study group. It is also because in 2014, RSU IS started a new international facial growth evaluation study where 10 year old healthy children were randomly selected. It was possible to include the first 35 participants from that study as a control group. The control group number was made up to the same the number in the study group, i.e. 35 children. It is difficult to find a control group with healthy individuals due to social and ethical aspects, but especially if certain investigations need be performed which required active collaboration of the child, for example, in active anterior rhinomanometry. In a literature review of nasal aesthetic evaluation methods in 143 selected articles, healthy children of different kind of ages were used as a control group in only 23 articles. Usually in these articles, the 2D photo was taken of the children (McComb & Coghlan, 1996; Kyrkanides et al., 1996; Kim et al., 2004; Daelemans et al., 2006; Russell et al., 2009; Meazzini et al., 2010; Fudalej et al., 2012; Russell et al., 2014) or 3D photo (Duffy et al., 2000; Hood et al., 2003; Hood et al., 2004; Ayoub et al., 2011; Zreaqat et al., 2012; Bell et al., 2014; Bugaighis et al., 2014a; Bugaighis et al., 2014b; Dadáková et al., 2016; Wu et al., 2016), in one case a video record was performed (Trotman et al., 2007), in one case facial gypsum casts (Russell et al., 2000), in one case clinical examination (Oosterkamp et al., 2007), and in 2 articles already available CT scans after facial traumas were used as a control group (Miyamoto & Nakajima, 2010; Miyamoto et al., 2012).

The control group used in our study was unique, not only because 3D photo was performed, but also nose function was assessed by active anterior rhinomanometry.
3.2. Surgical procedures

In last 20 years, much attention has been paid to primary nasal correction during primary cleft lip repair. Torre et al. (2000) believes that early nasal correction leads to more symmetric growth of the nasal cartilages. Also, Byrd and Salomon (2000) consider that primary nasal correction in patients with cleft-related nose deformity provides a more symmetric nose and better overall appearance in early life, which increasingly reduces physiological trauma in early school ages. However, Guyuron (2008) highlights that although early surgery reduces psychological problems, it does not exclude possible nasal deformity and the necessity for further nasal correction. Today, most nasal deformities are corrected during primary cleft lip repair. Also during last ten years, RSU IS cleft lip and palate centre had paid much attention to nasal septum reposition during primary cleft lip and nose repair. We have noticed that, after nasal septum reposition during primary cleft lip repair, nose growth was more symmetric, and that secondary nasal correction before school age was unnecessary. Unfortunately, many more years and patients are needed to prove that early nasal septum repositioning leads to more symmetrical nose growth and fewer operations to achieve desirable results.

In today's society, aesthetics is increasingly important. School is the first place where children meet their peers and start socializing. Taunting was reported to be a common phenomenon for children with clefts (Lorot-Marchand et al., 2015). Gosain and Fathi (2009) believe that secondary nasal correction in children with complete unilateral cleft lip and palate is often indicated before school age (6-8 years). At this age, social interactions of patients increase significantly and they begin to experience pressures from peers that create their self-image (Gosain & Fathi, 2009).
In the case of remarkable nasal deformity, patients with complete unilateral cleft lip and palate were having secondary rhinoplasty at an early school age (range 6-10 years) at our centre.

3.3. Nasal aesthetic and function evaluation methods

Aesthetics cannot be simply assessed by objective measures (Russell et al., 2009). The evaluation of facial forms can be classified into 2 groups – objective methods using measurements, and subjective methods using a scoring system based on examination findings and other items (Yamada et al., 1999). The most popular method remains 2D photography, which is simple, widely available, economical, and has been used for a long time with a large database. Therefore, 2D photos were also used in our study. Since the Eurocleft 1996-2000 project, 2D photogrammetry was set as a standard for data collection, making later comparison of data between European cleft centres possible (Shaw et al., 2001). It was decided to use retrospective nasal aesthetic evaluation in 2D photos, which was time-consuming because it was necessary to prepare materials in advance, train the evaluators, and provide continuously standardized conditions over several consecutive weeks. Therefore, it was necessary during the study to involve several experts, who took part in 2D photo randomisation and evaluation, so that inter-rater variability could be calculated. Nasal aesthetic evaluation in patients with clefts is complex; therefore 2 methods were used in 2D photos, i.e. the adapted and modified Anastassov and Chipkov (2003) scheme, and the visual analogue scale (VAS). The Anastassov and Chipkov (2003) clinical evaluation scale was specially adapted for nasal aesthetic evaluation in 2D photos. The same Anastassov and Chipkov (2003) scale was also used in the Baltic Cleft Network multicentre study. The results of the Baltic Cleft Network multicentre study
have not yet been published. Also, Zaleckas et al. (2011) used a similar evaluation scale in their study, where lip and nose aesthetic were evaluated. In the first part of our study, we were confronted with poor quality 2D photos, were often in inappropriate positions and blurred, or were simply unavailable. Mercado et al. (2011) in their multicentre study found that none of centres had used any kind of stabilizer during 2D photography to standardize the position of the head. Consequently, it was impossible to compare objectively surgical outcomes between different centres. In many centres, 2D photos were not complied in columna projection, which is highly necessary for nasal aesthetic evaluation (Mercado et al., 2011).

Discussions on the aesthetic evaluation of the nose in cleft lip and palate patients remains problematical. Many different evaluation methods have been published, which indicates that none has proved adequate. In many articles, different combinations of measurements and methods have been used for nasal aesthetic evaluation. In their review, Al-Omari et al. (2005) noted no single reliable method for aesthetic evaluation of the nose. However, 3D images had already become the most highly rated and remain so (Al-Omari et al., 2005). The recent trend for nasal aesthetic evaluation is for 3D laser scanning of face with 3D computer-assisted coordinates of soft tissue facial landmarks and angles, which is more objective method and can involve a smaller study group. Further 3D analysis with more patients could lead to the possible development of new surgical techniques that will improve facial symmetry (Stauber et al., 2008). Several authors have concluded that 3D stereo-photogrammetry was a more non-invasive, accurate, and achievable method for assessing facial form and surgical change. Nasal symmetry can be quantified and measured reliably with this tool (Devlin et al., 2007; Mori et al., 2005). However, this method is technologically more expensive and might not be available in some regions (Russell et al., 2011). Considering its advantages, accessibility and experience
in assessing nasal aesthetics in 2D photos, it was decided that in second part of the study 3D stereo-photometric examination of facial topographic surface would be used to compare nasolabial symmetry in children with complete unilateral cleft lip, alveolus and palate. This tool is simple, easy to use, patient friendly, non-invasive, quick and can be used on child patients.

Neither is there consensus about measurements. Despite many different anthropometric landmarks and measurements used for the aesthetic evaluation in patients with cleft lip and palate (Bagante & Akota, 2015), there are some landmarks and measurements which are used in most studies, e.g. the nasal tip, alar points and height of nares (Farkas et al., 1993; Duffy et al., 2000; Yamada et al., 2002; Ferrario et al., 2003; Hood et al., 2004; Devlin et al., 2007; Weinberg et al., 2009; van Loon et al., 2010; Ayoub et al., 2011; Zreaqat et al., 2012). In this study, we used the same anthropometric landmarks and distances as Farkas et al. (1993), with the modifications of van Loon et al. (2010) and Hood et al. (2004). In our opinion, this set of landmarks and distances could describe the shape of the nasolabial complex accurately in 3D photographs. This set of landmarks and distances might be proposed to assess routinely nasolabial appearance after surgery.

Marking anthropometric landmarks in 3D photos is quite time-consuming, therefore they were set as low as possible, but had to be enough for routine evaluation of nasolabial symmetry in clinical praxis. Like van Loon et al. (2010), we were confronted with accuracy in the subnasal region, the nostrils being the region of error because of the complex anatomy and the inability of the camera to capture dark holes perfectly. For standardisation, an automatized landmark pointer might be more promising. Automated methods already have been introduced, but they are not widely applied in practice. Mishima et al. (2002) used an accurate quantitative method for measuring external nasal forms from plaster facial casts to identify
noted that surprisingly small attention was paid to functional aspects of nasal deformation and functional results after reconstructive surgery, although it is well known that a significant number of patients with complete unilateral cleft lip, alveolus and palate suffer from nasal airway obstruction, which can have a significant effect on nasal physiology and quality of life (Cohen et al., 2003). Rhinomanometry is the most frequently used method to evaluate nasal quantitative function, measuring nasal resistance and minimal cross-sectional area (Fukushiro & Trindade, 2005). Andre et al. (2009), after analysing evidence-based studies, concluded that there is still no correlation between
objective rhinomanometry measurements and individual subjective nasal airway obstruction. The most frequent nasal airway obstruction is deviation of the septum. Since nasal septal deviation is a commonly observed anatomical variation in healthy individuals, clinicians must often decide whether it is clinically relevant to the symptoms of nasal airway obstruction. This means that many individuals with septal deviation do not have difficulties with nasal breathing (Kim et al., 2014). Although there is no consensus in this area, several authors used rhinomanometry before and after nasal correction, and concluded that rhinomanometry is a quite objective method for quantitative nasal function evaluation (Sandham & Murray, 1993; Dusková et al., 2002; Trindade et al., 2009; Huempfner-Hierl et al., 2009). Mani et al. (2010) stated that, not only objective measurements are important, but also individuals’ subjective complaints. This was also observed in our study, as many patients with complete unilateral cleft lip, alveolus and palate did not complain of respiratory distress, although rhinomanometric data showed obstructive signs. While Kunkel et al. (1997) found that for patients with clefts, while having many uncomfortable investigations during long-term multidisciplinary treatment, it is very important to use non-invasive investigation methods such as rhinomanometry. In our study, we did not have difficulties using rhinomanometry in children with complete unilateral cleft lip, alveolus and palate, and the control group. Children could easily understand and accomplished the task.

3.4. Interpretation of the study results

The sum of the results of nasal aesthetic evaluation, using the adapted and modified Anastassov and Chipkov (2003) evaluation scheme, were from 0 to 3 points. Most noted were positions such as “deformation of upper
part of nostril rim” and “long sill”, but there was high variability. Intra-rater variability between 2 evaluations was relatively low, indicating good reliability; but difference between 2 raters had high inter rater variability, indicating the subjectivity of the method.

Perhaps the number of patients included in the first part of the study and the potential 2D photo’s flaws affected the statistical confidence. Although evaluators were prepared and introduced to the adapted Anastassov and Chipkov (2003) nasal aesthetic evaluation scheme, individual positions could be interpreted differently. Nasal aesthetic evaluation in 2D photos in studies with larger patients’ groups showed good reliability; for example, Pitak-Arnnop et al. (2011) followed 50 patients, but Anastassov and Chipkov (2003) nasolabial aesthetic assessed clinically on patients, that excluded possible error of 2D photo. Also, Anastassov performed measurements only by himself, once, that excluded calculation of intra-rater and inter-rater variability.

Nasal aesthetic evaluation with VAS was very subjective, with variable results between evaluators, and also between the first and second measurements of one evaluator. Reliability of VAS evaluation is influenced only by human element, i.e. it is based on the principle “to like” or “to dislike”. Therefore, VAS has rarely been used in studies, and usually together with some other aesthetic evaluation method. VAS is a good method if non-specialist opinion is assessed; for example, patient self-assessment as Sinko et al. (2005) did in their study, where VAS was used to compare patients’ self-assessment with the assessment of the surgeons (Sinko et al., 2005). Oosterkamp et al. (2007) also used VAS for patients’ self-assessment of nasal aesthetic and function. Oti et al. (2014) used VAS to assess nasal aesthetic by parents of patients, laypersons and surgeons. However, there are studies where VAS showed good statistical reliability (Bongaarts et al., 2008; Desmedt et al., 2015; Stebel et al., 2016). The visual analogue scale could probably be used to
determine patient self-assessment and parent satisfaction with postoperative results, since it is convenient, inexpensive and easy to understand.

The study results on rhinomanometry were similar to other studies in the literature where nasal function of patients with clefts has been compared with the control group. Nasal flow in patients with complete unilateral cleft lip, alveolus and palate was reduced compared to healthy children (Kunkel et al., 1997; Fukushiro & Trindade, 2005; Mani et al., 2010). Rhinomanometric studies show an increase resistance due to septal deviation on the cleft side and decreased resistance on the non-cleft side. However, in 4 cases in children with complete unilateral cleft lip, alveolus and palate nasal flow through the cleft side was better than through the non-cleft side, inspite of nasal septal deviation, a condition called paradoxical nasal obstruction. This is explained by the reduced airflow on the contralateral side caused by an obstruction resulting from compensatory hypertrophy of the inferior turbinate (Ridgway et al., 2011). The disturbed nasal flow may also be related not only to the position of the nasal septum, but to other otorhinolaryngological organ disorders. Reiser et al. (2011) points out that in patients with clefts, internal nasal deformities, such as a deviated septum, hypertrophied turbinates, nasal polyps, posterior choanae deficiencies, adenoid tissue, tonsils, and narrow and/or decreased size of nares or intranasal strictures, can all cause obstruction of the nasal airway. According to rhinomanometric data in children with breathing disturbances, it could probably be suggested some additional nasal correction procedures, but it is not clear how much they might improve nasal breathing. Cohen et al. (2003) concluded that better functional and aesthetic results were obtained in patients who had undergone fewer surgical procedures. This was explained as the result of tissue contracture and the relative lack of elasticity of scarred tissue during breathing (Cohen et al., 2003). These authors also point out that in patients with complete unilateral cleft lip, alveolus and palate prior to nasal correction, it is
important to keep in mind previous procedures and scarring that might have produced additional iatrogenic deformities (Cohen et al., 2003). In our study, a direct relationship between the nasal airflow and the number of operations was not proven.

The study results regarding anthropometric measurements in 3D photos showed that there was no significant difference in measurements between both sides in patients with complete unilateral cleft lip alveolus and palate except for alar wing length (al-PRN), which indicates that nasolabial symmetry had been achieved after secondary nasal corrections before school age, as also for those children for whom secondary nasal correction was considered unnecessary. The difference of nasolabial anthropometric landmarks and distances in control group between the left and the right side was not statistically significant, thus the nose could be considered symmetrical. The difference between the cleft group and the control group was statistically significant in all anthropometric distances except the lateral lip length to cupid’s bow on both sides (ch-cph). The youngest participants in study group were 10 years old, the mean age being 14.7 (range 10-18 years), but the control group consisted only of 10 year old children, which could have made some difference in the results. However, we consider that it was possible to assess the difference in measurements between the cleft and the non-cleft side compared to the difference between both sides in the control group. Our results confirm the findings of others (Duffy et al., 2000; Yamada et al., 2002; Ferrario et al., 2003; Hood et al., 2004; Ayoub et al., 2011; Okawachi et al., 2011; Zreaqat et al., 2012).

Although statistically significant difference between the cleft and the non-cleft side in the study group was detected only in alar wing length (al-PRN), the symmetry index showed a statistically significant difference in symmetry of length of red lip from crista philtrum to midline (cph-cM), nose lateral length (al-N) and alar wing length (al-PRN). Also in the control group,
the symmetry index showed statistically significant asymmetry of nostril height (nl-nd), which went undetected during symmetry analysis in absolute measurements. Surprisingly, symmetry of nostril height in the study group, was achieved better than in the control group regarding children without cleft lip and palate. The symmetry index indicates more accurate symmetry, but it is an artificial mathematically created index. However, as Bell et al. (2014) point out that it is already generally accepted that the face is not completely symmetrical. Also Peck H. and Peck S. (1970) concluded in their study that a completely symmetrical face is not aesthetic during evaluation of facial symmetry of beauty contest winners, professional models and other beautiful women. Authors divided the face along the median plane and reprocessed photographically to yield composite faces. The right side was paired with its mirror image to yield one face and the left side was paired with its mirror image to produce another. After evaluation all 3 faces – the original and 2 artificial examples, the authors concluded that very small facial asymmetry can characterise an aesthetically attractive face (Peck H. & Peck S., 1970). However, the symmetry index is very helpful for nasolabial symmetry comparison of the study group and the control group because there is variation in age and stature, excluding direct comparison of anthropometric distances. Also, the symmetry index helps to determine typical post-operative nasolabial asymmetries in patients with clefts, and could be useful in evaluating surgical post-operative results. The Li et al. (2010) review states that many studies have analysed asymmetry of normal people and have shown that slight facial asymmetry is a natural phenomenon, absolute symmetry being uncommon. The authors also indicated that the aesthetic indexes used for evaluation of nasolabial region in healthy people was in a range of from 90 to 110%, and this range was considered to be giving a symmetric face. If the nasolabial symmetry index range was from 90 to 110% in patients with clefts, they concluded that
the surgery had been successful (Li et al., 2010). If we take into account their proposed symmetry index interpretation, then all patients with complete unilateral cleft lip alveolus and palate involved in our study had successful surgery outcomes because mean symmetry index was in a range of from 95 to 110%; in the control group, the index was in a range of from 96 to 103%.

Russell et al. (2009) point out that the clinical significance of the variables that are statistically different remains unclear. One guideline for clinical significance could be based on Lu (1965), who report that asymmetries greater than 3% are clinically detectable (Russell et al., 2009). Othman et al. (2014) propose that 5 mm asymmetry of face is an appropriate threshold to indicate clinical relevance. This means that a clinician can detect asymmetry of the face of patients with unilateral cleft lip and palate only if the difference is more than 5 mm that of normal patients (Othman et al., 2014).

Aesthetics might be the interpretation of an accumulation of many facial features, and it is possible that aesthetics cannot be assessed with specific objective measurements in patients with complete unilateral cleft lip, alveolus and palate (Russell et al., 2009). The degree of nasal airway obstruction varies and does not necessarily directly correlate with the degree of nasal deformity (Cohen et al., 2003). It was also observed in our study that there was no convincing relationship between nasolabial symmetry in 3D photos and nasal function.
CONCLUSIONS

1. Nasal aesthetic evaluation with the modified Anastassov and Chipkov (2003) evaluation scale had high intra-rater and inter-rater variability. The use of this scoring scale in 2D photos for a selected patient group was subjective and was not applicable for group comparisons.

2. Nasal aesthetic evaluation with the visual analogue scale (VAS) in 2D photos for a selected patient group was a subjective method.

3. Nasal flow after rhinoplasty through the cleft side was heavily obstructed, and through the non-cleft side was slightly obstructed in patients with complete unilateral cleft lip, alveolus and palate. In the control group, nasal flow through both the right and the left sides was only slightly obstructed. Rhinomanometry was a convenient and objective method for evaluating nasal function in children over 10 years of age.

4. Nasolabial symmetry was restored in children with complete unilateral cleft lip, alveolus and palate after reconstructive nose correction at early school age in 3D photo evaluation; however, symmetry between the study group and the control group differed. Symmetry index was helpful for comparing nasolabial symmetry between the study and the control groups.

5. 3D photographs with the proposed set of anthropometric landmarks for evaluation of nasolabial appearance seems to be a convenient and an accurate non-invasive way to follow patients after surgery in selected patients group, whereas nasal aesthetic evaluation in 2D photos with the chosen methods gave subjective results. Further 3D photographs should be used for evaluating nasal appearance.
PRACTICAL RECOMMENDATIONS

1. 3D photogrammetry is recommended for routinely evaluating postsurgical results in children with complete unilateral cleft lip, alveolus and palate from 10 years of age.

2. The proposed set of anthropometric landmarks and distances for evaluation of nasolabial symmetry might be routinely used in clinical practice.

3. Active anterior rhinomanometry is recommended for routinely evaluating nasal function in children with complete unilateral cleft lip, alveolus and palate from 10 years of age.
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