

# Speech Outcomes and Velopharyngeal Function in Children Undergoing Submucous Cleft Palate Repair

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**Objective:** Describe and compare the results of speech and velopharyngeal function in children with classic and occult submucous cleft palate undergoing interdisciplinary treatment at the Gantz Foundation.

**Methods:** The clinical history of all patients born between 2012 and 2017 with a diagnosis of classic or occult submucous cleft palate was retrospectively reviewed. Preoperative and postoperative medical, surgical, and speech and language history were collected.

**Results:** Twenty-eight cases diagnosed at the age of  $44.8 \pm 23.9$  months were included. Of these, 71.4% presented classic submucous cleft, and 28.6% occult. Before primary surgery, 7.1% had a diagnosis of the syndrome, and 21.4% were under study. A total of 39.3% had hearing difficulties and 21.4% used tympanic ventilation tubes. A total of 60.7% had language problems, 39.3% had compensatory articulation, 17.9% had absent hypernasality, and 21.4% had absent nasal emission. The team indicated primary palate surgery in 71.4%, of which 85% performed the surgery at the mean age of  $61.7 \pm 24.7$  months. The surgical technique was Furlow in 88.2% of the cases and intravelar veloplasty in the remaining 11.8%. Then, 3 cases underwent velopharyngeal insufficiency surgery; 2 of them eliminated hypernasality and reduced nasal emission. The age of diagnosis ( $P=0.021$ ) and the performance of velopharyngeal insufficiency surgery ( $P=0.029$ ) of the occult submucous cleft palate group was significantly later than the classic cleft palate group.

**Conclusions:** Language, hearing, compensatory articulation, hypernasality, and nasal emission problems were recorded. A high percentage required primary surgery. Of these, a low

proportion also required a velopharyngeal insufficiency surgery, which improved the velopharyngeal function of the children but did not completely adapt it. In this regard, early diagnosis is essential, as well as an analysis of each center primary closure protocol.

**Key Words:** Cleft palate, palatoplasty, pharyngoplasty, speech, submucous cleft palate, velopharyngeal insufficiency

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The submucous cleft palate (SMCP) is a congenital malformation where exist an imperfect muscular union through the velum but with a oral and nasal mucosa continuity.<sup>1,2</sup> Among the subtypes of SMCP, the classic triad is described, characterized by a bifid uvula, notch at the posterior end of the bony palate, and a pellucida zone in the midline of the soft palate.<sup>1</sup> Meanwhile, in the occult SMCP subtype does not present the triad, but exist a disorientation and malposition of the palatal musculature that leads to velar dysfunction.<sup>3,4</sup> The malformation extent varies, from the uvula muscle hypoplasia to some diastasis degree of the remaining palatine musculature, and is not directly proportional to the velopharyngeal gap size or to the hypernasality severity.<sup>4</sup> The pharyngeal musculature function and the space size occupied by the adenoids are compensatory factors that explain, in many patients with classic or occult SMCP, their normal speech.<sup>4</sup>

The classic SMCP has an estimated general population incidence ranging from 0.02% to 0.08%.<sup>5</sup> The general population occult SMCP incidence has not been determined and has only been investigated in those subjects referred by velopharyngeal insufficiency (VPI). Shprintzen published that 92% of the subjects with bifid uvula studied ( $n=25$ ) had other findings associated with occult SMCP.<sup>6</sup> The occult SMCP might be more prevalent than thought, considering that bifid uvula has been reported to have an incidence of 1% to 7.5%.<sup>7</sup> Occult SMCP associated with absent or deficient uvula muscle may be sufficient to cause VPI. The association of occult SMCP in patients with VPI has been studied: varying between 10% and 22% the occult SMCP and between 17% and 44% the classic SMCP.<sup>3,4</sup>

The SMCP could be present with or without VPI.<sup>8</sup> It has been reported that 90% are asymptomatic and do not require treatment.<sup>9</sup> Only 5% to 10% develop VPI, requiring to be studied.<sup>10,11</sup> Historically, they have been related to hypernasal speech that can even become unintelligible.<sup>1</sup> In the same way, one of the main speech and language difficulties that people with some type of cleft palate present is the production of compensatory articulations (CA), which affect the linguistic system and creates a different motor engram for speech sounds

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production, severely impacting speech intelligibility.<sup>12,13</sup> Early detection is important to allow timely speech therapy intervention and/or surgical repair before children develop compensatory speech mechanisms.<sup>14</sup>

In cases with VPI, surgery is usually indicated after 3 years old.<sup>9</sup> Three types of surgical management have been postulated: velar repair, pharyngoplasty, and combination procedures of the above.<sup>15–17</sup> However, there is little evidence to support which technique offers the optimal result without the need for another surgery.<sup>18</sup>

Studies have defined surgical success in relation to the lower percentage of patients requiring secondary surgery for residual VPI.<sup>10</sup> In that sense, a recent systematic review and a meta-analysis in the pediatric population with nonsyndromic SMCP, reported that the most common secondary surgical techniques were Furlow Z-plasty, intravelar veloplasty (IVV), and palatoplasty techniques as “straight line repair” (SLR) and pharyngeal flap (PF). The latter had the lowest reoperation rate and the best result as a primary technique (0%) with no persistence of VPI.<sup>19</sup>

In Chile, the comprehensive treatment of patients with cleft lip and palate is described in the “Explicit Health Guarantees” clinical guide for cleft lip and palate.<sup>20</sup> Similar to other countries, SMCP is still underdiagnosed with consequent alterations in children’s speech and/or surgical indications at older ages. For this reason, the purpose of this study is to describe the speech and velopharyngeal function of children with SMCP attended by an interdisciplinary care team for patients with craniofacial malformations in Santiago of Chile.

## METHODS

Retrospective review of clinical history.

### Participants

The study universe was 432 patients born between January 2012 and July 2017 who underwent their primary palate surgery and interdisciplinary treatment at Fundación Gantz in Santiago, Chile. The inclusion criteria established for the participants selection were medical diagnosis of syndromic or nonsyndromic SMCP; SMCP surgery at the Fundación Gantz; speech therapy intervention in the preschool stage or school stage; full access to the patient’s clinical record. Meanwhile, the exclusion criteria were patients with complete or incomplete cleft palate and completed unilateral or bilateral cleft lip and rare cleft.

This research was approved by the Facultad de Medicina of the Universidad de Chile Bioethics Committee, project No. 263-2020.

All the children treated at the Fundación Gantz followed an SMCP presurgical and postsurgical protocol, both for surgeries and for speech therapy evaluation, described below:

### Procedures and Instruments

#### Sociodemographic, Medical, and Surgical History

The information extracted from the medical records is shown in Supplemental Table 1, Supplemental Digital Content 1, <http://links.lww.com/SCS/F275>.

In relation to primary palate surgery, information was obtained regarding video nasopharyngoscopy previous performance; primary surgery need; age and type of surgery; and complications presence (hemorrhage, anemia, respiratory problem in the immediate postoperative period, dehiscence in the first few days, fistula 1 mo postoperative).

Finally, information was obtained on the need for secondary VPI surgery; secondary surgery age; the secondary surgery age and type of surgery; and the presence of complications.

### Speech Therapy Assessment

In this regard, information was extracted on: a language problem history before primary surgery; the first consultation age before and after primary surgery; and whether the team suggested primary surgery, among others.

According to Henningsson description,<sup>21</sup> the CA assessment recorded the absence or presence of CA, based on the results of the articulatory evaluation performed in vivo during the first consultation before primary surgery.

On the other hand, in the velopharyngeal function perceptual assessment, the hypernasality degree and nasal emission were obtained, as described by Henningsson et al.<sup>21</sup> In addition, in the nasal emission case, the “intermittent” option was added when the person is consciously capable of canceling nasal emission, but in spontaneous speech it is still appreciable. These evaluations were performed in person by a speech therapist with more than 15 years of experience in the therapy of people with cleft palate. The reports were made before and after the primary palate surgery and after the VPI secondary surgery.

Also, results were obtained from all participants who underwent a nasometry assessment, performed with a model 6450 Nasometer (Kay Elemetrics Corp., Lincoln Park, NJ). The nasalance value (acoustic correlate of nasality) obtained during the repetition of sustained vowels /a/ and /i/ and the production of a numerical series (counting from 1 to 10) were recovered before and after primary surgery and after VPI surgery.<sup>22,23</sup>

### Statistic Analysis

Data were collected with Google Sheets<sup>24</sup> and analyzed with Jamovi<sup>25</sup> and the R.<sup>26</sup> To compare the results between both groups and between surgeries, the Mann-Whitney *U* Test and Fisher exact test were used. Differences were considered significant when  $P < 0.05$ .

## RESULTS

Forty-six (10.64%) of the 432 patients born between January 2012 and July 2017 presented a diagnosis of SMCP; however, for this study, only 28 met the inclusion criteria. The results obtained are presented below.

### Background

Supplemental Table 1, Supplemental Digital Content 1, <http://links.lww.com/SCS/F275> shows the number (n) and percentage (%) of children for the different antecedents, both for the total and for the classic SMCP and occult SMCP groups. In this regard, of the 28 (100%) participants, 71.4% (20/28) were diagnosed with classic SMCP and 28.6% (8/28) with occult SMCP. The diagnosis of SMCP was made in the same number of boys and girls (50% each). Regarding a genetic diagnosis associated with SMCP, 7.1% (2/28) of the cases presented a diagnosis of chromosome 22q11.2 deletion syndrome. Meanwhile, 24.7% (6/28) of the children were still undergoing a genetic study at the time of diagnosis. For their part, before primary palate surgery, 39.3% (11/28) of the children had a history of hearing difficulty, and 21.4% (6/28) had used collars. Finally, when comparing the groups of children with classic SMCP and occult SMCP, no statistically significant differences were observed ( $P > 0.05$ ).

### Surgical Treatment and Speech Therapy Evaluation

Supplemental Table 2, Supplemental Digital Content 2, <http://links.lww.com/SCS/F276> presents the number and age of important milestones in the interdisciplinary treatment of

SMCP, both for the total number of children and for both groups. Only 60.7% (17/28) of the children underwent primary palate surgery; of these, 94.1% (16/17) attended a postsurgical speech assessment. Of which, 18.8% (3/16) underwent secondary surgery to correct residual VPI who attended 100% (3/3) postsurgical evaluation. Meanwhile, when comparing the ages of the different milestones, a statistically significant difference was only observed in the diagnosis of SMCP ( $P=0.021$ ), being later in the group of children with occult SMCP ( $61.0 \pm 18.4$ ).

In relation to the interdisciplinary approach parameters (surgeon-speech therapist). Supplemental Table 3, Supplemental Digital Content 3, <http://links.lww.com/SCS/F277> that before primary palate surgery, 60.7% (17/28) of the children had a history of language problems, 39.3% (11/17) CA, and 60.7% mild (9/28) and moderate (8/28) hypernasality, and 57.1% (16/28) nasal emission. However, hypernasality and nasal emission could not be evaluated in 21.4% (6/28) of the children, due to presenting AC at a level that made perceptual evaluation of velopharyngeal function impossible. In addition, the evaluation of hypernasality was complemented with the nasometry of the vowels /a/ and /i/ and the numerical series 1 to 10, observed in Supplemental Table 4, Supplemental Digital Content 4, <http://links.lww.com/SCS/F278>. Meanwhile, 32.1% (9/28) underwent a video nasopharyngoscopy before the surgery, of which one 77.7% (7/9) were occult and 22.3% (2/9) classic SMCP. In this regard, all the parameters analyzed before primary palate surgery did not differ significantly between the classic SMCP and occult SMCP groups ( $P>0.05$ ).

In relation to the primary surgery and postoperative results, such as seen in Supplemental Table 3, Supplemental Digital Content 3, <http://links.lww.com/SCS/F277>, a palate primary closure was indicated in 71.4% (20/28) of the children with SMCP. Of these, 85% (17/20) underwent primary surgery, all without complications. The Furlow technique was mostly performed (88.2%).

When analyzing the surgical technique by type of SMCP, it can be observed that in the case of occult SMCP operated with IVV, hypernasality, and nasal emission did not improve. However, of the 5 cases operated on with Furlow, 2 eliminated hypernasality and nasal emission, 1 maintained hypernasality and eliminated nasal emission, and another case maintained hypernasality and nasal emission. Meanwhile, one case was not evaluated in the presurgical stage, so the change cannot be determined.

In the 10 cases of classic SMCP operated with Furlow, it was observed that 3 eliminated hypernasality, and only 1 case, nasal emission. Meanwhile, 2 children were not evaluated in one of the stages, so it is not possible to compare them. The rest did not present changes in the perceptual evaluation. The case of classic SMCP operated with IVV did not present hypernasality and nasal emission in the postoperative evaluation.

After surgery, a video nasopharyngoscopy was performed in 23.5% (4/17) and a speech-language evaluation in 94.1% (16/17). In the latter, 56.3% of mild (4/16) and moderate (5/16) degree hypernasality was recorded, and 56.3% (9/16) of nasal emission. Similarly, Supplemental Table 4, Supplemental Digital Content 4, <http://links.lww.com/SCS/F278> presents the post-surgery nasometry values.

Supplemental Table 5, Supplemental Digital Content 5, <http://links.lww.com/SCS/F279> presents the presurgical and postsurgical comparison of hypernasality, nasal emission, and nasalance. The only significantly different value being nasalance obtained by counting from 1 to 10 ( $P=0.045$ ). On the basis of the results, the speech therapist suggested VPI surgery in 18.8% (3/16) of children at this stage. In this regard, between the classic SMCP and occult SMCP groups, there was a significant

difference between the speech therapist's suggestion of VPI surgery ( $P=0.036$ ) and performing video nasopharyngoscopy ( $P=0.006$ ), higher in the occult SMCP group.

Supplemental Table 6, Supplemental Digital Content 6, <http://links.lww.com/SCS/F280> shows the results of 17.6% (3/17) of the children who underwent primary closure, who underwent secondary VPI surgery, which corresponds 100% (3/3) to the occult SMCP group. Of these surgeries, 66.7% (2/3) were a PF, and 33.3% (1/3) had an increase in the posterior pharyngeal wall. None reported complications (0%). After surgery, 33.3% (1/3) presented moderate hypernasality and 100% (3/3) nasal emission. It should be noted that the nasal emission in 2 of the 3 cases (1 with flap and 1 with lipoinjection) was intermittent, unlike what was observed before surgery, where the 3 cases presented constant nasal emission. Meanwhile, the nasalance values are presented in Supplemental Table 4, Supplemental Digital Content 4, <http://links.lww.com/SCS/F278>. Of these results, only the percentage of nasalance obtained in the vowel /i/ differed significantly ( $P=0.016$ ) between the classic and occult SMCP groups. Lastly, when comparing the presurgical and postsurgical results, no significant differences were observed between the parameters (Supplemental Table 5, Supplemental Digital Content 5, <http://links.lww.com/SCS/F279>).

## DISCUSSION

The SMCP can be considered a separate diagnostic category from the traditional cleft palate. Its management remains challenging.<sup>4,27</sup> To describe the speech and velopharyngeal function of children with SMCP treated at our institution, we analyzed various variables.

According to the incidence of SMCP in patients with cleft palate, in people with nonsyndromic SMCP round between 3.2% and 7.7%,<sup>2,5,27-29</sup> In the present study, 10.64% of the patients with cleft palate presented a diagnosis of SMCP. This percentage corresponds to an incidence higher than previously reported, which could be due to the fact that in Chile, our institution is a national reference center, and in the event of suspicion or discovery of any sign of SMCP or VPI without cleft palate, many pediatricians, otolaryngologists, speech therapists, among other professionals make the referral. Regarding sex, the diagnosis of SMCP was made in 50% of boys and girls, similar to that reported by Smit et al (2021) and Denadai and colleagues, where the proportion of men/women was 58.3%/41.5% and 48.1%/51.9%, respectively.<sup>2,28</sup>

With respect to SMCP subtype, some studies consider that the classic SMCP must comply with Calnan triad, while patients with the occult variant have been identified based on various criteria: intraoral examination without hard palate notch, septum pellucidum, or bifid uvula, an inverted V-shaped pattern (concave or flattened surface) of the soft palate rising during opening or phonation; and/or a central width deficiency near the uvula region. This intraoral examination could be with or without nasopharyngoscopic evaluation as an additional diagnostic method to evaluate the structure (anatomy) of the velopharyngeal valve when there were vague clinical findings in symptomatic children older than 4 years (that is to say, collaborative patients).<sup>27</sup> In most cases, our team uses nasopharyngoscopic examination to confirm the suspicion of occult SMCP, after VPI diagnosis, even if there is no finding on intraoral inspection.

Of the 28 SMCP diagnosed, 71.4% were classic SMCP, and 28.6% with occult SMCP. This is consistent with the findings in nonsyndromic subjects, where the highest proportion is in the classic version, which has more evident clinical signs: in the Netherlands study on symptomatic SMCP, 92.8% had classic

SMCP and 7.1% occult.<sup>5</sup> In the publication of Taiwan, 78.2% presented the classic variant, and 21.7% occult.<sup>27</sup>

On the other hand, when analyzing the type of cleft in a population with syndromic association, the velocardiofacial syndrome (VCFS) is the most associated with occult SMCP and in second place to classic SMCP.<sup>7</sup> In a Chilean study, in 80 confirmed diagnoses of 22 deletion syndrome, it was confirmed that the proportion of occult SMCP (72.5%) is inverse, that is, greater than the classic SMCP (27.5%).<sup>30</sup>

Regarding the genetic diagnosis associated with SMCP, 7.1% (2/28) of the cases in our sample had VCFS. Meanwhile, 24.7% (6/28) were still undergoing a genetic study at the time of diagnosis. Many children are evaluated, but the families do not carry out the requested examinations, mainly for economic reasons, and those who carry them out do not necessarily return with the diagnostic confirmation. In a reference publication, from 56 children with nonsyndromic SMCP, 32.1% were diagnosed with a syndrome and 19.6% had other anomalies. VCFS was the most common (7.1%)<sup>14</sup> with the same proportion of our sample.

Have been published nonsyndromic SMCP mean ages at diagnosis at 44.0 months (3.66 y) and 3.9 years, both only from classic SMCP.<sup>5,16</sup> This is consistent with previously reported results of late diagnosis.<sup>5</sup> Meanwhile, Smit et al<sup>2</sup> reported a median time to diagnosis of 33 months without distinguishing between classic and occult SMCP. Our sample was diagnosed at 45 months. It must be considered that this figure includes classic and occult SMCP, where a statistically significant difference was observed in the diagnosis, being later in the group of children with occult SMCP (61 mo) versus the classic (38 mo), which was to be expected considering that in the first one, searchable intraoral clinical findings are not necessarily noted, but rather the suspicion is based on the nasal voice. In this sense, it has been recommended to take into account the existence of SMCP in children with speech, hearing and/or feeding problems, with repeated episodes of otitis media and nasal regurgitation.<sup>14</sup> However, it is not necessary to actively search for the existence of SMCP in patients without complaints.<sup>5</sup>

In relation to the age of the first speech therapy assessment, early detection reduces the likelihood of receiving secondary surgery.<sup>31</sup> It is crucial to start speech therapy before compensatory speech mechanisms develop.<sup>16</sup> Increasing expectations for final normal speech function.<sup>31</sup> In our study, the mean age of the first speech therapy consultation was 47 months, being lower in the classic SMCP group (42 mo) and higher in the occult SMCP group (58 mo), a difference that is consistent with the diagnostic age by group. The same report was not found in similar studies, but the age at which the speech problem was diagnosed, which in the publication by Jung et al<sup>31</sup> was an average of 48.5 months, figure very similar to ours.

Other characteristics analyzed in our study correspond to aspects of hearing, speech and velopharyngeal function of the kids. We report that before primary palate surgery, 39.3% (11/28) had a history of hearing difficulty, and 21.4% (6/28) required tympanic ventilation tubes. Similar to us, Ten Dam and colleagues registered a conductive loss in 39% of 28 patients. But their ventilation tubes installation percentage was 50% before SMCP diagnosis, which evidences a more conservative management by our team in the T tubes indication. Raby-Smith et al<sup>32</sup> published that in 57 patients who underwent SMCP repair, otologic disease was present in 54.4%.

In relation to the speech disturbances before primary surgery, in our study, 39.3% of the participants presented CA. A group of Korean researchers reported a similar value, of which 37.5% had CA before primary surgery, which decreased to

19.2% after the surgical intervention and speech therapy, but it is not clear who received it.<sup>31</sup> When consulting the studies cited in this review in detail highlights the work of Baek et al,<sup>33</sup> who reported 45% of CA; Park et al,<sup>34</sup> who reported 40% glottal articulation and 13% palatalized articulation, and Ten Dam et al<sup>5</sup> with a 56% of articulation problems. On the other hand, in this retrospective study, no information was obtained regarding the patients who received speech therapy and for how long, so future investigations should take these factors into account.

Continuing with the results of velopharyngeal function, in our study, before primary surgery, 57.1% of the cases present nasal emission, 32.1% mild hypernasality, and 28.6% moderate hypernasality. By contrast, regarding cases of hypernasality before primary surgery, Bezuhly et al<sup>35</sup> reported 55 individuals (67% mild, 27% moderate, and 5% severe), Brandão et al<sup>36</sup> reported 25 subjects (24% mild, 56% moderate and 20% severe), Ezzat et al<sup>37</sup> described 20 patients (60% mild, 25% moderate and 15% severe), and Ng et al<sup>11</sup> reported 17 cases (53% mild, 41% moderate, and 6% severe). For their part, Ten Dam et al<sup>5</sup> recorded 78% of hypernasality in 23 patients, and Jung et al<sup>31</sup> in 72 participants reported 76.4% of hypernasality and 36.1% of nasal emission. It should be noted that most of these works considered subjects with syndromic SMCP, which could partly justify the higher percentage of cases with hypernasality to different degrees.

According to the need for palate surgery in nonsyndromic SMCP, Jung et al<sup>31</sup> reported that 64% (46/72) of the children with SMCP required surgical repair. Another reference states an 86% need for surgery, after having been treated with speech therapy to improve speech (without exact data on its duration), to 1 surgical middle ages of 4.5 years. In agreement, the other 4 publications cite age ranges between 3.7<sup>5</sup> and 3.9 to 7.7 years.<sup>14</sup> In our sample, 61% of the children underwent primary palate surgery, a slightly lower data than those of reference, at a mean age of 5.2 years. Meanwhile, there was not a significant difference between the surgical age of the group with classic (5.1 y) and occult SMCP (5.3 y). This partially agrees with what is found in the literature. In a study in Taiwan, the mean surgical age was 6.2 years, being significantly older for occult SMCP (8.3 y) in relation to the classic one (5.6 y).<sup>27</sup> Our study is similar to this reference only in the age of operation of the group with classic SMCP since that of the occult group is much lower. Unfortunately, they do not show the diagnostic age, making it impossible to determine whether a later diagnosis caused this later surgical age.

In the last 20 years, the Furlow palatoplasty surgical technique has been the first-line management procedure for patients diagnosed with classic and occult SMCP associated with VPI.<sup>28</sup>

The results of 29 patients who underwent a superior base PF combined with IVV or Furlow palatoplasty for SMCP repair were reported showing a significant velopharyngeal closure correction in both groups.

The correction of the nasalance scores obtained according to the surgical procedure for the syllables /s/ and /k/ demonstrated a significantly greater benefit with the PF combined with the IVV than with the Furlow palatoplasty.<sup>38</sup>

Another publication compared 4 techniques for primary SMCP closure in 50 patients, concluding that the PF and pushback palatoplasty combined with a PF seem to be more reliable procedures than isolated pushback palatoplasty in nonsyndromic SMCP population.<sup>34</sup>

A recent systematic review and meta-analysis,<sup>19</sup> which included 383 children with nonsyndromic SMCP, shows that the most common surgical techniques were the Furlow double-opposing Z-plasty pharyngeal flap (PF), intervallary veloplasties

and palatoplasty techniques, called “SLR” and PF. The latter had the lowest reoperation rate and therefore the best result as a primary technique (0%) and no VPI persistence. The difference between surgical techniques was not statistically significant, but there was a notable disparity in the reported rates of secondary surgery by surgical technique, with 0% for PF to 17.8% for SLR.<sup>19</sup>

On the other hand, a relationship between surgical technique and size of the velopharyngeal space has been described, concluding that palatoplasty with Furlow Z-plasty has a very high success rate in patients with velopharyngeal spaces of 8 mm or less and less probabilities of successful when the velopharyngeal space exceeds 8 mm.<sup>39</sup> For patients who have significant signs of hypernasality, the contribution of the PF may be considered.<sup>38</sup>

By institutional management protocol, all our patients with nonsyndromic SMCP of both subtypes, underwent primary cleft closure. The nasopharyngoscopy is indicated only to confirm the suspected diagnosis of occult SMCP and not for surgical management. In our sample, nasopharyngoscopy was performed on 5 of the 8 children with occult SMCP. And although it is not a routine indication in our center, 4 out of 20 with classic SMCP also underwent nasopharyngoscopy. It is important, after this analysis, to follow clear diagnostic study guidelines, as suggested in other studies. A different scheme is applied in those with SMCP, chromosome 22 microdeletion, and severe VPI, where the indication for a custom flap has been protocolized after performing nasopharyngoscopy.<sup>40</sup>

Of the 17 children in our sample who required surgery, the Furlow technique was used in 15, of which 10 presented classic and 5 occult SMCP. The remaining 2 underwent IVV, 1 classic, and 1 occult SMCP. In this regard, in our center, the selection of the technique is made by the surgeons based on a clinical analysis of the characteristics of each case. If the local conditions of the veil show adequate tissues in quality and size, which allow the carving of the 4 flaps of the Z-plasty with good vitality of the same, the Furlow will be indicated. Whereas, if the tissues are very hypoplastic, with a risk of dehiscence and/or fistulas secondary to zetaplasty, IVV is preferred as it ensures better irrigation of the palatal flaps.

The Furlow technique was preferred. Its result was totally successful in 3 cases, 2 with occult, and 1 classic SMCP. The rest had favorable changes in VPI but not within the normal range. Others kept hypernasality and nasal emission. Although the IVV technique was completely successful for the patient with classic SMCP, this was not the case in the one with occult SMCP, who maintained its hypernasality and nasal emission. It is worth mentioning that the first was diagnosed at 9 months and operated at 18 months, whereas in the second case, both ages were over 76 months. Neither of the 2 children had an associated syndrome.

It has been published that VPI secondary surgery was recommended in 11.1% of 24 patients.<sup>27</sup> Another reference reports that 4 of 46 operated children (8.7%) required secondary surgery at an average age of 83.5 months.<sup>31</sup> Three patients from our study, corresponding to 17.6%, required secondary VPI surgery, a higher proportion than the references. At an average age of 74.6 months, lower than that cited as a reference.

All of our surgical group at this stage had an occult SMCP diagnosis, 2 of them received the Furlow technique as primary surgery (33 and 83 mo), and 1 case the IVV (78 mo). On the basis of the nasopharyngoscopy, a PF was indicated for 2 of them. The third child underwent an augmentation pharyngoplasty at 92 months, lipoinjection type of pharyngeal walls, and veil. This subject was left without hypernasality, only with intermittent nasal emission. Their nasometric results were 10% for the vowel /a/, 42% for the vowel /i/, and 42% for the numerical series (1–10).

In the same way, one of the children who underwent PF surgery at 48 months also eliminated hypernasality and only maintained intermittent nasal emission. Both results were considered favorable. The second patient with a PF, performed at 96 months, maintained the preoperative results of hypernasality and nasal emission, with an unfavorable result. She was operated primarily with the Furlow technique, at 6.9 years of age, having previously performed nasopharyngoscopy. We believe that, as the literature suggests, a delayed age of repair inherent to SMCP can perform operations that rely on a functional levator muscle with less favorable results. Gap size, which is often used in the post-palatoplasty VPI population to guide treatment, seems a logical and useful objective measure to guide the primary surgical technique of choice for SMCP,<sup>19</sup> management that, at the date of this review, was not part of our team’s protocol.

According to what was reported in a recent systematic review, the complications in the 16 articles were reported variably but generally low. Obstructive sleep apnea was not mentioned in the majority of articles (60%), and of those that did report, there were no cases identified. Similarly, only 1 case of hyponasality was detected (SP group). Fistula rates were also not declared by the majority (60%); only 1 case was identified (FP group). They conclude that it is difficult to make recommendations based on these data.<sup>19</sup> In another publication, 3 patients (1.4 percent) were reported who presented complications related to the surgery, including bleeding and partial rupture of the wound.<sup>28</sup>

Our surgical data does not record the presence of complications, neither in relation to primary or secondary surgery, considering: hemorrhage, anemia, respiratory problems in the immediate postoperative period, dehiscence in the first days, and/or postoperative fistula.

## CONCLUSIONS

Children with a diagnosis of SMCP treated at the Gantz Foundation were more cases of classic than hidden SMCP, with equal distribution of men and women, a significant number of cases with a genetic syndrome under study or diagnosed, and with a history of hearing problems similar to international reports, but with less installation of transtympanic ventilation tubes.

In relation to the speech and velopharyngeal function of the children with SMCP, before the primary surgery, a significant percentage had a history of language problems, CA, hypernasality, and nasal emission. These last 2 parameters observed in the VPI were complemented with the nasometry and the interdisciplinary team clinical evaluation, indicating a primary surgery in more than half of the individuals, being executed the majority of these with the Furlow technique and in smaller quantities with an IVV, with no reported complications. After surgery, most of the children attended speech therapy evaluation, a lower percentage of hypernasality and nasal emission was recorded; however, some cases required VPI secondary surgery correction, which was planned based on the video nasopharyngoscopy.

The VPI surgeries performed were the PF and subsequent pharyngeal wall augmentation, which improved velopharyngeal function but did not completely adequate it. In this regard, early diagnosis is essential, as well as analysis of the primary SMCP closure protocol, as a standard surgical management procedure for all cases.

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