

Prevalence of Hoarseness in the Cleft Palate Population

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Objective: To determine the prevalence of hoarseness in the cleft palate population.

Design: Retrospective chart review from a tertiary pediatric hospital's craniofacial clinic.

Patients: Nonsyndromic patients with cleft palate who had undergone cleft palate repair were eligible for inclusion. Patients were excluded if they had previously undergone a tracheostomy or if they had significant hearing loss. A total of 487 patients met the inclusion criteria.

Main Outcome Measures: Medical records were reviewed for demographic data, presence of hoarseness, velopharyngeal insufficiency, symptoms of gastroesophageal reflux disease, and laryngoscopic findings.

Results: Of the 487 patients, 27 (5.5%) had complaints of hoarseness: 13 boys and 14 girls. The average age at initial complaint was 4.6 years, with slight differences ac-

ording to sex: 4.2 years for boys and 5.0 years for girls. Of those with hoarseness, 19 (70%) had velopharyngeal insufficiency, and 8 (30%) had concomitant symptoms of possible gastroesophageal reflux disease. Eleven patients underwent either direct or flexible laryngoscopy: 9 (33%) had vocal fold nodules, and 2 (7%) had edema and/or mucosal thickening of the vocal folds.

Conclusions: The 5.5% prevalence of hoarseness in this study is similar to the reported prevalence of 6% to 34% in the normal pediatric population. These results suggest that there is no difference in the cleft palate population and that hoarseness is either underrecognized and/or underreported. More studies are needed to fully elucidate the true prevalence of hoarseness in the cleft palate population and any correlation of hoarseness with velopharyngeal insufficiency and/or gastroesophageal reflux disease.

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MANY RESEARCHERS HAVE noted an anecdotal increase in the symptom of hoarseness in patients with cleft palate compared with the normal population; however, the exact prevalence is not currently known. Previous reports indicate that the rates of dysphonia in the cleft palate population are 12% to 43%.¹⁻⁴ These rates are similar to, or slightly higher than, the rates of hoarseness in the normal pediatric population, which are reported to be 6% to 34%.^{3,5,6}

Researchers have provided many different hypotheses to explain this possible increase in hoarseness, with the most common being laryngeal compensation for abnormal velopharyngeal valving.^{7,8} Many patients with cleft palate also have velopharyngeal insufficiency (VPI) secondary to the orientation of the palatal muscles. Velopharyngeal insufficiency, which may persist after surgical correction of the cleft, can cause hypernasality of speech and difficulty with articulation of specific consonants owing to the inability to build suffi-

cient air pressure in the oral cavity. Studies report the incidence of hypernasality in patients with cleft palate to be 25% to 40%.² Patients with VPI often use compensatory mechanisms to increase articulation. Some of these mechanisms include middorsum palatal stops, posterior nasal fricatives, velar fricatives, pharyngeal stops, pharyngeal fricatives, and glottal stops.⁹ Glottal stops, in particular, have been implicated in vocal cord abnormalities and voice disturbances such as hoarseness.^{5,7,10-12} In fact, the Cleft Palate Foundation literature states, "Children with velopharyngeal inadequacy may also have a voice disorder. In this case, your child's voice may sound 'breathy' and may fatigue easily. This problem is usually caused by the strain that he or she puts on the vocal cords while trying to build the pressure necessary for normal speech."¹³ However, a more recent study⁹ did not find any relationship between hoarseness in patients with cleft palate and VPI, calling into question the long-held theory of laryngeal compensation as the source of hoarseness.

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Table 1. Prevalence of Hoarseness in the Cleft Palate Population by Age at Time of Initial Complaint

Age, y	No. of Hoarse Patients	No. (%)					
		Cleft Palate	Cleft Lip/Palate	VPI Only	GERD Only	Both VPI and GERD	No VPI or GERD
0-2	7	4 (57)	3 (43)	2 (29)	1 (14)	2 (29)	2 (29)
3-5	10	5 (50)	5 (50)	4 (40)	1 (10)	2 (20)	3 (30)
6-10	9	4 (44)	5 (56)	4 (44)	0	4 (44)	1 (11)
>10	1	0	1 (100)	1 (100)	0	0	0
Total	27	13 (48)	14 (52)	11 (41)	2 (7)	8 (30)	6 (22)

Abbreviations: GERD, gastroesophageal reflux disease; VPI, velopharyngeal insufficiency.

Other hypotheses that have been proposed to explain a possible increased prevalence of hoarseness include the type of palatoplasty used to correct the cleft palate and a possible role of patient sex.^{7,14} To date, there is no identified definitive cause of the hoarseness, suggesting that it is most likely multifactorial or attributable to some other underrecognized factor. While most of patients with hoarseness and cleft palate are treated with voice therapy and antireflux treatment, the benefit of these interventions is also unknown.

Direct laryngoscopy (DL) of patients with hoarseness and cleft palate has resulted in mixed findings, such as vocal cord nodules and vocal cord edema. Studies in the literature report an extremely varied incidence of these findings, ranging from 7% to 84%.^{5,7,10,15} A more recent study reported no change in the prevalence of hoarseness before and after speech surgery,⁹ bringing into question the underlying causative factors.

Patients with cleft palate are possibly at increased risk for vocal dysfunction as a result of factors such as articulation disorders, VPI, laryngeal compensation, and distorted anatomy. The prevalence of hoarseness among these patients is not fully understood. The goal of this study was to look at the prevalence of hoarseness in our tertiary children's hospital craniofacial clinic and to try to determine whether there was any relationship between hoarseness and VPI.

METHODS

Institutional review board approval was obtained to search the database of the Children's Hospital of Pittsburgh Craniofacial Clinic, Pittsburgh, Pennsylvania, which contained information on 839 patients who were seen in the clinic from January 1, 2005, through August 1, 2009. Inclusion criteria for the study included nonsyndromic patients with a cleft palate (with or without a cleft lip) who had undergone palatal repair. Patients and/or families were questioned directly and filled out questionnaires that specifically asked about symptoms of hoarseness, VPI, and gastroesophageal reflux disease (GERD). A diagnosis of VPI was reached after the patient was examined by a speech pathologist (a member of the multidisciplinary craniofacial team). A definitive diagnosis of GERD was not made, but a history of symptoms associated with GERD (eg, nighttime cough, frequent spitting up, arching of back after feedings, and acidic taste in mouth) was taken from the questionnaire. Medical records were reviewed for the following data: age at initial complaint of hoarseness and presence of hoarseness before and after palate repair. The average and median ages were determined

Table 2. Characteristics of Hoarse Patients by Sex

Variable	Male	Female
No. of hoarse patients	13	14
Average age (median), y	4.2 (3.0)	5.0 (5.5)
Positive DL findings	5	6
VPI alone	7	4
GERD alone	2	0
Both VPI and GERD	2	6

Abbreviations: DL, direct laryngoscopy; GERD, gastroesophageal reflux disease; VPI, velopharyngeal insufficiency.

for the initial complaint of hoarseness as well as for DL and/or flexible fiberoptic laryngoscopy (FFL). Laryngoscopic findings as dictated in the surgical and clinical notes were reviewed.

RESULTS

A total of 487 patients met the inclusion criteria for the study. Of those patients, 27 (5.5% [**Table 1**]) had complaints of hoarseness. The average age at initial complaint of hoarseness was 4.6 years (median age, 5.0 years). There was a slight difference in age at onset noted between sexes; however, this difference was not statistically significant owing to the small number of patients ($P = .26$ for boys and $P = .23$ for girls). Boys had an earlier average age at diagnosis (4.2 years; median age, 3.0 years), with girls being slightly older (average age, 5.0; median age, 5.5 years [**Table 2**]). There was also an almost equal distribution between patients with a cleft palate only ($n = 13$) and patients with a cleft lip and a cleft palate ($n = 14$). Overall, 19 patients were also diagnosed as having VPI (70%); 10 patients (37%) had symptoms suggestive of GERD; and 21 patients (78%) had VPI and/or GERD. Only 6 patients (22%) did not have either VPI or GERD-like symptoms.

Direct laryngoscopy or FFL was performed on 11 patients (41% of patients with complaints of hoarseness), with an average age of 4.9 years at the first procedure (median age, 6.0 years). Indications for laryngoscopy were not always clearly identifiable from the clinical notes. Nine of the 11 patients (33%) were found to have vocal fold nodules, and 2 (7%) had edema or mucosal thickening of the vocal folds (**Table 3**). There was 1 patient who had vocal fold nodules on initial DL and was noted to have resolution of the nodules on subsequent DL but had

Table 3. Vocal Fold (VF) Abnormalities as Determined by Direct Laryngoscopy (DL) and/or Flexible Fiberoptic Laryngoscopy (FFL) According to Age at Initial Procedure

Age, y	No. of Patients Undergoing DL/FFL	No. of DL/FFL Procedures	VF Nodules	VF Edema and/or Mucosal Thickening
0-2	5	7	4	1
3-5	2	2	1	1
6-10	4	7	4 ^a	1 ^a
>10	0	0	0	0
Total, No. (%)	11 (41)	16	9 (33)^a	3 (11)^a

^aOne patient had VF nodules on initial DL and VF edema and mucosal thickening on subsequent DL.

edema and mucosal thickening of the vocal folds instead.

COMMENT

There is a broad range in the reported prevalence of hoarseness in the general pediatric population (6%-34%) as evidenced by the varied results of multiple different studies.^{3,5,6} The prevalence of hoarseness in the cleft population is also varied (12%-43%)¹⁻⁴ and is similar to, or possibly slightly higher than, normative values. With such large variations in both the normal and the cleft populations, it has been difficult to determine whether there is indeed an increased prevalence in the cleft population. The prevalence of 5.5% that was determined in this study is consistent with the lower range in normal patients and is below that reported for the cleft population. This finding suggests either that there is no difference between these patient populations or that the true prevalence in this cleft population is underreported or underrecognized.

When the patients were examined by age, there was fairly equal distribution among the different age groups of patients younger than 10 years, with a marked decrease in symptoms reported in the older patients. This decrease may be attributable to a decrease in the reporting of the symptoms in the older patients, to a decrease in office visits after the patients have become stable from a cleft palate perspective, to a combination of these issues, or to other factors.

There was an essentially equal distribution between male and female patients. There was a small but insignificant difference in age between sexes at initial complaint of hoarseness, with boys presenting earlier than girls (4.2 years [$P=.26$] vs 5.0 years [$P=.23$], respectively). The number of patients with VPI and/or symptoms of GERD were almost equal between sexes. Abnormal DL and FFL findings were also essentially equal between sexes (5 boys and 6 girls). Overall, there were no significant differences between boys and girls in this small population.

The main hypothesis for hoarseness in patients with cleft palate has historically been secondary to laryngeal compensation associated with VPI. However, a more recent study did not show a correlation between VPI and hoarseness in patients with cleft palate.⁹ In our patients, 19 (70%) had VPI and 8 (30%) did not. The number of

patients in our study was not large enough to determine a significant correlation between VPI and hoarseness, but our findings do suggest that there may be an association. Also, our lack of a definitive diagnosis of GERD prevented us from being able to draw any conclusions about a possible correlation of GERD and hoarseness, but, again, our findings suggest a possible association. Additional studies are needed to address this relationship.

Indications to perform DL or FFL were not clear from the patient records; however, only 11 of the hoarse patients (41%) underwent DL or FFL (either in the operating room or in the clinic). Interestingly, all patients who underwent DL or FFL were noted to have abnormal findings, which included vocal fold nodules and edema and/or mucosal thickening of the vocal folds. Vocal fold nodules were the most common finding. In the patients with abnormal findings on laryngoscopy, 8 (73%) also had a diagnosis of VPI, and the remaining 3 (37%) did not. The fact that every patient who underwent DL or FFL had abnormal findings raises a question as to whether there was selection bias toward those patients with more significant symptoms to undergo laryngoscopy or whether additional abnormalities were missed because laryngoscopy was not performed on all hoarse patients. These data also suggest that the presence of abnormal findings on DL or FFL may be more highly associated in patients with VPI.

In conclusion, the prevalence of hoarseness in the cleft palate population is not fully known. Multiple studies have suggested that it is greater than normal pediatric levels, but normal levels are also not very well established, and reports indicate significant variation, ranging from 6% to 34%.^{3,5,6} In our study, the prevalence of hoarseness was 5.5%, similar to reported normative values. This suggests that either the prevalence in this cleft palate population is similar to that in the normal pediatric population or that the prevalence in the cleft population is underreported and/or underrecognized. All of the patients who underwent DL or FFL had abnormal findings, and most patients were also diagnosed as having VPI. There was also an increase in the number of patients with symptoms consistent with possible GERD, but the lack of a definitive diagnosis of GERD makes it difficult, if not impossible, to draw any conclusions based on this finding. Because patients with cleft palate may be at risk for hoarseness, they may warrant more in-depth investigation and a lower threshold for DL or FFL

as well as for treatment of VPI and/or possible GERD. Further studies are needed to more fully elucidate the true prevalence of hoarseness in the cleft palate population.

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