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**LIPOMODELAGE PHARYNGÉ DANS L'INSUFFISANCE
VÉLOPHARYNGÉE : UN TRAITEMENT DE PREMIÈRE INTENTION ?
COMPARAISON AU LAMBEAU PHARYNGÉ POSTÉRIEUR A PÉDICULE
INFÉRIEUR. ÉTUDE RÉTROSPECTIVE DE 70 CAS.**

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TABLE DES MATIÈRES

ABRÉVIATIONS.....	1
TABLE OF FIGURES.....	3
TABLE OF TABLES.....	4
INTRODUCTION	5
ARTICLE	10
ABSTRACT.....	11
INTRODUCTION	13
MATERIALS AND METHODS	17
Data collection	17
Patients.....	17
PFI ± PL group.....	18
VPP Group.....	18
Surgical Procedures	19
<i>PFI</i>	19
<i>VPP with an inferiorly based pharyngeal flap and an uvula island flap</i>	21
<i>Palatal lengthening</i>	22
<i>Associated procedures</i>	24
<i>Previous procedures on the velopharyngeal sphincter</i>	24
<i>Exploration of velopharyngeal insufficiency</i>	24
<i>Statistical Tests</i>	27

RESULTS	28
Patients.....	28
Previous procedures on the velopharyngeal sphincter.....	29
Associated procedures	30
Speech and language assessment.....	33
Cases of moderate to severe VPI	36
PFI ± PL group.....	37
 DISCUSSION	 40
 CONCLUSION	 44
 DISCUSSION	 45
 RÉFÉRENCES.....	 50

ABRÉVIATIONS

ABRÉVIATIONS FRANÇAISES

ATT : aérateurs transtympaniques

AV : allongement vélaire

EVA : échelle visuelle analogique

IC95% : intervalle de confiance à 95%

IRM : imagerie par résonance magnétique

IVP : insuffisance vélopharyngée

LP : lipomodelage pharyngé

LPP : lambeau pharyngé postérieur

NF1 : neurofibromatose de type 1

SAOS : syndrome d'apnées obstructives du sommeil

SPR : séquence de Pierre Robin

SVCF : syndrome vélo-cardio-facial, syndrome de délétion 22q11

SVP : sphincter vélo-pharyngé

UVPP : uvulovélopharyngoplastie

VIV : véloplastie intra-vélaire

ENGLISH ABBREVIATIONS

CI95%: confidence interval 95%

IVV: intravelar veloplasty

MRI: magnetic resonance imaging

NF1: type 1 neurofibromatosis

OSA: obstructive sleep apnea

PL: palatal lengthening

PFI: pharyngeal fat injection

PPF: posterior pharyngeal flap

PRS: Pierre Robin sequence

SLA: speech and language assessment

VCFS: velocardiofacial syndrome, 22q11 deletion syndrome

VELO: VPI Effects on Life Outcome

VPI: velopharyngeal insufficiency

VPIQL: VPI Insufficiency Quality of Life

VPP: velopharyngoplasty

TABLE OF FIGURES

<i>Figure 1.</i> Pharyngeal fat injection in a child previously treated with PFI and V-Y pushback palatal lengthening _____	21
<i>Figure 2.</i> VPP with an inferiorly based pharyngeal flap in a patient previously treated for a cleft palate _____	22
<i>Figure 3.</i> Uvula island flap with V-Y plasty _____	23
<i>Figure 4.</i> Z plasty of oral mucosa for velar lengthening _____	23
<i>Figure 5.</i> Speech and Language assessment carried out by a speech and language therapist with extensive experience in evaluating VPI _____	26

TABLE OF TABLES

<i>Table 1.</i> Borel-Maisonny score: subjective evaluation of hypernasality and intelligibility	27
<i>Table 2.</i> Patient's characteristics	28
<i>Table 3.</i> Previous procedures on the velum in the two groups	30
<i>Table 4.</i> Details of procedures carried out in the PFI ± PL group	31
<i>Table 5.</i> Evolution of the Borel-Maisonny score in the PFI ± PL group	33
<i>Table 6.</i> Evolution of the Borel-Maisonny score in the VPP group	34
<i>Table 7.</i> Comparison of the results between the two groups: Borel-Maisonny score, intelligibility, nasality, nasal air emission, duration of hospitalization and resumption of oral intake	35
<i>Table 8.</i> Comparison of the results between the two groups for moderate to severe VPI	37
<i>Table 9.</i> Phonatory results after second PFI	38
<i>Table 10.</i> Comparison of the results for PFI alone and PFI + PL	39

INTRODUCTION

L'insuffisance vélo-pharyngée (IVP) se définit par une occlusion incomplète du sphincter vélo-pharyngé due à une inadéquation anatomique entre la longueur réelle du voile et celle nécessaire à son contact ferme avec les parois pharyngées postérieure et latérales. Ce sphincter vélopharyngé est constitué des muscles élévateurs et tenseurs du voile du palais, palatoglosses, palatopharyngiens, uvulaire, salpingopharyngiens et constricteur supérieur du pharynx. Sa fermeture joue un rôle dans la phonation, la succion, la protection des fosses nasales lors de la déglutition et des vomissements, l'audition et la respiration (1,2).

La principale cause d'IVP est la présence d'une fente vélaire (3), isolée ou non, syndromique ou non, ouverte ou sous-muqueuse ainsi que les conséquences de sa fermeture chirurgicale. Dans ce cas, l'IVP peut s'expliquer par une rigidité vélaire cicatricielle, un voile court et/ou la persistance d'un défaut musculaire responsable d'une incompétence vélaire. Outre l'insuffisance vélopharyngée, de cause structurelle, peut exister un degré plus ou moins important d'incompétence vélopharyngée, fonctionnelle, avec hypocontractilité vélaire et/ou pharyngée due à une faiblesse musculaire constitutionnelle, à un défaut de réparation chirurgicale de l'intégrité musculaire du voile ou à une atteinte neurophysiologique en cas de syndrome associé.

Le cas des fentes vélares sous-muqueuses est un peu particulier. Cliniquement, celles-ci se présentent fréquemment par une luette bifide, et toujours par un défaut musculaire vélaire médian, palpable et parfois visible à son aspect bleuté et pellucide. Ce terme a été controversé, la fente sous-muqueuse congénitale étant en réalité comparable au résultat obtenu après fermeture chirurgicale d'une fente vélopalatine, la fente osseuse n'étant pas comblée. Discrètes cliniquement, leur diagnostic est souvent tardif, parfois lors de la décompensation d'une IVP après adénoïdectomie, contre-indiquée pour cette raison de façon relative en cas de fente sous-muqueuse connue.

En l'absence de fente vélaire, une IVP peut être secondaire à différentes atteintes neurologiques, anatomiques et/ou iatrogènes (4). Des défauts de l'intégrité vélaire ont pu être reliés à plus de 400 syndromes, le syndrome de délétion 22q11 étant le plus fréquemment retrouvé (5). Après bilan génétique, seuls 14% des IVP non liées à une fente demeurent sans cause retrouvée (6).

Les causes iatrogènes sont également relativement fréquentes, avec une incidence importante d'IVP post-adénoïdectomie, en particulier en cas de voile court ou de fente sous-muqueuse non diagnostiquée (7,8). Des IVP après avancée maxillaire par ostéotomie de Lefort 1 ont également été décrites chez des patients atteints de fentes labio-maxillo-palatines (9).

Sur le plan phonatoire, l'insuffisance vélo-pharyngée se manifeste par un nasonnement et une déperdition nasale d'air, accompagnés ou non de phénomènes compensatoires articulatoires (10). Lorsque l'intelligibilité est affectée, l'altération de la communication des patients crée une cascade de conséquences psycho-sociales (11,12) pouvant participer au contexte d'isolement et de moqueries chez les patients porteurs de séquelles faciales de fentes labio-maxillo-palatines (13). Lorsqu'une rééducation orthophonique intensive est insuffisante à corriger tout à fait les conséquences phonatoires de cette IVP, plusieurs techniques chirurgicales sont disponibles pour améliorer l'occlusion vélopharyngée et la phonation : palatoplasties d'allongement vélaire, uvulovélopharyngoplasties (UVPP), et plus récemment, injection pharyngée de graisse autologue ou lipomodélage pharyngé postérieur (LP). Toutes ces techniques ont pour objectif de réduire la distance existante entre le bord postérieur du voile du palais et la paroi pharyngée postérieure afin de faciliter leur contact ferme lorsque nécessaire, notamment lors de la phonation.

De nombreuses techniques de palatoplasties d'allongement vélaire ont été décrites et utilisées depuis des décennies. La technique du push-back a été décrite dans les années 1920 pour la fermeture primaire des fentes vélopalatines, utilisant alors une incision en fer à cheval. L'inconvénient de cette technique est de laisser une zone crantée palatine étendue, antérieure et latérale, responsable d'une rétraction cicatricielle majeure entravant la croissance maxillaire (14) et favorisant l'apparition de fistules buconasales. Cette technique est aujourd'hui principalement utilisée dans le traitement secondaire de l'IVP après la puberté, favorisant alors une incision sulculaire et une fermeture en V-Y, le V étant placé de part et d'autre du canal nasopalatin. Le push-back est alors associé à un lambeau pharyngé postérieur afin de prévenir la récurrence par rétraction cicatricielle (15,16). Par ailleurs, cette procédure ne permet pas la transversalisation des muscles vélaire dans le même temps.

La double plastie en Z, décrite par Leonard Furlow en 1978, consiste en la réalisation d'un double Z, musculomusculaire oral et musculo nasale, dans le but de créer une sangle musculaire de maintien et l'allongement du voile. Cette technique peut être utilisée dans la

chirurgie primaire de fermeture des fentes vélaires comme dans le traitement secondaire de l'IVP (17–20).

Le lambeau de luvette en îlot a initialement été décrit par Delaire et Markus comme une technique additionnelle au moment de la fermeture de la fente du palais osseux en cas de voile court (21). Cette technique permet un recul de la muqueuse orale associé à une véloplastie intravélaire et une fermeture en V-Y, les muscles vélaires restant solidaires de la muqueuse orale lors du recul (Figure 3).

De nombreuses autres techniques ont été décrites, notamment l'utilisation d'un lambeau musculomuqueux de muscle buccinateur (22–25).

Les UVPP avec lambeau pharyngé postérieur, souvent associées à un push-back, sont les actuels gold standards dans le traitement de l'IVP sévère (26–30). Le principe de ces techniques est de réduire le sphincter vélapharyngé en joignant le voile du palais et la paroi pharyngée postérieure : en 1865, Passavant suturait directement le voile du palais à la paroi postérieure, et Schoenborn réalisait le premier lambeau pharyngé postérieur à pédicule inférieur en 1875. Le principe du lambeau pharyngé postérieur est de réduire mécaniquement le passage nasal d'air en cloisonnant une surface large en deux orifices latéraux plus étroits. La technique fut développée au cours des années, notamment avec le lambeau à pédicule supérieur, aujourd'hui largement utilisé, sans qu'aucune supériorité entre pédicule supérieur et inférieur n'ait pu être mise en évidence en termes d'efficacité (31,32).

La sphinctéroplastie dynamique est également une technique largement répandue. Initialement décrite en 1950 par Wilfred Hynes (33), sa première version décrivait l'élévation de deux lambeaux musculomuqueux pharyngés latéraux comprenant muqueuse et muscles salpingopharyngiens. Il y ajoute en 1953 les muscles palatopharyngiens ainsi que des fibres du constricteur supérieur du pharynx (34). Hynes a ensuite spécifié la technique d'élévation des lambeaux mais surtout le site de leur implantation en 1967 (35), qu'il préconise le plus haut possible. Miguel Orticochea publia une version modifiée de cette technique en 1968 (36), dans laquelle les lambeaux sont pédiculés plus bas et sur une surface plus étendue afin de conserver leur innervation donc leur contractilité, et leurs extrémités suturées sous un petit lambeau pharyngé postérieur. Les complications obstructives de cette technique restent similaires à celle des lambeaux pharyngés postérieurs (37).

La technique d'UVPP réalisée dans notre service depuis une vingtaine d'années associe un lambeau pharyngé postérieur inséré en arrière de la luvette, un recul du voile par

lambeau de luvette en îlot selon Delaire (21,38) (Figure 2), et un allongement du voile par suture des piliers postérieurs lorsque jugé nécessaire. Cependant, cette technique est responsable en période post-opératoire de douleurs importantes, parfois d'inconfort à type de sensation d'oppression pharyngée et d'étouffement, ainsi que d'un risque de saignement post-opératoire non négligeable (39) et d'obstruction des voies aériennes supérieures avec apparition d'un syndrome d'apnées obstructives du sommeil (SAOS) (40–42,39).

Les pharyngoplasties d'augmentation ont été décrites comme alternatives à ces techniques plus invasives dans le traitement de l'IVP depuis que Robert Gersuny eut l'idée, en 1900, d'injecter de la Vaseline dans le palais et le pharynx de deux patients atteints de fente afin de réduire leur déperdition nasale d'air (43). De nombreux matériaux ont par la suite été utilisés : paraffine (44,45), silicone (46), collagène (47), Teflon (48), Proplast (49), Gortex (50), hydroxyapatite (51). En plus de leurs complications propres, ces matériaux présentent tous des risques de réaction immunitaire, d'infection, de migration et d'extrusion. L'emploi de graisse autologue dans l'augmentation de la paroi pharyngée postérieure a initialement été décrite par Gaza en 1926, par abord cervical externe. À partir de 1996, Dejonckere and al. ont utilisé des injections de graisse autologue dans le traitement de l'IVP, avec de bons résultats en termes de nasonnement (52). Aujourd'hui, le récent développement de la technique de prélèvement et d'injection de graisse autologue grâce à sa codification par Sydney Coleman (53,54) ainsi que l'accès à des techniques précises d'évaluation telles que l'IRM (55–57) font du lipomodelage pharyngé (LP) postérieur une technique accessible et sûre dans le traitement de l'IVP.

Actuellement utilisé seul dans le traitement des IVP légères, le LP postérieur peut être associé à un allongement vélaire dans le traitement des IVP modérées à sévères. L'efficacité du LP associé à une palatoplastie ou une pharyngoplastie dans les IVP modérées à sévères a été évaluée dans une étude (58), avec de bons résultats rapportés en terme de nasonnement et d'intelligibilité. Cette étude comporte cependant plusieurs biais, en particulier l'absence d'évaluation ou de suivi par un orthophoniste, qui la rendent difficilement généralisable. Par ailleurs, les résultats du LP n'ont encore jamais été comparés à ceux des UVPP.

L'objectif principal de cette étude était de comparer les résultats phonatoires du lipomodelage pharyngé postérieur, associé ou non à une palatoplastie d'allongement vélaire, à ceux de l'UVPP avec lambeau pharyngé à pédicule inférieur dans le traitement de l'IVP.

L'objectif secondaire était d'évaluer ces résultats dans le traitement des IVP modérées à sévères afin d'établir un algorithme décisionnel.

ARTICLE

Could Pharyngeal Fat Injection with Palatal Lengthening be a first-line treatment of velopharyngeal insufficiency? Comparison with Velopharyngoplasty with inferiorly based pharyngeal flap. A retrospective study of 70 cases.

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ABSTRACT

Introduction: Velopharyngeal insufficiency (VPI) affects the communication of patients concerned and their quality of life. Its main cause is the presence of a cleft palate, either syndromic or not. When speech therapy is not sufficient to correct the phonatory consequences of VPI, several surgical techniques are available to try to improve intelligibility: palatal lengthening (PL), velopharyngoplasty (VPP) and more recently, pharyngeal fat injection (PFI). VPP with pharyngeal flap is generally considered as the gold standard in the treatment of moderate to severe VPI, while PL and PFI, alone or in combination, are reserved for mild VPIs. VPP is an effective but invasive surgical procedure leading to significant postoperative pain and the risk of obstructive sleep apnea (OSA) syndrome. PFI, with or without PL, is a less invasive and a well-tolerated procedure, but its efficacy in moderate to severe VPI has not yet been compared to that of VPP. The main objective of this study was to compare the phonatory results of patients with VPI, treated either with PFI, associated or not with PL, or with VPP with an inferiorly based pharyngeal flap.

Material and methods: This retrospective study was comprised of seventy patients with VPI that required surgery. Thirty-five patients were treated with VPP with an inferiorly based pharyngeal flap and 35 patients with PFI more or less associated with PL. All patients were evaluated pre-operatively and 6 months postoperatively by the same speech and language therapist at the University Hospital of Nantes according to the following criteria: Borel-Maisonny score, intelligibility, nasality and nasal air emission.

Results: There was no significant difference between the patients in the two groups according to age at treatment, sex, the presence of a syndrome, the presence of a cleft, or preoperative Borel-Maisonny scores. There was no postoperative complication in any of the patients. The Borel-Maisonny score and all the main speech parameters - intelligibility, nasality, nasal air emission - were improved in both groups, with no significant difference between them. The delay of resumption of oral intake and duration of hospitalization were significantly reduced in the PFI $\pm$ PL group compared to the VPP group.

Conclusion: PFI, whether associated or not with PL, produced phonatory results comparable to those of the VPP with an inferiorly based pharyngeal flap in the treatment of moderate to

severe VPI. PFI $\pm$ PL was responsible for lower postoperative morbidity, resulting in a significant reduction in the delay of resumption of oral intake and duration of hospitalization. A prospective study is now needed to confirm these findings and to define, when appropriate, PFI as a first-line treatment for not only mild but moderate to severe VPI.

INTRODUCTION

Velopharyngeal insufficiency (VPI) is defined by incomplete occlusion of the velopharyngeal sphincter due to an anatomical inadequacy between the actual length of the velum and the length necessary to allow its firm contact with the pharyngeal walls. The muscles that constitute the velopharyngeal sphincter are the levator veli palatini, tensor veli palatini, palatoglossus, palatopharyngeus, musculus uvulae, salpingopharyngeus, and superior pharyngeal constrictor (59). The combined action of these velar and pharyngeal muscles thus plays a role in phonation, suction, protection of the nasal cavity during swallowing and vomiting, hearing, and breathing (1).

The main cause of VPI is the presence of a cleft palate (3), isolated or not, whether syndromic or not, overt or submucous and the consequences of its surgical closure. In the latter case, VPI can be explained by a scarred or short velum and/or a persisting muscular defect creating a velar incompetence. In addition to velopharyngeal insufficiency, there may be a greater or lesser degree of velopharyngeal incompetence with velar and/or pharyngeal hypocontractility due to constitutional muscular weakness, lack of surgical repair of the muscular integrity of the velum, or neurophysiological impairment in the case of an associated syndrome. A submucous cleft palate can induce a perceptible hypernasality even in the event of complete closure of the velopharyngeal sphincter by reducing the thickness of the velum (1).

In the absence of cleft palate, VPI may be secondary to various neurological, anatomical and/or iatrogenic disorders (4). Defects in velum integrity have been linked to more than 400 syndromes, with 22q11.2 deletion syndrome the most common finding (5). After genetic assessment, only 14% of non-cleft VPIs remain without an established cause (6).

Iatrogenic causes are also relatively frequent, with a significant incidence of post-adenoidectomy VPI, especially in patients with a short velum or undiagnosed submucosal cleft (7,8). VPI after maxillary advancement using a LeFort 1 osteotomy has also been described in cleft patients (9).

On the phonatory level, VPI is mainly manifested by hypernasality and nasal air emission. Those signs can be accompanied by compensatory mechanisms such as glottal stops or pharyngeal fricatives, that may affect intelligibility, leading to impaired communication and its cascade of psycho-social consequences (11,12), participating to patients with clefts

facing isolation and mockery (13). When intensive speech therapy is insufficient to completely correct the phonatory consequences of VPI, several surgical techniques are available to improve intelligibility: palatal lengthening (PL), velopharyngoplasty (VPP), and, more recently, pharyngeal fat injection (PFI). All these techniques aim to reduce the distance between the posterior border of the velum and the posterior pharyngeal wall, allowing firmer contact between them when necessary, in particular during speech.

Concerning palatal lengthening, numerous techniques have been described and used for decades. The push back technique has been described as early as the 1920s for cleft palate repair using a horseshoe palatal incision. The disadvantage of this technique is to leave large anterior and lateral denuded areas on the hard palate, resulting in major palatal scar contraction, impaired maxillary growth (14) and oral fistula. Push-back is now commonly associated with a posterior pharyngeal flap in the treatment of VPI after puberty, using a sulcular incision and a V-Y closure (15,16), the V being placed on both sides of the nasopalatine canal.

The double-opposing Z-plasty has been first described in 1978 by Leonard Furlow. This procedure consists in the dissection and redirection of the palatal muscles to produce an overlapping muscle sling and lengthen the velum without using tissue from the hard palate. This technique can be used for primary hard palate closure without pushback or lateral relaxing incisions or for treatment of VPI (17–20).

The uvula island flap has been described by Markus and Delaire as an additional procedure to be performed at the time of primary hard palate closure, in case of short velum (21). It consists in an oral mucosa push-back associated with a intravelar veloplasty, the muscles and the uvula mucosa remaining attached together during the V-Y plasty (Figure 3).

Numerous other procedures have been described such as buccinator myomucosal flaps (22–25).

The VPP with posterior pharyngeal flap, possibly combined with palatal push-back, is the current gold standard in the treatment of moderate to severe VPI (27–30). This procedure is the evolution of techniques aiming to narrow the velopharyngeal sphincter by joining the soft palate to the posterior pharyngeal wall: in 1865, Passavant sutured the posterior border of the velum to the posterior pharyngeal wall and Schoenborn created the first inferiorly based pharyngeal flap in 1875 following the same principle. The posterior pharyngeal flap reduces

the nasal air passage by dividing the large velopharyngeal sphincter into two smaller lateral orifices on both sides of the flap. No significant difference in the results has been shown between superiorly and inferiorly based pharyngeal flaps so far.

Sphincter pharyngoplasty is another commonly used technique: two superiorly based lateral pharyngeal flaps are raised, transposed medially and sutured to each other along the posterior pharyngeal wall. First described in 1950 by Wilfred Hynes (33), it is mostly used for the treatment of velopharyngeal incompetence, ideally for patients with good sagittal motion but poor pharyngeal lateral motion. Its first version mentioned the elevation of the salpingopharyngeus muscle, when the second version in 1953 included the bulk of the palatopharyngeus and fibers of the superior pharyngeal constrictor (34). Hynes then specified the flap elevation and its inset in 1967 (35). Orticochea published a modified version of this technique in 1968 (36), where flaps' pedicles were longer to allow better contractility. The flaps were sutured under a small posterior pharyngeal flap. He later modified the location of the inset to be more superior, which is today a widely spread surgical technique for treatment of VPI. As for the posterior pharyngeal flap, OSA syndrome remains the main long-term complication of these techniques (37).

The VPP procedure carried out in our department for the last two decades associates an inferiorly based pharyngeal flap with an uvula island flap (21,38) (Figure 2) and the suture of the posterior tonsillar pillars, when appropriate. However, this procedure is responsible in the postoperative period for significant pain, sometimes including discomfort like pharyngeal oppression and choking sensation, and can also carry the risk for significant bleeding and obstruction of the upper airways, with the onset of obstructive sleep apnea (OSA) syndrome (40–42,39).

Pharyngeal wall augmentation techniques have been used as an alternative to more invasive surgical procedures for the treatment of VPI since Robert Gersuny first had the idea to inject Vaseline in the palate and the pharynx of two cleft patients to reduce nasal air emission in 1900 (43). Numerous materials have then been used: paraffine (44,45), silicone (46), collagen (47), Teflon (48), Proplast (49), Gortex (50), calcium hydroxyapatite (51). In addition to their own complications, they all present the risk of foreign body reaction, infection, migration and extrusion. The use of autologous fat to thicken the posterior pharyngeal wall was first described by Gaza in 1926 using an external cervical approach. From 1996, Dejonckere and al. used autologous fat injections to treat VPI with good results on nasality (52). Today, the more recent development and clear codification of fat grafting

technique and its assessment, in particular by Sydney Coleman (53,54) and Charles Filip (55–57), made it an accessible and safe treatment for VPI.

PFI, associated or not with PL, is responsible for low morbidity. Its efficacy in moderate to severe VPI, when combined with velopharyngoplasty or palatoplasty, has been evaluated in one study (58) showing good results in terms of nasality and intelligibility, although none of the patients underwent evaluation or treatment by a speech and language therapist, questioning the indication for surgical treatment. However, results after PFI have never been compared with that of VPP.

The main objective of this study was to compare the phonatory results of PFI, whether or not associated with PL, with that of VPP with an inferiorly based pharyngeal flap and an uvula island flap in the treatment of VPI. The secondary objective was to evaluate these results in moderate to severe VPIs in order to establish a decision algorithm.

MATERIALS AND METHODS

Data collection

Seventy patients presenting with mild to severe VPI requiring surgery in the Oral and Maxillofacial Surgery Department of Nantes University Hospital between 2012 and 2017 were included in the study. The data collection was performed using the computerized and paper files of the patients included and only concerned strict routine care. Data were analyzed retrospectively. This non-interventional study did not require the approval of an ethics committee, according to the Articles L. 1121-1 and R 1121-2, paragraph 1 of the French Public Health Code. The information collected was:

- initial characteristics of patients: age, sex, presence of a syndrome, presence and characteristics of a cleft, presence of hearing disorders, retained cause of VPI, primary IVV in cases of cleft palate
- date of completion of the VPP or the first PFI
- in cases of PFI: volume of injected fat, injection site(s), need for a second injection
- operative report
- results of the speech and language assessment (SLA) preoperatively and at 6 months postoperatively
- number of postoperative days until resumption of oral intake and duration of postoperative hospitalization
- occurrence of complications: onset of airways obstruction, signs of infection, and bleeding complications requiring surgical revision.

Patients

This study included 70 French-speaking patients divided into two groups: the PFI $\pm$ PL group treated by PFI combined with PL in 80% and isolated in 20% of cases, and the VPP group treated by VPP associated with an inferiorly based pharyngeal flap and an uvula island flap.

All patients with a cleft, including those whose primary surgery had been performed in another center, were managed according to the following surgical schedule, adapted to each type of cleft:

- Delaire primary cheilorhinoplasty and soft palate repair, more or less associated with an intravelar veloplasty (IVV) according to the Sommerlad technique (60,61), at the age of 5-6 months
- Direct repair of the hard palate between 15 and 18 months of age
- Alveolar cleft repair by gingivoperiostoplasty associated with a cancellous bone grafting between 5 and 7 years of age.

PFI ± PL group

The PFI ± PL group consisted of thirty-five patients who were aged on average 9 years on average (3-29) with a median at 7 years at the time of their first PFI. Of these 35 patients, 23 (65.7%) were born with a cleft and 12 (34.3%) without a cleft. The retained causes of VPI in the absence of cleft were 7 idiopathic VPIs, 2 congenital VPIs associated with non-cleft syndrome and 3 post-adenoidectomy iatrogenic VPIs. Of the 23 patients with a cleft, 3 had a submucosal occult cleft palate and 20 had an open cleft: 14 cleft palate and 6 cleft lip and palate including one without maxillary cleft. Seven out of 23 patients (30,4%) had undergone a primary IVV and two patients a secondary IVV.

In the PFI ± PL group, 13 patients had an identified genetic syndrome confirmed by a modified karyotype with 5 cases of 22q11.2 deletion syndromes (velocardiofacial syndrome, VCFS), 4 Robin sequences (PRS), 1 neurofibromatosis type 1, 1 Van Der Woude syndrome associated with Cowden syndrome, 1 CAMT-1 syndrome, and 1 Klinefelter syndrome. Two patients had a global psychomotor delay with normal karyotype.

VPP Group

The VPP group consisted of thirty-five patients whose VPI had been treated with VPP with an inferiorly based pharyngeal flap and an uvula island flap. These patients were selected from all the patients who had been treated with VPP and posterior pharyngeal flap for VPI between 2012 and 2017 in the Department of Maxillofacial Surgery and Stomatology of the University Hospital of Nantes in order to be comparable to the PFI ± PL group according to

sex and age of care: 16 males and 19 females, averaging 6.6 years (3-29) with a median of 6 years when the VPP with pharyngeal flap was performed.

Eight out of 35 patients (22.9%) were born without a facial cleft: the retained causes of VPI were 2 idiopathic VPIs, 5 congenital VPIs associated with a non-cleft syndrome, and 1 iatrogenic post-tonsillectomy VPI. Of the 27 patients with a cleft (77.1%), 3 (11.1%) had a submucosal occult cleft palate and 24 (88.9%) had an open cleft palate: 16 cleft palate, 8 cleft lip and palate including 1 with no associated maxillary cleft.

In the VPP group, 10 patients had an identified genetic syndrome and 3 patients had delayed acquisition, associated in one case with global hypotonia, with normal karyotype. The identified syndromes were 3 22q11.2 deletion syndromes, 3 Robin sequences including 1 Stickler syndrome, 1 Cornelia de Lange syndrome, 1 fetal sodium valproate syndrome, 1 West syndrome and 1 Stilling-Duane retraction syndrome.

Five of the eight patients with non-cleft VPI had a syndrome, whether identified or not: 2 22q11.2 deletion syndromes, Cornelia de Lange syndrome, West syndrome and psychomotor retardation associated with global hypotonia.

Surgical Procedures

PFI (Figure 1)

The surgical procedure was performed under general anesthesia. The patient was in supine position with the head in hyperextension and with oral endotracheal intubation. The intubation tube and the tongue were held back using a Dott and Kilner mouth gag (62).

Abdominal, gluteal or knee-fat removal was the first part of the procedure: after sterile draping and local superficial infiltration with 0.05% epinephrine serum, a 3mm cutaneous incision was made in the umbilicus, gluteal fold or the inner side of the knee using a cold blade 11. A liposuction cannula attached to a sterile 10 ml syringe maintained under negative pressure was introduced through this incision. Approximately 10 to 15 ml of fat were aspirated.

The sample syringes were then centrifuged at 3000 rpm for 3 minutes to separate the fat from the serum, blood and lysed adipocytes using the Coleman technique (54). These were then removed manually from the syringe. The isolated fat was then transferred to 1 ml sterile syringes. The second step of the procedure consisted of the intra-oral injection of the fat using a 20-gauge reinjection cannula, manually bent until the optimum angle for injection was obtained, taking care not to obstruct the passage of fat. After inspection and palpation of the posterior pharyngeal wall in search of aberrant courses of the internal carotid arteries, epinephrine serum was injected in the nasopharyngeal mucosa, and a 1 millimeter incision of the mucosa was performed on the midline at the level of the anterior part of atlas vertebra.

The reinjection cannula was then introduced through this incision, after retraction of the soft palate with a malleable blade, to perform a superficial intramuscular injection, directly under the pharyngeal mucosa. The fat was first injected into the median line of the posterior pharyngeal wall, then into the paramedian areas in order to obtain a satisfactory circular reduction of the velopharyngeal orifice. In some cases, it could be completed by the median and paramedian injection into the posterior part of the soft palate and into the posterior tonsillar pillars in case the immediate result was considered insufficient and the quantity of fat harvested allowed it. The immediate surgical result was assessed directly. Upon removal of the cannula, the incision was closed with an absorbable thread stitch to prevent the escape of the injected fat.

Patients were routinely hospitalized for at least 1 night to monitor and relieve postoperative pain, monitor resumption of oral intake, and onset of airway obstruction. Since PFI was considered an autologous tissue transplant, all patients received perioperative and postoperative amoxicillin-clavulanic acid orally for 5 days.

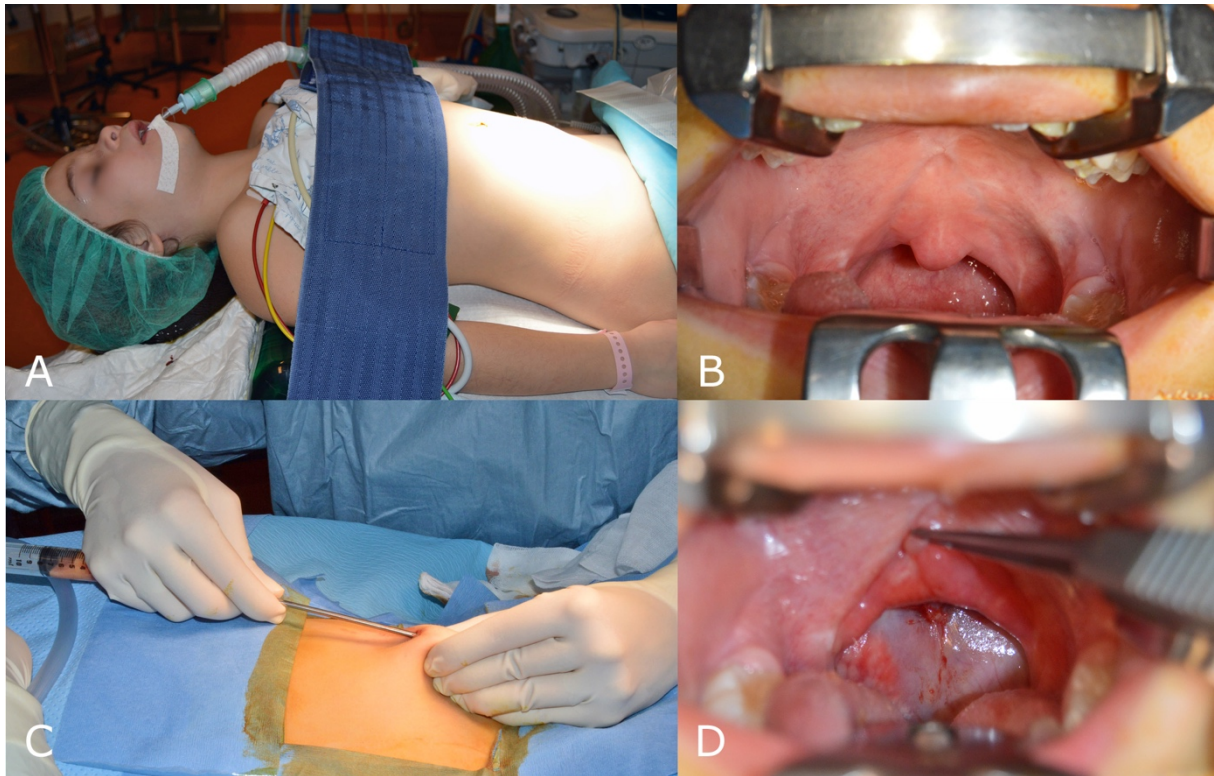


Figure 1 Pharyngeal fat injection in a child previously treated with PFI and an uvula island flap with V-Y pushback of velum. A. Supine position with the head in hyperextension and with oral endotracheal intubation. B. Exposition of the oropharynx with a Kilner Dott mouth gag. The Y scare can be observed on the velum C. Harvesting of the peri-umbilical fat. d. Direct visualization of the injected fat in the posterior pharyngeal wall, after retraction of the uvula.

VPP with an inferiorly based pharyngeal flap and an uvula island flap (Figure 2)

The installation was similar to that used for PFI. After forming a denuded area behind the uvula by mucosal excision, an uvula island flap with a V-Y plasty of the velum was performed with detachment of the muscles from the nasal mucosa, leaving them attached to the oral mucosa during the V-Y plasty, according to Delaire's technique.

An inferiorly based pharyngeal musculo-mucosal flap was then raised and attached behind the uvula and laterally to the posterior pillars of the velum. The palatal plane was then closed with a V-Y plasty to lengthen the velum (38).

Bilateral nasopharyngeal tubes were put in place at the end of the procedure and left in place for the two post-operative nights to prevent a mucosal synechia.

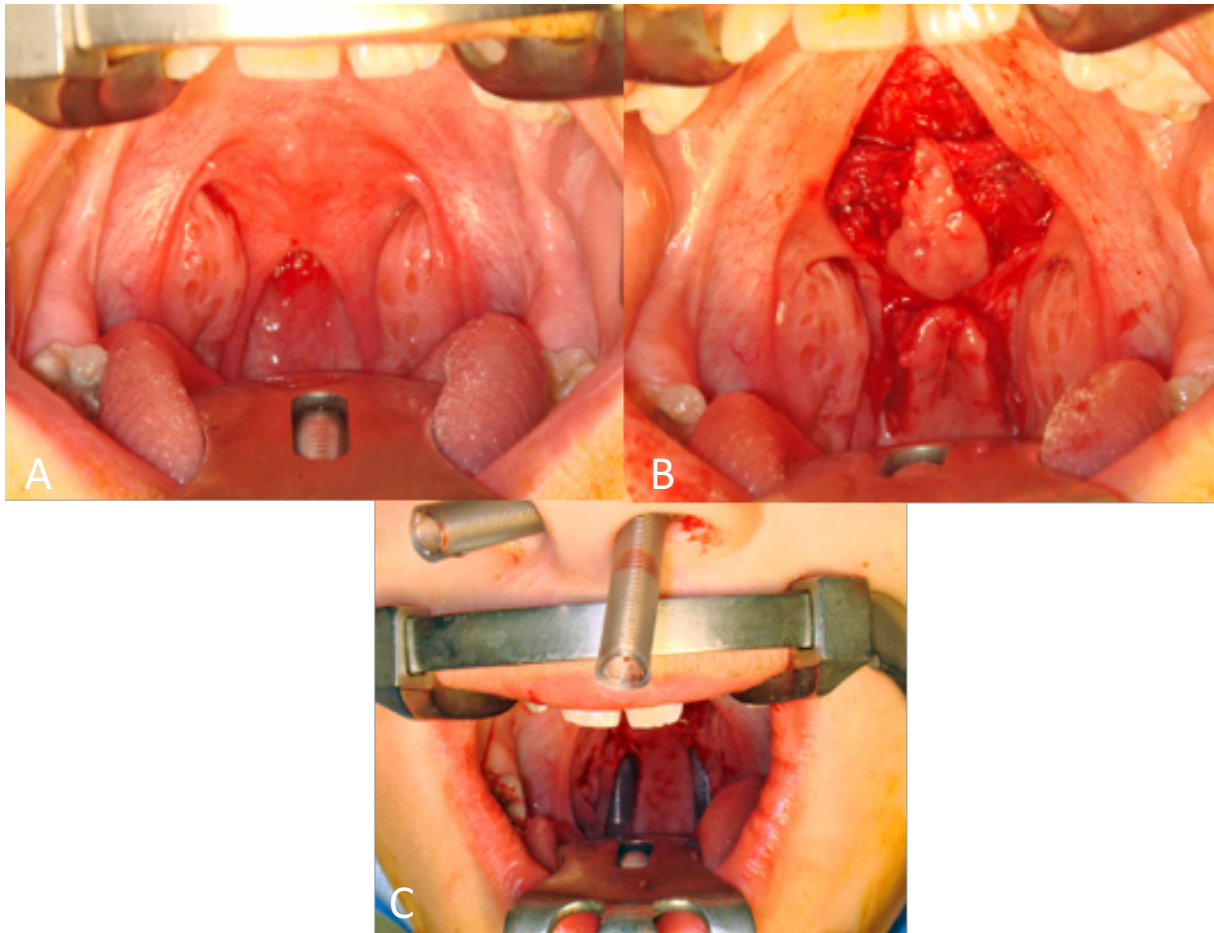


Figure 2. VPP with an inferiorly based pharyngeal flap and an uvula island flap according to Delaire's technique in a patient previously treated for a cleft palate. A. Exposition of the oropharynx with a Kilner Dott mouth gag B. Uvula island flap with V-Y pushback of the velum muscles. Suture of pharyngeal flap to the posterior part of the uvula. C. Inferiorly based posterior pharyngeal flap with nasopharyngeal tubes in place.

Palatal lengthening

Palatal lengthening was associated with PFI using either an uvula island flap with a V-Y plasty of the velum (Figure 3) or an IVV according to the Sommerlad technique (60,61) associated with a Z plasty of the oral mucosa according to Talmant (Figure 4).

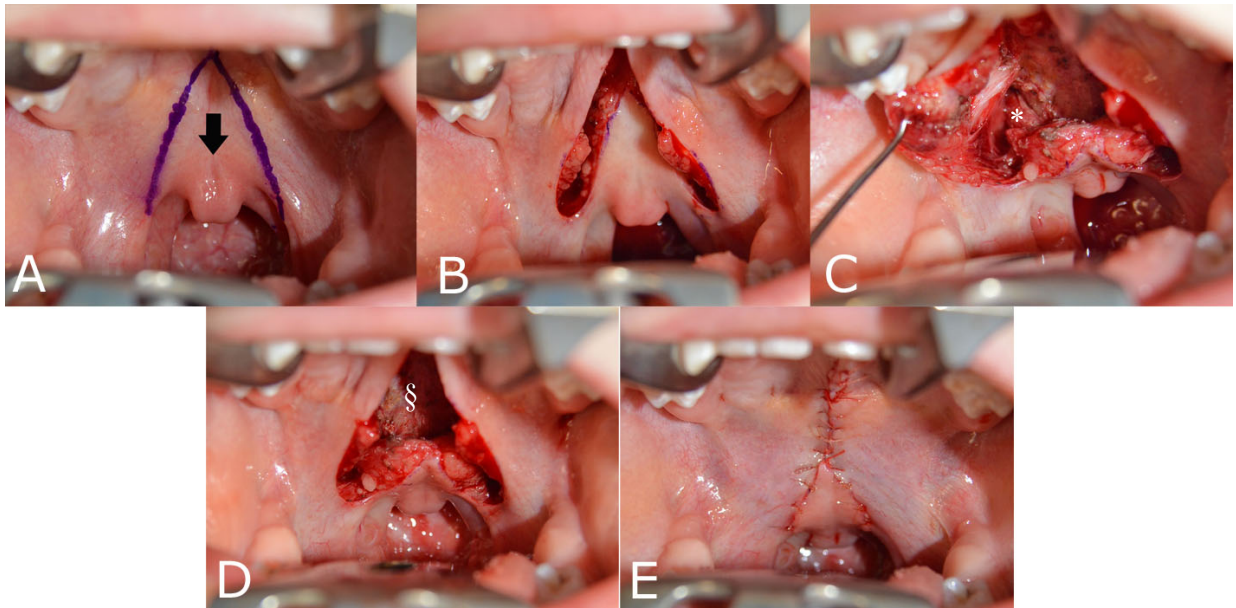


Figure 3. Uvula island flap with V-Y plasty. A. Exposition of the velum using a Dott and Kilner mouth gag; drawing of the incisions. The arrow represents the direction of the uvula island flap B. Incision of palatal mucosa. C. Subperiosteal dissection and intravelar veloplasty. The * locates the levator veli palatini muscle. D. Push-back of the velum once the muscles were separated from the nasal mucosa. The § locates the nasal mucosa. E. Lengthened velum after V-Y closure.

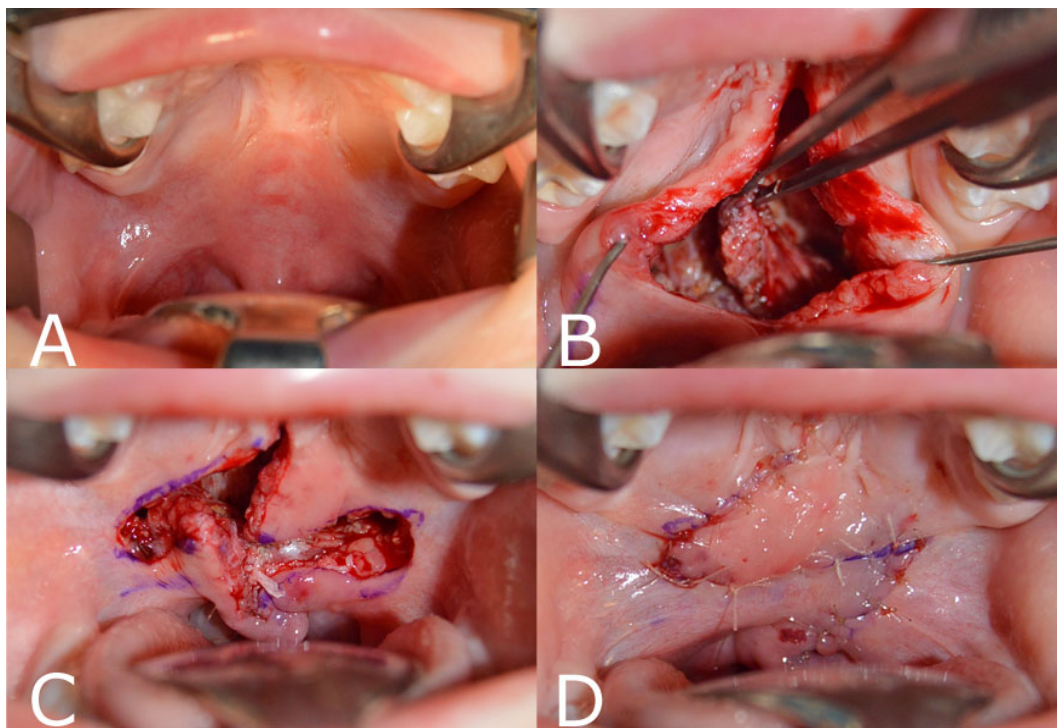


Figure 4. Z plasty of oral mucosa and IVV for velar lengthening. A. Exposition of the oropharynx: short velum and scar from a previous cleft palate repair. B. Individualization of left velar muscles through intravelar veloplasty. C. Z flaps after push-back of the muscles. D. Lengthened velum after closure.

Associated procedures

The associated procedures were classified as follows:

- the principal associated procedures directly related to the action researched on the velopharyngeal sphincter and/or the phonation, isolated or associated with each other: V-Y plasty of the velum with an island uvula flap, VIV according to the Sommerlad technique with Z plasty of the oral mucosa, re-closure of the cleft palate, removal of a previous posterior pharyngeal flap; in this group, we added the closure of a bucconasal fistula in three patients: associated with a IVV in 2 cases and a re-closure of the velum in 1 case.
- Secondary associated procedures resulting in widening of the velopharyngeal sphincter: adenoidectomy, tonsillectomy
- the secondary procedures that theoretically had no consequence on the velopharyngeal sphincter but were considered in order to take into account their role in postoperative pain and potentially delayed oral intake

Previous procedures on the velopharyngeal sphincter

Prior procedures to VPI or VPP were also noted if these were related to the velopharyngeal sphincter: VPP, PFI, PL, IVV, re-repair of cleft palate.

Exploration of velopharyngeal insufficiency

The analysis of the function of the velopharyngeal sphincter was performed by a clinical examination and a pre-operative speech and language assessment preoperatively and at 6 months postoperatively.

The clinical examination was performed by the surgeon in charge of the procedure.

The examination of the velum included:

- assessment of the soft palate at rest
 - length
 - distance to posterior pharyngeal wall, depth of the cavum
 - occult submucosal cleft palate signs: bifid uvula, mucosal notch, medial weakness on palpation

- appearance of the mucosa (soft, scarred)
- assessment of the soft palate during speech (vowel "A" maintained):
 - contractility
 - appearance of the velum (soft, scarred)
 - deviation of the uvula
 - adaptive hypertrophy of the pharyngeal muscles: Passavant's ridge, lateral pharyngeal muscles

A more specific clinical examination by a genetician and X-rays were also performed when an associated syndrome was suspected.

The oral examination was systematically completed by an ENT examination, with hearing disorders assessment, otoscopy, and nasofibroscope if required.

The speech and language assessment was performed by a speech and language therapist with extensive experience evaluating patients with VPI and cleft palate (S. Balandier). It systematically included a specific history and a classification of the cleft palate where appropriate. Data concerning the SLA were collected in a simplified form for decision making (Figure 5).

The clinical examination included static and dynamic observation of the oral sphere to diagnose morphological and functional abnormalities of the lips, nose, teeth, tongue, palate, velum and cavum. The examination of the articulation and the speech was done by the repetition of phonemes, syllables, sentences, and a text, as well as by the naming of images, the narrative on images for the youngest patients, the reading of a text, recitation, song, conversation, and comparing the pre- and post-operative recording. The quality of the articulation, the timbre of the voice, the intelligibility and the flow were studied. The diagnosis was then made, reflected by the Borel-Maisonny score (Table 1).

Regional University Hospital of Nantes
MAXILLOFACIAL SURGERY AND STOMATOLOGY DEPARTMENT

First name: _____
 Name: _____
 Date of birth:..../..../.... Date of examination:..../..../....

ORTHOPHONIC EVALUATION OF PHONATION

☐ After primary surgery
☐ After speech therapy
☐ After secondary surgery

Hearing: assessed: yes no
 good: yes no
 follow-up: yes no

~~~~~

|                           |      |      |          |        |
|---------------------------|------|------|----------|--------|
|                           | None | Mild | Moderate | Severe |
| <b>Nasality</b>           |      |      |          |        |
| <b>Nasal air emission</b> |      |      |          |        |
| <b>Dysphonie</b>          |      |      |          |        |

|                        |      |              |     |
|------------------------|------|--------------|-----|
|                        | Good | Intermediate | Bad |
| <b>Intelligibility</b> |      |              |     |

|                     |        |           |                |
|---------------------|--------|-----------|----------------|
|                     | Normal | Disturbed | Very disturbed |
| <b>Articulation</b> |        |           |                |

|                       |                              |         |        |           |
|-----------------------|------------------------------|---------|--------|-----------|
| <b>Oral vowels</b>    | <i>open</i><br><i>closed</i> | a<br>ou | è<br>é | o<br>i    |
| <b>Nasal vowels</b>   |                              | an      | on     | in        |
| <b>Plosives</b>       | <i>thud</i><br><i>loud</i>   | p<br>b  | t<br>d | k<br>g    |
| <b>Fricatives</b>     | <i>thud</i><br><i>loud</i>   | f<br>v  | s<br>z | ch<br>j   |
| <b>Other phonemes</b> |                              | l<br>r  | m<br>n | gn<br>ill |

**Compensatory articulation productions:** no yes      **Glottal stops** ☐  
**Facial muscle compensation:** ☐ none      **Pharyngeal fricatives** ☐  
☐ nasal wings only  
☐ nose and face, light  
☐ nose and face, moderate  
☐ nose and face, severe

**Evaluation by entourage:** ☐ no difficulty to be understood  
☐ sometimes has to repeat  
☐ often has to repeat  
☐ phonatory fatigability

**Borel-Maisonny score:**

|                                  |                                |                                   |                                |
|----------------------------------|--------------------------------|-----------------------------------|--------------------------------|
| <input type="checkbox"/> Ph I    | <input type="checkbox"/> + art |                                   |                                |
| <input type="checkbox"/> Ph II b | <input type="checkbox"/> + art | <input type="checkbox"/> Ph III b | <input type="checkbox"/> + art |
| <input type="checkbox"/> Ph II m | <input type="checkbox"/> + art | <input type="checkbox"/> Ph III m | <input type="checkbox"/> + art |

**Observations:** .....

*Sophie Balandier - Orthophoniste*

Figure 5. Speech and Language assessment form used by the speech and language therapist with extensive experience in evaluating VPI.

*Table 1. Borel-Maisonny score: subjective evaluation of hypernasality and intelligibility*

| <b>Score</b> | <b>Definition</b>                                                            |
|--------------|------------------------------------------------------------------------------|
| <b>I</b>     | Normal phonation, no nasal air emission                                      |
| <b>I-II</b>  | Good phonation, intermittent nasal air emission, good intelligibility        |
| <b>IIb</b>   | Phonation with continuous nasal emission but good intelligibility            |
| <b>IIm</b>   | Phonation with continuous nasal emission and poor intelligibility            |
| <b>IIIb</b>  | Phonation with continuous compensatory articulation but good intelligibility |
| <b>IIIm</b>  | Phonation with continuous compensatory articulation and poor intelligibility |

### **Statistical Tests**

The statistical tests used were non-parametric tests: Wilcoxon-Mann-Whitney for continuous variables, Fisher for non-ordinal categorical variables and Kolomogorov-Smirnov for ordinal categorical variables.



## RESULTS

### Patients

There was no significant difference between the characteristics of the patients in the two groups regarding age at treatment, sex, number of syndromes, presence of a cleft, presence of an associated auditory disorder and assumed cause of VPI (Table 2. Patient's characteristics).

*Table 2. Patient's characteristics*

|                        |    | PFI ± PL |           |    | VPP  |           |         |  |
|------------------------|----|----------|-----------|----|------|-----------|---------|--|
|                        | N  | %        | CI95%     | n  | %    | CI95%     | p-value |  |
| Age at time of surgery |    |          |           |    |      |           |         |  |
| (years)                |    | 6        | 7-11      |    | 6    | 5.8-7.4   | 0,1326  |  |
| Sex                    |    |          |           |    |      |           |         |  |
| Male                   | 19 | 54.3     | 38.2-69.5 | 16 | 45.7 | 30.5-61.8 | 0.63294 |  |
| Female                 | 16 | 45.7     | 30.5-61.8 | 19 | 54.3 | 38.2-69.5 |         |  |
| Syndrome               |    |          |           |    |      |           |         |  |
| No                     | 20 | 57.1     | 40.9-72   | 22 | 62.9 | 46.3-76.8 | 0.80752 |  |
| Yes                    | 15 | 42.9     | 28-59.1   | 13 | 37.1 | 23.2-53.7 |         |  |
| Hearing disorder       |    |          |           |    |      |           |         |  |
| No                     | 28 | 80       | 64.1-90   | 28 | 84.8 | 69.1-93.3 | 0.75312 |  |
| Yes                    | 7  | 20       | 10-35.9   | 5  | 15.2 | 6.7-30.9  |         |  |
| Cleft palate           |    |          |           |    |      |           |         |  |
| No                     | 12 | 34.3     | 20.8-50.8 | 8  | 22.9 | 12.1-39   | 0.42786 |  |
| Yes                    | 23 | 65.7     | 49.2-79.2 | 27 | 77.1 | 61-87.9   |         |  |
| Cause                  |    |          |           |    |      |           |         |  |
| Idiopathic             | 4  | 11.4     | 4.5-26    | 2  | 5.7  | 1.6-18.6  | 0.59698 |  |
| Congenital             | 5  | 14.3     | 6.3-29.4  | 5  | 14.3 | 6.3-29.4  |         |  |
| Cleft with syndrome    | 10 | 28.6     | 16.3-45.1 | 8  | 22.9 | 12.1-39   |         |  |
| Cleft without syndrome | 13 | 37.1     | 23.2-53.7 | 19 | 54.3 | 38.2-69.5 |         |  |
| Iatrogenic             | 3  | 8.6      | 3-22.4    | 1  | 2.9  | 0.1-14.5  |         |  |

|                        |               | PFI ± PL |      |           | n  | VPP  |           | p-value |
|------------------------|---------------|----------|------|-----------|----|------|-----------|---------|
|                        |               | N        | %    | CI95%     |    | %    | CI95%     |         |
| Primary IVV            |               |          |      |           |    |      |           |         |
|                        | No            | 27       | 77.1 | 61-87.9   | 23 | 67.6 | 50.8-80.9 | 0.42786 |
|                        | Yes           | 8        | 22.9 | 12.1-39   | 11 | 32.4 | 19.1-49.2 |         |
| Previous procedure     |               |          |      |           |    |      |           |         |
|                        | None          | 26       | 74.3 | 57.9-85.8 | 33 | 94.3 | 81.4-98.4 | 0.0372  |
| With action on the VPS |               | 5        | 14.3 | 6.3-29.4  | 2  | 5.7  | 1.6-18.6  |         |
|                        | Adenoidectomy | 4        | 11.4 | 4.5-26    | 0  | 0    | 0-9.9     |         |

*CI95%: confidence interval 95%; IVV: intravelar veloplasty; n: number of patients; PFI: pharyngeal fat injection; PL: palatal lengthening; VPP: velopharyngoplasty; VPS: velopharyngeal sphincter.*

### Previous procedures on the velopharyngeal sphincter

Previous procedures to PFI or VPP were also noted if they were related to the velopharyngeal sphincter and are summarized in Table 3.

In the PFI ± PL group, those procedures were:

- 1 Orticochea sphincteroplasty with an IVV
- 1 VPP with an uvula island flap
- 1 PFI with IVV and PL (in another center without any operative report available)
- 1 IVV
- 1 re-closure of the velum using the Malek technique

The time between previous previous procedures and PFI ± PL was 35.8 months on average (8-84 months).

In the VPP group with inferiorly based posterior pharyngeal flap, 2 patients had undergone a PL with an uvula island flap and V-Y plasty, respectively 44 and 52 months before the posterior pharyngeal flap was performed (48 months on average).

Table 3. Previous procedures on the velum in the two groups

| Procedure                        | PFI ± PL group (n) | VPP group (n) |
|----------------------------------|--------------------|---------------|
| <b>PL with uvula island flap</b> | 1                  | 2             |
| <b>PFI + IVV + PL</b>            | 1                  | 0             |
| <b>IVV</b>                       | 1                  | 0             |
| <b>Re-repair of the velum</b>    | 1                  | 0             |
| <b>Orticochea + IVV</b>          | 1                  | 0             |
| <b>Total</b>                     | 5                  | 2             |

*IVV: intravelar veloplasty; n: number of patients; PFI: pharyngeal fat injection; PL: palatal lengthening; VPP: velopharyngoplasty.*

### Associated procedures

Procedures carried out in the PFI ± PL group are summarized in Table 4:

- PFI was associated with IVV using the Sommerlad technique with Z plasty of the oral mucosa in 40% of the cases (14/35), with PL using an uvula island flap and V-Y plasty in 37% of these cases (13/35) and a re-repair of the velum with closure of bucconasal communication without PL in 1 case (3%)
- PFI was isolated in 20% of cases (7 patients)
- the secondary actions that theoretically did not have any consequence on the velopharyngeal sphincter were: 1 genioplasty, 1 bilateral ear tubes insertion, 1 gingivoperiostoplasty with cancellous bone grafting, and 1 secondary cheiloplasty

The eight PFIs without PL were performed in patients whose velum length was deemed sufficient in pre- and intra-operative clinical examination:

- 3 patients with a VPI with a normal velum, idiopathic in 1 case, associated with a NF1 in 1 case and post-adenoidectomy in the third case
- 2 patients, with cleft palate, had undergone IVV 10 and 13 months before the first PFI. This IVV had been associated with a dynamic sphincteroplasty Orticochea for one of them who was suffering from Pierre Robin Sequence.
- 1 patient with psychomotor retardation had undergone an IVV with PFI and PL in another center, 19 months before their first PFI in our department

- 1 patient with a velar cleft within the scope of a Van der Woude's syndrome associated with Cowden syndrome had undergone a PL with an island uvula flap and V-Y plasty of the oral mucosa 13 months before the first PFI
- 1 patient with a right cleft lip and palate operated on multiple times in another center (no operative report available) and whose first PFI was performed at the age of 27

*Table 4. Details of procedures carried out in the PFI ± PL group*

| <b>Cause</b>               | <b>Syndrome</b> | <b>Prior procedure</b>  | <b>Procedure</b> |
|----------------------------|-----------------|-------------------------|------------------|
| <b>1</b> Cleft             | 0               | 0                       | PFI+PL           |
| <b>2</b> Cleft             | 0               | 0                       | PFI+PL           |
| <b>3</b> Idiopathic        | 0               | 0                       | PFI+PL           |
| <b>4</b> Cleft/Syndromic   | PRS             | IVV + Orticochea        | PFI              |
| <b>5</b> Cleft             | 0               | 0                       | PFI+PL           |
| <b>6</b> Cleft             | 0               | 0                       | PFI+PL           |
| <b>7</b> Cleft/Syndromic   | VCFS            | 0                       | PFI+PL           |
| <b>8</b> Cleft/Syndromic   | VCFS            | 0                       | PFI+PL           |
| <b>9</b> Cleft             | 0               | IVV                     | PFI              |
| <b>10</b> Syndromic        | NF1             | 0                       | PFI              |
| <b>11</b> Cleft            | 0               | VPP                     | PFI+PL           |
| <b>12</b> Adenoidectomy    | 0               | 0                       | PFI+PL           |
| <b>13</b> Idiopathic       | 0               | 0                       | PFI+PL           |
| <b>14</b> Cleft            | 0               | 0                       | PFI+PL           |
| <b>15</b> Cleft/Syndromic  | VCFS            | 0                       | PFI+PL           |
| <b>16</b> Cleft            | 0               | 0                       | PFI              |
| <b>17</b> Idiopathic       | 0               | 0                       | PFI+PL           |
| <b>18</b> Cleft /Syndromic | VDW + Cowden    | Uvula island flap + V-Y | PFI              |
| <b>19</b> Syndromic        | CAMT-1          | 0                       | PFI+PL           |
| <b>20</b> Cleft            | 0               | 0                       | PFI+PL           |
| <b>21</b> Idiopathic       | 0               | 0                       | PFI              |
| <b>22</b> Syndromic        | Klinefelter     | 0                       | PFI+PL           |
| <b>23</b> Cleft            | 0               | 0                       | PFI              |
| <b>24</b> Cleft/Syndromic  | VCFS            | 0                       | PFI+PL           |

|           |                 |                            |                |        |
|-----------|-----------------|----------------------------|----------------|--------|
| <b>25</b> | Adenoidectomy   | 0                          | 0              | PFI+PL |
| <b>26</b> | Cleft/Syndromic | PRS                        | 0              | PFI+PL |
| <b>27</b> | Syndromic       | VCFS                       | 0              | PFI+PL |
| <b>28</b> | Cleft           | 0                          | 0              | PFI+PL |
| <b>29</b> | Cleft/syndromic | Psychomotor<br>retardation | 0              | PFI+PL |
| <b>30</b> | Cleft/Syndromic | PRS/Stickler               | 0              | PFI+PL |
| <b>31</b> | Cleft           | 0                          | 0              | PFI+PL |
| <b>32</b> | Cleft/Syndromic | PRS                        | 0              | PFI+PL |
| <b>33</b> | Cleft           | 0                          | 0              | PFI+PL |
| <b>34</b> | Syndromic       | Psychomotor<br>retardation | PFI + IVV + PL | PFI+PL |
| <b>35</b> | Adenoidectomy   | 0                          | 0              | PFI    |

*IVV: intravelar veloplasty; n: number of patients; PFI: pharyngeal fat injection; PL: palatal lengthening; PRS: Pierre Robin sequence; VCF: velocardiofacial syndrome; VDW: Van Der Woude.*

In the VPP group :

- 33 out of 35 patients underwent uvula island flaps with V-Y plasty associated with posterior pharyngeal flap and 2 patients also with closure of buconasal communication
- 4 patients underwent a bilateral tonsillectomy at the same time
- the secondary procedures without action on the velopharyngeal sphincter had been: 1 gingivoperiostoplasty, 1 glossoplasty, 1 secondary cheiloplasty, 2 bilateral ears tubes, and 1 tooth extraction with cauterization of the Kiesselbach's plexus.

An uvula island flap with V-Y plasty was not performed in two patients: the first because the flap was made simultaneously with repair of the cleft palate, and the second because VPP with an uvula island flap and V-Y plasty had already been carried out beforehand and resulted in a velum of satisfactory length.

## Speech and language assessment (SLA)

Preoperative Borel-Maisonny scores were comparable between the two groups and distributed as follows: 17 IIB (48.6%), 12 IIm (34.3%), 1 IIb (2.9%) and 5 IIIm (14.2%) in the PFI  $\pm$  PL group and 16 IIB (45.7%), 10 IIm (28.6%), 1 IIb (2.9%) and 8 IIIm (22.8%) in the VPP group (Table 5, Table 6). There was no significant difference between the two groups ( $p=0.69$ ).

The postoperative Borel-Maisonny score was improved in both groups with no significant difference between them (Tables 5, 6 and 7). It was distributed as follows: 1 I (3%), 1 I-II (3%), 25 IIB (71%), 5 IIm (14%), 2 IIb (6%) and 1 IIIm (3%) in the PFI  $\pm$  PL group and 4 I (11%), 0 I-II, 24 IIB (69%), 1 IIm (3%), 2 IIb (6%) and 4 IIIm (11%) in the VPP group. The evolution of Borel-Maisonny scores was classified as "improvement", "stability" or "aggravation". There was no significant difference in the evolution of the Borel-Maisonny score between the two groups according to this evolution criterion. The Borel-Maisonny score was improved in 34.3% and stable in 65.7% of cases in the PFI  $\pm$  PL group. It was improved in 48.6%, stable in 48.6% and aggravated in 2.9% (1 patient) cases in the VPP group. The case involving a worsening of the score in the VPP group involved a patient with an isolated incomplete cleft palate who had received a primary IVV and then a posterior pharyngeal flap associated with an uvula island flap and buconasal fistula closure. The Borel-Maisonny score had evolved from IIm to IIb because of the presence of a glottal stop during the postoperative assessment. Nasality and nasal air emission stagnated as intelligibility improved, moving from "average" to "good".

Table 5. Evolution of the Borel-Maisonny score in the PFI  $\pm$  PL group

|              |      | Postoperative |      |    |     |     |      |
|--------------|------|---------------|------|----|-----|-----|------|
|              |      | I             | I-II | Ib | IIm | IIb | IIIm |
| Preoperative | I    | 0             | 0    | 0  | 0   | 0   | 0    |
|              | I-II | 0             | 0    | 0  | 0   | 0   | 0    |
|              | Ib   | 0             | 1    | 16 | 0   | 0   | 0    |
|              | IIm  | 1             | 0    | 7  | 5   | 0   | 0    |
|              | IIb  | 0             | 0    | 0  | 0   | 1   | 0    |
|              | IIIm | 0             | 0    | 2  | 0   | 1   | 1    |

Table 6. Evolution of the Borel-Maisonny score in the VPP group

|                     |              | Postoperative |      |     |      |      |       |
|---------------------|--------------|---------------|------|-----|------|------|-------|
|                     |              | I             | I-II | IIb | IIIm | IIIb | IIIIm |
| <b>Preoperative</b> | <b>I</b>     | 0             | 0    | 0   | 0    | 0    | 0     |
|                     | <b>I-II</b>  | 0             | 0    | 0   | 0    | 0    | 0     |
|                     | <b>IIb</b>   | 4             | 0    | 12  | 0    | 0    | 0     |
|                     | <b>IIIm</b>  | 0             | 0    | 8   | 1    | 1    | 0     |
|                     | <b>IIIb</b>  | 0             | 0    | 1   | 0    | 0    | 0     |
|                     | <b>IIIIm</b> | 0             | 0    | 3   | 0    | 1    | 4     |

The IIb phonations seemed the most difficult to improve in both groups: they stagnated in 94% (16/17) of cases in the PFI  $\pm$  PL group and 75% (12/16) in the VPP group. IIIm phonations evolved to I phonation in one case (7.6%), to IIb in 54% (7/13), and stagnated in 38.5% of cases in the PFI  $\pm$  PL group. They evolved to phonation IIb in 80% (8/10), stagnated in 10% and worsened in 10% of cases in the VPP group. IIIIm phonations evolved to phonation IIb in 37,5% (3/8), to IIIb in 12,5% (1/8) and stagnated in 50% of cases in the VPP group. They evolved to phonation IIb in 50% (2/4), to IIIb in 25% (1/4) and stagnated in 25% of cases in the PFI  $\pm$  PL group. In both groups, all postoperative IIIIm phonations were already IIIIm preoperatively.

The intelligibility, nasality and nasal air emission were improved postoperatively in both groups, with no significant difference between them (Table 7).

The intelligibility was improved in 42.9% and stable in 54.3% of the cases in the PFI  $\pm$  PL group and improved in 34.3% and stable in 62.9% of the cases in the VPP group; there was a worsening of intelligibility in one patient in each group post-operatively. In the case of the PFI  $\pm$  PL group, it was a patient presenting with a complete isolated unilateral cleft lip and palate, with a IIb phonation and whose intelligibility had evolved from "good" to "average". The Borel-Maisonny score remained stable and the other parameters had improved (nasality, nasal air emission) excepted articulation, which had remained very disturbed, and could explain the impairment of intelligibility. In the VPP group, this was a patient with a complete cleft palate in a context of Depakine fetal poisoning syndrome, with IIIm phonation and whose

intelligibility had evolved from "average" to "poor". All the other speech parameters (Borel-Maisonny score, nasality, nasal air emission, articulation) had improved.

Nasality was improved in 71.4% and stable in 28.6% of cases in the PFI ± PL group and improved in 80% and stable in 20% of cases in the VPP group.

Nasal air emission was improved in 77.1%, stable in 20% and worsened in 2.9% (1 patient) cases in the PFI ± PL group. It was improved in 83% and stable in 17% of cases in the VPP group.

The resumption of oral intake was significantly faster in the PFI ± PL group compared to the VPP group (median 1, 95% CI (0-1) vs median 1, 95% CI (1-2),  $p < 0.05$ ) (Table 7).

The duration of hospitalization was significantly reduced in the PFI ± PL group compared to the VPP group (median of 2, 95% CI (1-2) vs median of 2, 95% CI (2-2),  $p < 0.05$ ) (Table 7).

*Table 7. Comparison of the results between the two groups: Borel-Maisonny score, intelligibility, nasality, nasal air emission, duration of hospitalization and resumption of oral intake*

|                      |    | PFI ± PL |           |    | VPP  |           |         |  |
|----------------------|----|----------|-----------|----|------|-----------|---------|--|
|                      | n  | %        | CI95%     | n  | %    | CI95%     | p-value |  |
| Borel-Maisonny score |    |          |           |    |      |           |         |  |
| Improvement          | 12 | 34.3     | 20.8-50.8 | 17 | 48.6 | 33-64.4   | 0.86743 |  |
| Stability            | 23 | 65.7     | 49.2-79.2 | 17 | 48.6 | 33-64.4   |         |  |
| Aggravation          | 0  | 0        | 0-9.9     | 1  | 2.9  | 0.1-14.5  |         |  |
| Intelligibility      |    |          |           |    |      |           |         |  |
| Improvement          | 15 | 42.9     | 28-59.1   | 12 | 34.3 | 20.8-50.8 | 1       |  |
| Stability            | 19 | 54.3     | 38.2-69.5 | 22 | 62.9 | 46.3-76.8 |         |  |
| Aggravation          | 1  | 2.9      | 0.1-14.5  | 1  | 2.9  | 0.1-14.5  |         |  |
| Nasality             |    |          |           |    |      |           |         |  |
| Improvement          | 25 | 71.4     | 54.9-83.7 | 28 | 80   | 64.1-90   | 0.31997 |  |
| Stability            | 10 | 28.6     | 16.3-45.1 | 7  | 20   | 10-35.9   |         |  |
| Aggravation          | 0  | 0        | 0-9.9     | 0  | 0    | 0-9.9     |         |  |
| Nasal air emission   |    |          |           |    |      |           |         |  |
| Improvement          | 27 | 77.1     | 61-87.9   | 29 | 82.9 | 67.3-91.9 | 0.11484 |  |
| Stability            | 7  | 20.0     | 10-35.9   | 6  | 17.1 | 8.1-32.7  |         |  |
| Aggravation          | 1  | 2.9      | 0.1-14.5  | 0  | 0    | 0-9.9     |         |  |



|                                                         | PFI ± PL |   |       | VPP |   |       |         |
|---------------------------------------------------------|----------|---|-------|-----|---|-------|---------|
|                                                         | n        | % | CI95% | n   | % | CI95% | p-value |
| <b>Postoperative nights</b>                             | 2        |   | 1-2   | 2   |   | 2-2   | 2e-05   |
| <b>Resumption of oral intake (postoperative nights)</b> | 1        |   | 0-1   | 1   |   | 1-2   | 1e-05   |

*CI95%: confidence interval 95%; IVV: intravelar veloplasty; n: number of patients; PFI: pharyngeal fat injection; PL: palatal lengthening; VPP: velopharyngoplasty.*

### Cases of moderate to severe VPI

We considered the VPI with preoperative Borel-Maisonny scores of II<sub>m</sub>, III<sub>b</sub> and III<sub>m</sub> as moderate to severe in order to study in a descriptive way their postoperative evolution in both groups (Table 8). This involved 18 patients in the PFI ± PL group and 19 in the VPP group. We did not perform a statistical test on this data because of the small number of patients.

In this subgroup, the Borel-Maisonny score was improved in 61% and stable in 39% of cases in the PFI ± PL group. It was improved in 68.4%, stable in 26.3% and worsened in 5.3% (1 patient) cases in the VPP group.

The intelligibility was improved in 55.6% and stable in 44.4% of cases in the PFI ± PL group and improved in 52.6%, stable in 42.1% and worsened in 5.3% (1 patient, previously described) cases in the VPP group.

Nasality was improved in 71.4% and stable in 28.6% of cases in the PFI ± PL group and improved in 80% and stable in 20% of cases in the VPP group.

Nasal air emission was improved in 61.1%, stable in 33.3% and worsened in 5.6% (1 patient) cases in the PFI ± PL group. It was improved in 84% and stable in 16% of cases in the VPP group.

Table 8. Comparison of the results between the two groups for moderate to severe VPI

|                             | PFI ± PL |      |           | VPP |      |           |
|-----------------------------|----------|------|-----------|-----|------|-----------|
|                             | n        | %    | CI95%     | n   | %    | CI95%     |
| <b>Borel-Maisonny score</b> |          |      |           |     |      |           |
| Improvement                 | 11       | 61.1 | 38.6-79.7 | 13  | 68.4 | 46-84.6   |
| Stability                   | 7        | 38.9 | 20.3-61.4 | 5   | 26.3 | 11.8-48.8 |
| Aggravation                 | 0        | 0    | 0-9.9     | 1   | 5.3  | 0.3-24.6  |
| <b>Intelligibility</b>      |          |      |           |     |      |           |
| Improvement                 | 10       | 55.6 | 33.7-75.4 | 10  | 52.6 | 31.7-72.7 |
| Stability                   | 8        | 44.4 | 24.6-66.3 | 8   | 42.1 | 23.1-63.7 |
| Aggravation                 | 0        | 0    | 0-9.9     | 1   | 5.3  | 0.3-24.6  |
| <b>Nasality</b>             |          |      |           |     |      |           |
| Improvement                 | 10       | 55.6 | 33.7-75.4 | 16  | 84.2 | 62.4-94.5 |
| Stability                   | 8        | 44.4 | 24.6-66.3 | 3   | 15.8 | 5.5-37.6  |
| Aggravation                 | 0        | 0    | 0-9.9     | 0   | 0    | 0-9.9     |
| <b>Nasal air emission</b>   |          |      |           |     |      |           |
| Improvement                 | 11       | 61.1 | 38.6-79.7 | 16  | 84.2 | 62.4-94.5 |
| Stability                   | 6        | 33.3 | 16.3-56.3 | 3   | 15.8 | 5.5-37.6  |
| Aggravation                 | 1        | 5.6  | 0.3-25.8  | 0   | 0    | 0-9.9     |

CI95%: confidence interval 95%; IVV: intravelar veloplasty; n: number of patients; PFI: pharyngeal fat injection; PL: palatal lengthening; VPP: velopharyngoplasty..

### PFI ± PL group

Forty-nine PFI were performed on 35 patients. The average total volume of fat injected was 6.9 ml (2-12). Fourteen out of 35 patients (40%) required a second PFI, with an average duration of 17.7 months (6-39) between the first and the second PFI. During the first PFI, the injection was exclusively in the posterior pharyngeal wall in 11 cases (31.4%), in the posterior pharyngeal wall and the velum in 23 cases (65.7%), and exclusively in the velum in 1 case due to a perforation of the posterior pharyngeal in the adenoidectomy scar. During the second PFI, the injection was exclusively in the posterior pharyngeal wall in 4 cases (31%) and in the

posterior pharyngeal wall and the velum in 9 cases (79%). In the 32 PFI that were both posterior and intravelar, the mean volume was 5.7 ml in the first site and 1.6 ml in the second site respectively.

Of the 14 patients who received a second PFI, 11 post-operative speech therapy assessments were retrieved. Of these 11 patients, two (18.2%) had improved and nine (81.8%) retained their pre-operative Borel-Maisonny score. The intelligibility was improved in 2 and stable in 9 patients. The patients with improvement in their Borel-Maisonny scores and those with improvement in their intelligibility were not the same. Nasality was improved in 5 (45.4%), stable in 4 (36.4%) and worsened in 2 patients. Nasal air emission was improved in 2, stable in 8 (72.7%) and worsened in 1 (9.1%) cases after the second PFI (Table 9).

*Table 9. Phonatory results after second PFI*

|                             | PFI 2 |      |           |
|-----------------------------|-------|------|-----------|
|                             | n     | %    | CI95%     |
| <b>Borel-Maisonny score</b> |       |      |           |
| Improvement                 | 2     | 18.2 | 5.1-47.7  |
| Stability                   | 9     | 81.8 | 52.3-94.9 |
| Aggravation                 | 0     | 0    | 0-25.9    |
| <b>Intelligibility</b>      |       |      |           |
| Improvement                 | 2     | 18.2 | 5.1-47.7  |
| Stability                   | 9     | 81.8 | 52.3-94.9 |
| Aggravation                 | 0     | 0    | 0-25.9    |
| <b>Nasality</b>             |       |      |           |
| Improvement                 | 5     | 45.4 | 21.3-72.0 |
| Stability                   | 4     | 36.4 | 15.2-64.6 |
| Aggravation                 | 2     | 18.2 | 5.1-47.7  |
| <b>Nasal air emission</b>   |       |      |           |
| Improvement                 | 2     | 18.2 | 5.1-47.7  |
| Stability                   | 8     | 72.7 | 43.4-90.2 |
| Aggravation                 | 1     | 9.1  | 0.5-37.7  |

*CI95%: confidence interval 95%; n: number of patients; PFI: pharyngeal fat injection.*

The results for PFI alone and PFI+PL are summarized in Table 10.

*Table 10. Comparison of the results for PFI alone and PFI + PL*

|                             | PFI |      |           | PFI+PL |      |           |
|-----------------------------|-----|------|-----------|--------|------|-----------|
|                             | n   | %    | CI95%     | n      | %    | CI95%     |
| <b>Borel-Maisonny score</b> |     |      |           |        |      |           |
| Improvement                 | 3   | 37.5 | 13.7-69.4 | 10     | 37   | 21.5-55.8 |
| Stability                   | 5   | 62.5 | 30.6-86.3 | 17     | 63   | 44.2-78.5 |
| Aggravation                 | 0   | 0    | 0-32.4    | 0      | 0    | 0-12.4    |
| <b>Intelligibility</b>      |     |      |           |        |      |           |
| Improvement                 | 3   | 37.5 | 13.7-69.4 | 12     | 44.4 | 27.8-62.7 |
| Stability                   | 5   | 62.5 | 30.6-86.3 | 14     | 51.9 | 34.0-69.2 |
| Aggravation                 | 0   | 0    | 0-32.4    | 1      | 3.7  | 0.2-18.3  |
| <b>Nasality</b>             |     |      |           |        |      |           |
| Improvement                 | 6   | 75   | 40.9-92.8 | 19     | 70.4 | 51.5-84.1 |
| Stability                   | 2   | 25   | 7.1-59.1  | 8      | 29.6 | 15.8-48.5 |
| Aggravation                 | 0   | 0    | 0-32.4    | 0      | 0    | 0-12.4    |
| <b>Nasal air emission</b>   |     |      |           |        |      |           |
| Improvement                 | 6   | 75   | 40.9-92.8 | 21     | 77.8 | 59.2-89.4 |
| Stability                   | 2   | 25   | 7.1-59.1  | 5      | 18.5 | 8.2-36.7  |
| Aggravation                 | 0   | 0    | 0-32.4    | 1      | 3.7  | 0.2-18.3  |

*CI95%: confidence interval 95%; n: number of patients; PFI: pharyngeal fat injection; PL: palatal lengthening.*

## DISCUSSION

From the orthophonic point of view, PFI associated or not with PL and VPP with an inferiorly based pharyngeal flap gave comparable phonatory results in the treatment of the VPI: Borel-Maisonny score, intelligibility, nasality and nasal air emission were improved in both groups with no significant difference between them. In the case of moderate to severe VPI, the present study has shown descriptively a comparable evolution of Borel-Maisonny scores (61.1% in the PFI  $\pm$  PL group vs 68.4% in the VPP group) and intelligibility (55.6% in the PFI  $\pm$  PL group vs 52.6% in the VPP group) in both groups. Postoperative nasal air emission remained greater in the PFI  $\pm$  PL group (61.1% of improvements) compared to the VPP group (84.2%), which was expected mechanically. Nasality also appeared to be improved more frequently in the VPP group (84.2 vs 55.6%). These results confirm the complex relationship between nasal air emission, nasality and actual intelligibility, which is not linear and includes numerous factors, in particular articulation. This could explain the lack of efficacy of a second PFI in our cases concerning intelligibility and BM score.

Our study also showed that PFI  $\pm$  PL allows a rapid resumption of oral intake and phonation, and therefore shortens the period of hospitalization, and is then implicitly probably less painful than VPP. It could avoid the feeling of "oppression" and "suffocation" often felt after the realization of a posterior pharyngeal flap. The absence of nasopharyngeal tubes after PFI also plays an important role in improving postoperative comfort and faster resumption of oral intake, since feeding is often refused by children when wearing them. In terms of public health, the shortening of hospitalization as well as the decrease in the consumption of high-level analgesics, which is implied, results in a significant decrease in the cost of care.

From a technical point of view, pharyngeal fat injection is simple to perform. Sampling, like injection, has very little surgical risk. Of 65 PFI procedures performed in our department since 2011, we have never had any postoperative complications. Logically, given the mechanics of the procedure, the risk of obstructive syndrome is less important than in posterior pharyngeal flaps, which is evidenced in the literature (63). In addition, the PFI does not traumatize the pharyngeal muscles and does not create a new pharyngeal scar, hindering neither the maxillary growth (64) nor the performance of a subsequent surgery. It can for the same reason be associated with a palatal lengthening.

Bois et al. (65) suggested the PFI as an interesting and effective alternative to the posterior pharyngeal flap in cases of aberrant internal carotid courses, present in 5% of the

general population and highly prevalent in patients with 22q11.2 micro deletion syndrome (66). Le Boulanger and al. also recommended PFI when flap was contraindicated because of aberrant internal carotid arteries (67). This has to be further evaluated regarding the risk of fat embolism during fat injection (53,68,69). Filip and al. realize a systematic preoperative MRI to identify major vessels and an intraoperative ultra-sound Doppler assessment of the posterior pharyngeal wall to detect smaller vessels. They consider it a contraindication to fat injection in the posterior pharyngeal wall as long as the injection is not precisely controlled (70,57).

One of the main unsolved issues in PFI is the volume loss of injected fat. This phenomenon is known but is difficult to quantify and largely unpredictable in the pharynx and the soft palate. Leuchter and al. observed with nasofibroscopy an early resorption of 30 to 50% of the injected fat in the first two postoperative months. The quantity of fat then seemed to remain stable, as the phonatory results (71). We also experienced the same findings. After a transitory and remarkable improvement of speech during the first 2 weeks following the PFI (notably caused by edema and fat graft) patients usually complain from hypernasality reappear, then finally observe a progressive improvement of intelligibility with early intensive speech therapy. However, long-term effects of PFI on the reduction of the velopharyngeal distance measured on MRI have been shown but this reduction seems to not be linked to the improvement of any speech parameters, which would make its quantification clinically irrelevant (55,56). The literature is controversial about the correlation between the reduction of the velopharyngeal distance and/or gap and the improvement of speech parameters (72). Then the question of preventing and limiting it remains. The appropriate indication for the procedure and the surgical technique are important factors (73–76) but how fat grafting works at the cellular level remains to be fully clarified (77).

Several studies have shown that the effectiveness of PFI was limited in time. Eliküçük and al. observed a decrease in the postoperative improvements concerning nasality after 18 to 24 months (78). As our study was limited to the postoperative orthophonic assessment at 6 months, we could not have demonstrated such a development, even if the need for second fat injection in 40% of patients in the PFI ± PL group suggests a possible alteration of the results over time.

When a first PFI is not sufficient to improve the VPI enough, a second PFI can be performed. This path of action is satisfactory considering the low morbidity of the intervention and the possibility of nasality and intelligibility improvement after a second PFI

(79). In our cases, nasality was the main improved speech parameter after a second VPI (45.2% or 5/11 patients). Four of these patients had a cleft.

The efficacy of repeated PFI is a major argument in the logic of PFI use as a first-line treatment in the VPI, in opposition to the pharyngeal flap, which doesn't allow the realization of later surgical pharyngeal procedure in case of lack of efficacy. As the improvement of a second PFI seems to concern mainly clefts patients that have previously received a single fat injection (11), It could be interesting to evaluate the long term quality of life in the in the patients that benefited from one or more injections.

Indeed the phonatory consequences of VPI directly affect the quality of life of patients, which remains the main final parameter to take into account. In the case of patients with clefts, VPI additionally contributes to them frequently finding themselves the sources of teasing, social rejection and suffering from loss of self-confidence. The VELO questionnaire is a tool resulting from the modification of the VIQL questionnaire that enables the evaluation of the quality of life of patients with VPI, as well as their parents' feelings, if any (80,81). It has been shown that the management of mild VPI by PFI significantly improves quality of life according to the VELO questionnaire (11).

It would be interesting to evaluate this effect in cases of moderate and severe VPI, and to compare, as suggested by Feragen et al. (82), the subjective improvement of the quality of life to the objective Borel-Maisonny score. In their study, these authors suggest that the satisfaction of the child with respect to his language would be significantly related only to the measure of nasality by the speech therapist. When analyzing the questionnaires of the patient's parents, their evaluation of the social and psychological difficulties of their child was significantly correlated with the speech intelligibility measurement.

Another unknown is the fate of grafted fat in cases of weight gain: one case of sleep apnea syndrome was reported in 2009 in an 8-year-old child with ulnar-mammary syndrome who received an PFI and experienced a rapid onset of excess weight (63). It was nevertheless the only reported case of sleep apnea after PFI compared to the large description of OSA syndrome after VPP (40–42).

The present study has several biases requiring consideration. First, the number of patients was limited in both groups. VPP patients were selected among all patients treated in the maxillofacial surgery department of Nantes University Hospital between 2012 and 2017 so that they would be comparable to patients in the PFI ± PL group in age, sex, presence of

cleft palate and syndrome. Hence that was not a matching but a selection of "convenience" that could limit the ability to generalize the present results.

Speech and language assessments were performed by a single speech therapist, which may also have led to an evaluation bias. However, this therapist had extensive experience evaluating patients with velopharyngeal insufficiency and cleft palate. Evaluation by the entourage, mainly the parents, was also used and had a great - although subjective - value in determining the score of intelligibility. Ideally, phonation should be blindly assessed by several speech therapists using voice - and video - recordings of patients as recommended by Sommerlad et al.

In the same way, the small number of patients in this study did not allow us to study the results according to the presence of a syndrome or not, but it has been shown in the literature that the prognosis was poorer in syndromic patients presenting VPI and that the path of care might have to be adapted .

A longer-term prospective study is now needed to determine the place of PFI alone or associated with PL in the treatment of moderate to severe VPI and its potential to increase the quality of life for the patients concerned.



## CONCLUSION

The present study showed that PFI, with or without PL, allowed speech improvement comparable to those of VPP with an inferiorly based pharyngeal flap, including in the treatment of mild to severe VPI. It also showed that this procedure was less invasive and allowed a faster resumption of oral intake as well as reduction of hospital stay, due to less postoperative pain and discomfort.

## DISCUSSION

Du point de vue orthophonique, le lipomodelage pharyngé associé ou non à un allongement vélaire et l'UVPP avec lambeau pharyngé postérieur à pédicule inférieur donnaient des résultats phonatoires comparables dans le traitement de l'IVP : score de Borel-Maisonny, intelligibilité, nasonnement et déperdition nasale étaient améliorés dans les deux groupes sans différence significative entre ceux-ci.

Notre étude a également montré que dans le sous-groupe des patients atteints d'IVP modérée à sévère (phonations II<sub>m</sub>, III<sub>b</sub> et III<sub>m</sub> selon le score de Borel-Maisonny), l'amélioration du score de Borel-Maisonny (61.1% d'améliorations dans le groupe PFI ± PL et 68,4% dans le groupe VPP) et de l'intelligibilité (55.6% dans le groupe PFI ± PL vs 52.6% dans le groupe VPP) était comparable dans les deux groupes en post-opératoire. La déperdition nasale était améliorée de façon plus importante dans le groupe VPP (84,2%) par rapport au groupe PFI ± PL (61,1%), ce qui était attendu sur le plan mécanique. Le nasonnement semblait également amélioré d'avantages dans le groupe VPP (84.2 vs 55.6%). Ces résultats montrent bien la relation complexe qu'entretiennent déperdition nasale, nasonnement et intelligibilité. Celle-ci n'est pas linéaire et prend en compte de nombreux facteurs, en particulier l'articulation. Cette notion pourrait expliquer, au moins en partie, la faible efficacité des secondes injections de graisse dans notre série en termes d'amélioration du score de Borel-Maisonny et de l'intelligibilité.

Notre étude montre également que le LP ± allongement vélaire est un geste moins invasif permettant la réduction du délai de reprise alimentaire post-opératoire, traduisant une douleur post-opératoire moins importante, ainsi que celle de la durée d'hospitalisation. Cette technique semble éviter le sentiment d'oppression et de suffocation parfois ressenti après UVPP avec lambeau pharyngé postérieur, et majoré par la présence de sondes nasopharyngées en post-opératoire immédiat. Leur absence après LP joue un rôle important dans la diminution de l'inconfort post-opératoire et la reprise plus rapide de l'alimentation orale, souvent refusée par les enfants le temps de leur port.

En termes de santé publique, le raccourcissement de l'hospitalisation, donc la diminution de la consommation d'antalgiques de palier élevé - que nous n'avons pas pu démontrée ici de façon chiffrée en raison de nombreuses données manquantes, notamment liées au jeune âge de certains patients - se traduit par une diminution significative du coût de prise en charge. Cette durée d'hospitalisation pourra probablement être réduite d'avantages à

l'avenir, avec la mise en place d'un protocole de surveillance et d'éducation des patients et des parents permettant idéalement la pratique ambulatoire du lipomodelage pharyngé postérieur, même lorsqu'associé à une palatoplastie d'allongement vélaire.

Sur le plan technique, l'injection pharyngée de graisse est simple à réaliser. Le prélèvement, comme l'injection, présentent très peu de risques chirurgicaux. Parmi 65 LP±AV réalisés dans le service depuis 2012, nous n'avons jamais rencontré de complication post-opératoire infectieuse, hémorragique ou obstructive. Le risque de syndrome obstructif des VAS est dans la littérature moins important que dans les lambeaux pharyngés postérieurs, ce qui s'envisage facilement sur le plan mécanique. De plus, le lipomodelage ne traumatise pas les muscles vélopharyngés et ne crée pas de nouvelle cicatrice pharyngée, n'entravant ni la croissance maxillaire ni la réalisation d'une chirurgie vélaire, palatine et/ou pharyngée ultérieure. Il peut pour la même raison être associée à une palatoplastie d'allongement vélaire.

Bois et al. (65) ont suggéré que le LP était une alternative intéressante au lambeau pharyngé postérieur chez les patients présentant un trajet aberrant des carotides internes le contre-indiquant. La prévalence de ces trajets carotidiens aberrants est de 5% dans la population générale mais beaucoup plus importante en cas de syndrome de délétion 22q11 (66). Leboulanger and al. ont également réalisé un LP en cas de trajet carotidien aberrant (67). Cette indication doit être envisagée prudemment, le seul réel grave risque du LP restant l'embolie graisseuse durant l'injection (53,68,69). Filip and al. (56,57) considèrent quant à eux toute déviation des vaisseaux comme une contre-indication au LP tant que le site d'injection ne sera pas précisément guidé. Ils recourent à une IRM pré-opératoire systématique afin d'identifier les principaux vaisseaux puis à une échographie-Doppler per-opératoire de la paroi pharyngée postérieure afin de détecter les vaisseaux secondaires.

L'une des principales questions non résolues de l'injection de graisse autologue est celle de la résorption de la graisse injectée (75,76,83). Ce phénomène, connu et décrit dans d'autres applications de la greffe de graisse autologue, est difficilement quantifiable et largement imprévisible dans le pharynx et le voile du palais. Leuchter et al. (71) ont observé par nasofibroscopie une résorption précoce de 30 à 50% de la quantité de graisse injectée à deux mois post-opératoires. Cette quantité semblait se stabiliser après cette phase de résorption rapide, ce qui semblait confirmer par des résultats phonatoires durables. Notre expérience est similaire : après une amélioration extrêmement rapide, si ce n'est immédiate,

de la phonation dans les deux premières semaines postopératoires, les patients remarquent souvent une réapparition partielle de leur nasonnement, qui s'améliore finalement progressivement avec la reprise précoce d'une rééducation orthophonique intensive. Puis ces résultats acquis après rééducation semblent s'installer dans le temps, malgré la probable résorption quantitative de la graisse injectée. L'amélioration transitoire spectaculaire initiale est probablement, en plus de la graisse injectée, créée par l'œdème post-opératoire, et s'accompagne souvent de ronflements nocturnes, eux-aussi transitoires.

L'efficacité du LP sur le long terme sur la réduction de la distance vélopharyngée mesurée par IRM a été montrée, mais cette réduction ne semble être significativement liée à l'amélioration d'aucun paramètre phonatoire majeur, rendant sa quantification cliniquement non pertinente (55,56). La corrélation entre réduction de la distance vélopharyngée et/ou de la surface de l'orifice vélopharyngé et l'amélioration de la phonation est un objet de controverse dans la littérature (72). Ainsi, si la nécessité de la quantifier ne semble pas évidente, restent la question de la prévenir et de la limiter afin d'optimiser théoriquement les résultats phonatoires. Pour l'instant, une indication opératoire adaptée et la technique de prélèvement et d'injection, aujourd'hui bien codifiée, semblent les principaux facteurs de réussite (73–76) mais le mécanisme d'action de la greffe de graisse autologue à l'échelle cellulaire reste à être totalement élucidé (77).

Outre cette phase de résorption précoce, il a été montré que l'efficacité du lipomodelage pharyngé était limitée dans le temps sur le long terme. Eliküçük and al. (78) ont observé chez leurs patients la réapparition d'un nasonnement après 18 à 24 mois après LP. Notre étude se limitant au bilan orthophonique post-opératoire à 6 mois, nous n'avons pas pu mettre en évidence une telle évolution, même si la nécessité de deuxième lipomodelage chez 40% des patients du groupe LP laisse deviner l'altérabilité des résultats dans le temps.

Lorsque le premier LP n'est pas suffisant pour corriger l'IVP de façon satisfaisante, une deuxième injection peut être réalisée (40% des cas dans notre étude). Cette conduite thérapeutique est satisfaisante compte-tenu de la faible morbidité de l'intervention et du taux de réussite de cette deuxième injection (79). Dans notre étude, le seul paramètre nettement amélioré après seconde injection était le nasonnement (45.2% or 5/11 patients). Quatre des cinq patients concernés par cette amélioration étaient porteurs d'une fente vélaire.

La possibilité de répéter les injections de manière efficace est un argument majeur dans la logique de proposer le LP comme traitement de première intention dans la prise en charge de l'IVP quel que soit sa sévérité initiale : contrairement à la réalisation d'une VPP avec lambeau pharyngé postérieur, le LP permet la réalisation ultérieure, en cas d'efficacité

insuffisante, d'une seconde voire d'une troisième injection, et/ou d'une VPP. L'évaluation de l'efficacité des injections répétées devra être réalisée de manière prospective sur un plus grand nombre de patients. Il sera intéressant, dans le même temps, d'évaluer et de comparer l'évolution de la qualité de vie sur le long terme chez les patients ayant bénéficié d'une ou de plusieurs injections et leur perception de la répétition des interventions, l'amélioration de celle-ci semblant concerner principalement les patients atteints de fentes et ayant bénéficié d'une unique injection de graisse (11).

Les conséquences phonatoires de l'IVP altèrent directement la qualité de vie des patients atteints, paramètre final majeur à considérer. Dans le cas des patients atteints de fente, elles participent à un ensemble de séquelles fréquemment sources de moqueries, de rejet social et de perte de confiance en soi. Le questionnaire VELO est un outil résultant de la modification du questionnaire VIQL permettant l'évaluation de la qualité de vie des patients atteints de VPI, mais aussi du ressenti des parents le cas échéant (80,81). Il a déjà été montré que la prise en charge d'IVP légères par LP permettait d'améliorer la qualité de vie des patients de manière significative selon le questionnaire QOL (11). Il serait intéressant d'évaluer cet effet en cas d'IVP modérée à sévère, et de comparer, comme le suggèrent les auteurs, l'amélioration subjective de la qualité de vie à celle, plus objective, du score de Borel-Maisonny. Cependant, l'étude de Feragen et al. (82) semble suggérer que la satisfaction de l'enfant par rapport à son langage ne serait significativement liée qu'à la mesure du nasonnement par l'orthophoniste. Quant à l'estimation des difficultés sociales et psychologiques de leur enfant par les parents, elle était significativement corrélée à la mesure orthophonique de l'intelligibilité.

Il faut noter qu'une autre inconnue est le devenir de la graisse greffée en cas de prise de poids secondaire : un cas de SAOS a été rapporté en 2009 chez un enfant de 8 ans ayant bénéficié d'un LP pour le traitement d'une IVP décompensée après adénoïdo-amygdalectomie dans un contexte de syndrome ulnaire-mammaire après apparition rapide d'un surpoids (63). Cette complication n'a été décrite qu'une fois après LP, contrairement aux multiples cas de SAOS publiés après UVPP (40–42).

Notre étude comporte plusieurs biais nécessitant considération. Tout d'abord, le nombre de patients était restreint. De plus, nous avons sélectionné les patients du groupe UVPP parmi tous les patients traités par UVPP avec lambeau pharyngé postérieur pour IVP dans le service de chirurgie maxillo-faciale du CHU de Nantes entre 2012 et 2017 afin qu'ils

soient comparables aux patients du groupe LP sur l'âge et le sexe, réalisant alors non pas un appariement mais une sélection « de commodité » pouvant limiter la généralisabilité.

Ensuite, les bilans orthophoniques ont été réalisés par une seule orthophoniste, ce qui a également pu entraîner un biais d'évaluation. Cependant, cette orthophoniste avait une longue expérience dans l'évaluation spécialisée de l'IVP, en particulier chez les patients porteurs de fente. De plus, l'inclusion dans l'évaluation phonatoire de l'estimation de l'intelligibilité du patient par son entourage présentait une plus-value précieuse. Toutefois, l'évaluation phonatoire devrait idéalement être réalisée par plusieurs orthophonistes et en aveugle, basée sur des enregistrements audio et vidéo des patients, comme recommandé par Filip et al. (57).

De la même façon, le petit nombre de patients ne nous a pas permis d'étudier les résultats en fonction de la présence d'un syndrome ou non, mais il a été montré dans la littérature que le pronostic des IVP chez les patients syndromiques présentant une atteinte neurophysiologique responsable d'une incompétence vélaire plus ou moins évidente était plus sombre et pouvaient nécessiter un parcours de soins personnalisé (84,85).

À présent, une étude prospective sur le long terme incluant un nombre de patients plus importants est nécessaire afin de confirmer la place à donner au lipomodelage pharyngé postérieur, associé à un allongement vélaire si nécessaire, dans le traitement des IVP modérées à sévères et d'évaluer l'amélioration de la qualité de vie des patients concernés.

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**RÉSUMÉ : Lipomodelage pharyngé dans l'insuffisance vélopharyngée : un traitement de première intention ? Comparaison à l'uvulovélopharyngoplastie avec lambeau pharyngé postérieur à pédicule inférieur.**  
**Étude rétrospective de 70 cas.**

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**Introduction :** L'insuffisance vélopharyngée (IVP) affecte la communication des patients concernés et leur qualité de vie. Sa principale cause est la présence d'une fente vélaire, syndromique ou non. Lorsque la rééducation orthophonique ne suffit pas à corriger les conséquences phonatoires de cette IVP, plusieurs techniques chirurgicales sont disponibles pour tenter d'améliorer l'intelligibilité : véloplasties d'allongement, uvulovélopharyngoplasties (UVPP) et plus récemment, lipomodelage pharyngé (LP). Aujourd'hui, l'UVPP est le gold standard dans le traitement de l'IVP modérée à sévère, tandis que palatoplastie et LP sont réservés aux IVP légères. L'UVPP est une procédure efficace mais invasive induisant des douleurs post-opératoires importantes ainsi qu'un risque de troubles de la croissance maxillaire et de syndrome d'apnées obstructives du sommeil (SAOS). Le LP associé ou non à un allongement vélaire est peu invasif et bien toléré, mais son efficacité dans les IVP modérées à sévères n'a pas été comparée à celle de l'UVPP. L'objectif principal de cette étude était de comparer les résultats phonatoires du LP associé ou non à un allongement vélaire, et de l'UVPP avec lambeau pharyngé à pédicule inférieur.

**Matériel et méthodes :** Cette étude rétrospective a porté sur soixante-dix patients présentant une IVP : trente-cinq patients ont été traités par UVPP avec lambeau pharyngé à pédicule inférieur et 35 patients par lipomodelage pharyngé postérieur plus ou moins associé à un allongement vélaire. Tous les patients ont été évalués en pré-opératoire et à 6 mois post-opératoires par un orthophoniste spécialisé, selon le score de Borel-Maisonny, l'intelligibilité, le nasonnement et la déperdition nasale.

**Résultats :** Les patients des deux groupes étaient comparables selon l'âge à la prise en charge, le sexe, la présence d'un syndrome et d'une fente et les scores de Borel-Maisonny pré-opératoires. Il n'y eu aucune complication post-opératoire. Le score de Borel-Maisonny et tous les principaux paramètres phonatoires étaient améliorés dans les deux groupes sans différence significative entre ces derniers. Le délai de reprise alimentaire et la durée d'hospitalisation étaient significativement réduits dans le groupe LP par rapport au groupe UVPP.

**Conclusion :** le LP associé ou non à un allongement vélaire permettait d'obtenir des résultats phonatoires comparables à ceux de l'UVPP avec lambeau pharyngé à pédicule inférieur dans le traitement de l'IVP. Le LP était responsable d'une morbidité post-opératoire moins importante se traduisant par la réduction significative du délai de reprise alimentaire et de la durée d'hospitalisation. Une étude prospective est nécessaire pour confirmer ces résultats et définir la place du LP comme traitement de première intention de l'IVP modérée à sévère.

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**MOTS-CLÉS :** insuffisance vélopharyngée ; lipomodelage pharyngé ; uvulovélopharyngoplastie ; score de Borel-Maisonny ; nasonnement.