



Cleft palate, treatment and complications

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ABSTRACT

Introduction: The purpose of this study is to determine the incidence of velopharyngeal insufficiency (VPI), fistulae and recurrence development in patients seen by the Isfahan Cleft Care Clinic and also determine the association of gender, age at repair, and cleft type with the incidence of each.

Materials and Methods: In this retrospective study, 320 children who had undergone a primary cleft palate surgery and who had come to the cleft lip and palate in Isfahan in 2010-2017 were studied based on the medical records and information about each patient. Then all patients were evaluated by standard speech assessment methods for severity of hypernasalitis and screened for clinical manifestations of fistula and recurrence.

Results: According to the results of this study, the incidence of velopharyngeal insufficiency after initial repair was 78.1% and most of these patients had severe form. The results also showed that by increasing age at surgery also increased the intensity of velopharyngeal insufficiency, and the severity of this complication has nothing to do with gender. In the case of fistula and recurrence of cleft showed that, in patients who are undergoing the new procedure to be significantly less than other patients.

Conclusion: According to the study, palatoplasty complications such as velopharyngeal insufficiency, fistula, and recurrence were less common in patients treated with microsurgery. Therefore, it can be concluded surgical repair of cleft palate should be performed before 12 months ages and in microsurgery methods. It seems that follow up of these patients after surgery for monitoring of speech complications is necessary.

Keywords: Cleft palate; Velopharyngeal insufficiency; Fistula; Palatoplasty; Microsurgery.

Introduction

Congenital anatomical disorders of the mouth and palate, including cleft palate, is one of the most common causes of disruption of speech skills. The harm caused by communication disorders threatens all children born with cleft lip and palate. Because comprehensibility of speech decreases in these patients under the influence of hypernasalitis and increased openness of the palatopharyngeal valve. Velopharyngeal sufficiency is

achieved during speaking, when the soft palate approaches the posterior pharyngeal wall, it also prevents air and fluid from returning to the nasal cavity and it helps the air inside the mouth to make speech sounds [1]. The goal of repairing the cleft palate is to create a healthy and perfect palate for speaking and eating without difficulty. Surgery should lead to the development of sufficient palatopharyngeal function, while ensuring the least disturbance in max-

illary bone growth. Fistula and velopharyngeal insufficiency (VPI) are two unpleasant complications after surgery [2,3]. Velopharyngeal insufficiency is a structural defect in which the shortening and irregularity of the soft palate prevents its complete closure against the posterior wall of the pharynx while speaking [4]. The rate of VPI reports after surgery is about 10-40% in the articles [1]. Various results of cleft palate repair depend on various factors such as severity and type of cleft palate, surgical technique, and age of the patient during recovery. All of these factors affect the success rate of cleft palate repair [5].

Contrary to the relatively long history of cleft palate surgery, there is little consensus on the best techniques and even less consensus on the best surgical time [6]. And there are currently no standardized protocols to resolve the problem of time to repair the cleft palate to achieve optimal speech and prevent Maxillary abnormal growth [7]. In Iran, due to recent advances in the management of patients with cleft palate and the timing of treatment interventions, it seems that the number of patients with severe speech problems has decreased; however, there are few studies and information in this regard. In this regard, in order to increase the awareness of the members of the cleft palate team, especially surgeons and speech and language pathologists, we decided to do a study on this issue in the community of patients with cleft palate.

Material and Methods

In this retrospective study, 320 children who had undergone a primary cleft palate surgery and who had come to the cleft lip and palate in Isfahan in 2010-2017 and had inclusion criteria, were studied based on the medical records and information about each patient includes the patient's gender, age in time of surgery, surgical technique, type of cleft palate, surgeon's name and parent's call number, and post-surgical complications (including velopharyngeal insufficiency, fistula and relapse), from patients' medical history was extracted. In the case of patients with varying time of soft and hard cleft palate repair, the age of their soft palate cleft surgery was recorded. Then, we contacted the parents of these patients, and after justifying them about the goal of the study, they were invited to accompany their child to the cleft lip and palate clinic at the Faculty of Rehabilitation Sciences of Isfahan University of Medical Sciences. After referral to the speech and language pathologist, speech and voice resonance were evaluated by screening tests and the severity of hypernasalitis was determined. Voice resonance evaluation

was performed using "Speech screening test in unofficial Persian language". This test contains a list of words and sentences that are susceptible to pressure that the patient is asked to repeat. Since perceptual evaluations are more useful and non-invasive in clinical assessment for children, and the perceived quality of speech is the main criterion for determining medical decisions, this type of assessment is the golden standard and the most common way of evaluating speech [8,9]. In the current study, the CAPS-A speech evaluation test was performed using a global-Persian version of its system, which has acceptable reliability and validity [8].

The grading of hypernasalitis at the single-word level was characterized by grading from 0 to 3 by examining the single-word section and the explanations corresponding to each degree of severity: normal (0), mild (1), moderate (2), severe (3). The physical examination of the palate was performed by the physician to detect the complication of the fistula after surgery. It should be noted that in this study, the old method refers to the method in which the repair of the cleft palate was performed without separate reconstruction of the muscles and in the form of blind. But in the new technique, the muscles of the palate are repaired separately using a microsurgery.

At the end of this study, in order to improve the treatment process and develop proper speech skills of patients and prevent communication deficiencies, those patients who suffered from velopharyngeal insufficiency and speech impairment were referred to speech and language pathologists for the treatment of speech disorders. After collecting and eliminating the defect, the data were entered into the computer and analyzed with the updated version of SPSS software. Mann-Whitney and Kruskal-Wallis statistical tests and Spearman correlation coefficient were used to compare the groups of patients. The results were expressed in terms of tables and charts and were reported for quantitative variables with central indexes and statistical dispersion. In this study, P value less than 0.05 was considered statistically significant. It should be noted that this research project has been approved by the Ethics Committee of Najafabad Islamic Azad University, IR.IAU.NAJAFABAD. REC.1395.006.

Results

The age range of patients was between 3 and 12 years old with a mean of 6.9 and a standard deviation of 1.9 years (Table 1). 171 (53.4%) of the children were boys and 149 (46.6%) were girls. Frequency distribu-

tion of sex, type of cleft palate and surgical technique are presented in Table 2. 25.9% of children had fistula complications and 7.5% had recurrence after surgery (Table 3). Velopharyngeal insufficiency was severe in 44.4% of children. It should be noted that from 320 children examined, 250 children (78.1%) had post-operative velopharyngeal insufficiency (Table 4).

Spearman correlation coefficient showed that the velopharyngeal insufficiency severity was not significantly correlated with the current age of children ($P=0.41$), but had a direct correlation with the age of surgery ($P=0.007$). In other words, with increasing age of surgery, the intensity of velopharyngeal insufficiency after surgery increased (Table 5). The Mann-Whitney test showed that the intensity of velopharyngeal insufficiency was not significantly different between boys and girls ($P=0.60$). (Table 6, Diagram 1). The Mann-Whitney test showed that the intensity of velopharyngeal insufficiency in children undergoing new surgical techniques was significantly lower than those who operated with old surgical techniques ($P<0.001$). (Table 7, Diagram 2).

Kruskal-Wallis test showed that intensity of velopharyngeal insufficiency had a significant relationship with the type of cleft palate ($P=0.001$). So that the number of severe velopharyngeal insufficiency in children with sub mucosal-cleft palate type (SMCP) was significantly higher than the type of Hard/Soft-cleft palate type (HSCP), in the type of HSCP type was significantly more than the type of Bilateral cleft lip/palate (BCLP) and the type of BCLP cleft was significantly more than two other cleft palate (Table 8). Chi-square test showed that the frequency of fistula ($P<0.001$) and recurrence ($P=0.004$) in children undergoing a new surgical technique were significantly lower than that of children with the old surgical technique (Table 9, Diagram 3). Chi-square test showed that there was a significant relationship between the frequency of fistula and the type of cleft palate ($P=0.008$), but the Chi-square test with correlation coefficient showed that the frequency of recurrence with the type of cleft palate was not significant ($P=0.46$) (Table 10).

| Variable | Average | Standard deviation | Minimum | Maximum |
|---------------------------------|---------|--------------------|---------|---------|
| Age (years) | 6.9 | 1.9 | 3 | 12 |
| Age at time of surgery (months) | 15.5 | 12 | 6 | 88 |

Table 1. Statistical indexes of patients' age and age at time of surgery.

| Variable | Type | Number | Percentage |
|------------------------------|------------------------------------|--------|------------|
| Sex | Boy | 171 | 53.4 |
| | Girl | 149 | 46.6 |
| Type of cleft lip and palate | Soft cleft palate (SCP) | 74 | 23.1 |
| | Hard/Soft cleft palate (HSCP) | 80 | 25 |
| | Bilateral cleft lip/palate (BCLP) | 63 | 19.7 |
| | Unilateral cleft lip/palate (UCLP) | 83 | 25.9 |
| | (SMCP) Sub mucosal cleft palate | 20 | 6.3 |
| Surgical Technique | Old | 192 | 60 |
| | New | 128 | 40 |

Table 2. Frequency distribution of sex, type of cleft palate and surgical technique used in patients.

| Intensity of velopharyngeal insufficiency | Number | Percentage |
|---|--------|------------|
| Normal | 70 | 21.9 |
| Mild | 71 | 22.1 |
| Moderate | 37 | 11.6 |
| Severe | 142 | 44.4 |

Table 3. Frequency distribution of intensity of velopharyngeal insufficiency after surgery in children.

| Complication | Number | Percentage |
|--------------|--------|------------|
| Fistula | 83 | 25.9 |
| Recurrence | 24 | 7.5 |

Table 4. Frequency distribution of complications of fistula incidence and recurrence in children.

| Variable | Intensity of velopharyngeal insufficiency | |
|------------------------|---|-------|
| | R | P |
| Age | 0.046 | 0.41 |
| Age at time of surgery | 0.151 | 0.007 |

Table 5. Spearman correlation coefficients between intensity of velopharyngeal insufficiency and age and age at time of surgery.

| Intensity of velopharyngeal insufficiency | Gender | | P-value |
|---|--------------------------|-------------------------|---------|
| | Girl Number (percent) | Boy Number (percent) | |
| Normal | 35 (23.5) | 35 (20.5) | 0.60 |
| Mild | 28 (18.7) | 43 (25.1) | |
| Moderate | 15 (10.1) | 22 (12.9) | |
| Severe | 71 (47.7) | 71 (41.5) | |

Table 6. Frequency distribution of velopharyngeal insufficiency by gender.

| Intensity of velopharyngeal insufficiency | Age | | P-value |
|---|-------------------------|-------------------------|---------|
| | New Number (percent) | Old Number (percent) | |
| Normal | 54 (42.2) | 16 (8.4) | P<0.001 |
| Mild | 41 (32) | 30 (15.7) | |
| Moderate | 16 (12.5) | 21 (11) | |
| Severe | 17 (13.3) | 124 (64.9) | |

Table 7. Frequency distribution of intensity of velopharyngeal insufficiency by type of surgical technique.

| Intensity of velopharyngeal insufficiency | SMCP | UCLP | BCLP | HSCP | SCP | P-value |
|---|---------|-----------|-----------|-----------|-----------|---------|
| | Normal | 3 (15) | 19 (22.9) | 9 (14.3) | 16 (20) | |
| Mild | 2 (10) | 27 (32.6) | 15 (23.8) | 8 (10) | 19 (25.7) | |
| Moderate | 2 (10) | 9 (10.8) | 9 (14.3) | 9 (11.2) | 8 (10.8) | |
| Severe | 13 (65) | 28 (33.7) | 30 (47.6) | 47 (58.8) | 24 (32.4) | |

Table 8. Frequency distribution of the intensity of velopharyngeal insufficiency by the type of cleft palate.

| Variable | New | Old | P-value |
|------------|------------------|------------------|---------|
| | Number (percent) | Number (percent) | |
| Fistula | 16 (12.5) | 66 (34.6) | P<0.001 |
| Recurrence | 3 (2.3) | 21 (11) | |

Table 9. Frequency distribution of fistula and recurrence by type of surgical technique.

| Variable | SMCP | UCLP | BCLP | HSCP | SCP | P-value |
|------------|------------------|------------------|------------------|------------------|------------------|---------|
| | Number (percent) | Number (percent) | Number (percent) | Number (percent) | Number (percent) | |
| Fistula | 1 (5) | 19 (22.9) | 25 (39.7) | 24 (30) | 14 (18.9) | 0.008 |
| Recurrence | 1 (5) | 3 (3.6) | 5 (7.9) | 7 (8.8) | 8 (10.8) | 0.46 |

Table 10. Frequency distribution of fistula and recurrence by cleft palate type.

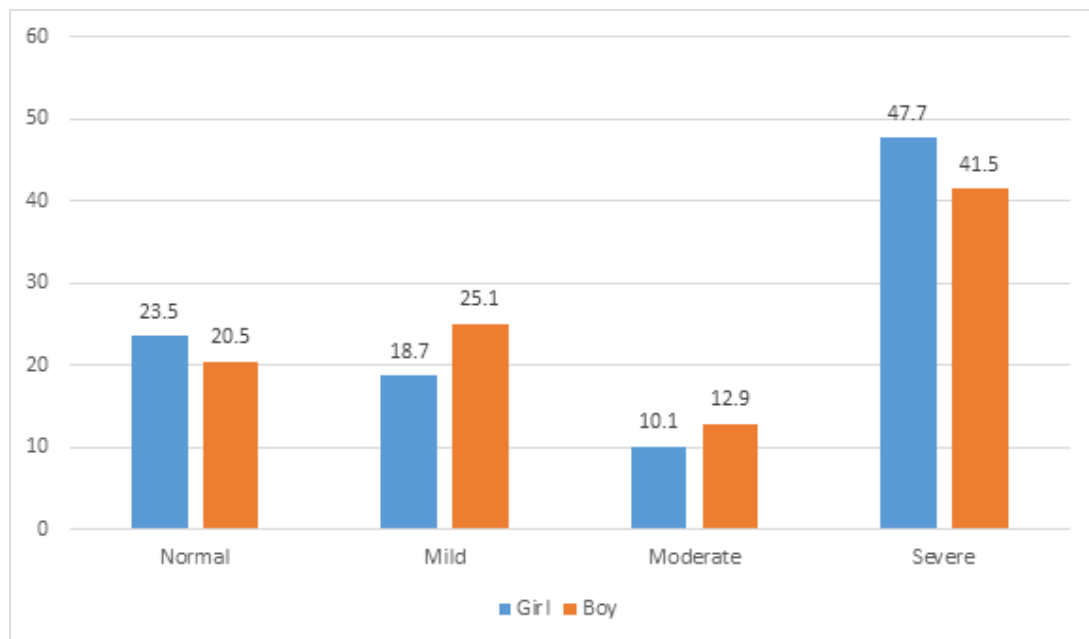


Diagram 1. Percentage of velopharyngeal insufficiency severity by gender.

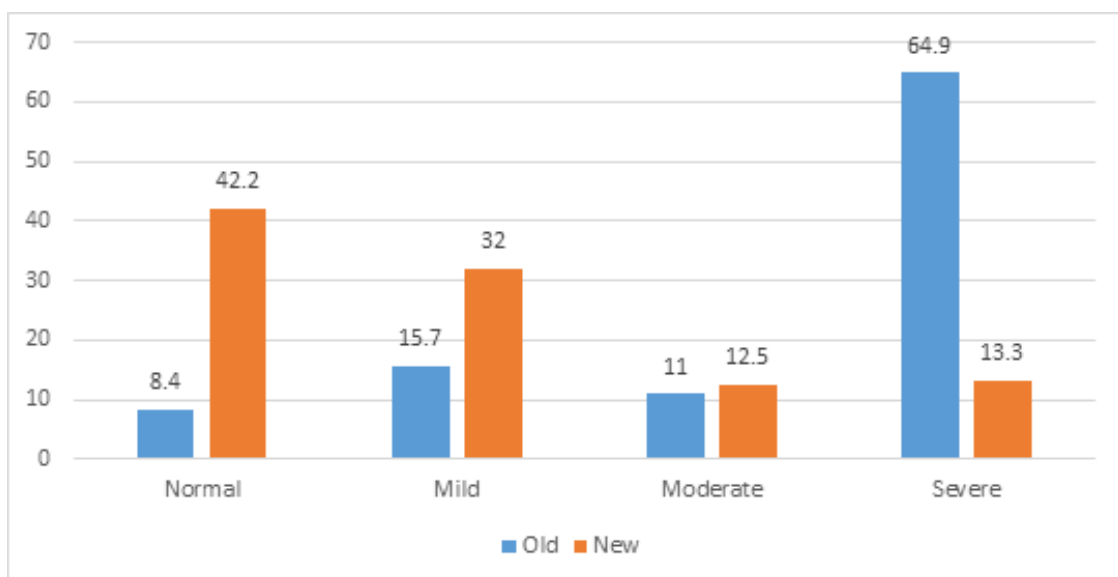


Diagram 2. Frequency distribution of intensity of velopharyngeal insufficiency by type of surgical technique.

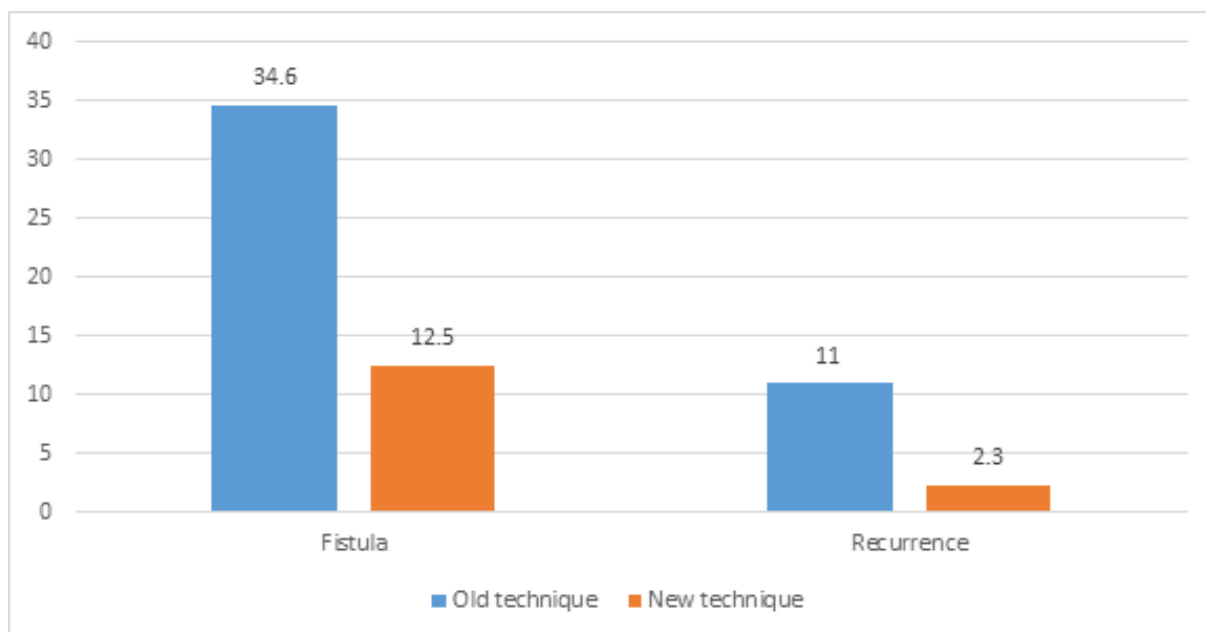


Diagram 3. Frequency percentage of fistula and recurrence by type of surgical technique.

Discussion

In the present study, the incidence of velopharyngeal insufficiency after the initial repair of the cleft palate was 78.1% and 21.9% (70 persons) were normal speech patients. Mild hypernasality was seen in 22.1% (71 cases) and moderate in 11.6% (37 cases) and severe type in 44.4% (142 cases). This is very high compared to some other studies [10,11]. Since the anatomical and functional abnormalities associated with cleft palate and their surgical repair are the main causes of speech problems, in the event that, Levator veli palatini and Tensor veli palatini muscles are not correctly positioned during the initial surgical procedure, only a small number of patients achieve their normal speech, and, given that surgical techniques and surgeon's experience and skills play a significant role in determining the child's speech skills, one of the main points of agreement between the specialists is that the results of the speech are the result of a successful surgical technique [12].

Therefore, surgical protocols and surgical techniques are the main and most important issues to be reviewed and studied [13]. Some studies have shown that in examining the speeches of patients with cleft palate, the age range of patients is effective in the results, and therefore the age range should not be widespread [13,14]. In the present study, the age range was between 3 and 12 years old and probably contributed to the results of the study. The results showed that the intensity of velopharyngeal insufficiency was not sig-

nificantly related to the age of the present or the age at time of the children's speech evaluation ($P=0.41$), but had a significant relationship with the age of the surgery ($P=0.007$). This means that the best speech results and proper operation of the velopharyngeal valve are achieved when surgery is performed at an earlier age and as soon as possible. Surgery must be performed before the child develops the verbal misconceptions [15]. However, the risk of impaired maxillofacial growth and the medial area of the face in early surgery and at lower age is unavoidable, and factors such as the size of the cleft palate and the child's health status do not allow surgery at that age [16,17]. The purpose of early surgery is that surgery should be performed before the child begins to speak, before 12 months of age [18]. Even some sources point out that surgery should be performed before 6 months of age [7]. This is unlike studies that stated that there is no significant relationship between the age of surgery and the incidence of hypernasalitis [10,19,20].

An important question is whether the child's sex affects the rate of his velopharyngeal function after the initial surgery. In the present study, no significant relationship was found between the intensity of velopharyngeal insufficiency and the gender of the patient ($P=0.6$). Other studies also indicated that there was no significant relationship between velopharyngeal function and gender [21,22]. From the embryological point of view, the cleft palate appears only in the absence of a cleft lip in most cases in the female sex, while the cleft palate along with the cleft lip, which often requires

secondary surgery, is found in the male; this may be a good justification for the impact of gender on the severity of hypernasalitis [23]. The results indicated that intensity of velopharyngeal insufficiency was significantly correlated with the type of cleft palate ($P=0.001$). So that the intensity of velopharyngeal insufficiency in children with SM cleft palate was significantly more than the type of HS cleft palate, and also in the type of HS cleft palate more than type of BCLP, and in the type of BCLP cleft there were more than two other types. Some other studies have confirmed the findings of this study [21,22]. It is noteworthy that there is a difference between the type of cleft and the width of the cleft. However, the width of the cleft palate is not very effective in speech results, and what is important is the connection between the vomer and palate, which is effective in verbal outcomes [9,14]. Syndromes and some accompanying anomalies also affect the outcome of studies [13,14,24].

The results also showed that intensity of velopharyngeal insufficiency had a significant relationship with surgical technique ($P<0.001$). In this way, the intensity of velopharyngeal insufficiency was significantly lower in children who operated with new surgical techniques than those who had been surgically treated with old techniques. Most patients with normal speech or mild hypernasalitis were surgically treated using new techniques. Contrary to our findings, some studies have shown that surgical techniques and surgeon's skills have no significant effect on clinical results [25].

Conclusion

According to the results of this study, it can be concluded surgical repair of cleft palate should be performed at younger ages (Preferably before 12 months) and in newer methods. Microsurgery palatoplasty seems to have had better results in patients. It seems that follow up of these patients after surgery for monitoring of speech complications is necessary. However, it seems that the different types of cleft palate, the amount of surgeon's experience and skill, and the age range of the patients studied can affect the difference in results in various studies.

Conflict of Interest

There is no conflict of interest to declare.

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