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Original article

Retropharyngeal lipostructure in the treatment of velopharyngeal insufficiency: A prospective study and update

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ABSTRACT

Introduction: Retropharyngeal lipostructure is a recent procedure in velopharyngeal insufficiency (VPI), offering an effective alternative to heavier surgery.
Objectives: To update and assess retropharyngeal lipostructure as a treatment for VPI in the University Hospital Center of Rouen (France).
Type of study: Single-center prospective study, from May 2012 to May 2014.
Patients and methods: Six patients (4 girls, 2 boys) presenting with VPI were treated by retropharyngeal lipostructure. Age at surgery ranged between 6 and 12 years. Four of the patients bore a 22q11 microdeletion. Treatment was indicated in case of Borel-Maisonny type 2b ($n=2$) or 2 m ($n=4$) despite well-conducted speech therapy and of $\geq 50\%$ velopharyngeal sphincter closure on nasal endoscopy. Patients were assessed preoperatively and at 3 months, by a multidisciplinary team. Borel-Maisonny type was assessed by a speech therapist. Nasality was measured on assisted vocal evaluation (EVA®). Sphincter closure was assessed on dynamic MRI.
Results: Between 6 and 8 cm^3 autologous fat was injected. At 3 months, 4 children showed 1-grade improvement in Borel-Maisonny type. Nasality decreased systematically, from a mean 14.5% preoperatively to 10.5% postoperatively. MRI showed improvement in all cases, with complete closure in occlusive vowels in 3 children.
Conclusion: EVA® and MRI provide precise objective assessment of VPI. Retropharyngeal lipostructure is a simple, relatively non-invasive, reproducible technique, providing good results in VPI.

1. Introduction

Velopharyngeal insufficiency (VPI) is a complex pathology related to the anatomy and physiology of the velopharyngeal sphincter (VPS). It appears when contraction of the sphincter, comprising soft palate and lateral and posterior pharyngeal walls, does not achieve complete closure of the nasopharyngeal space, resulting in nasal air leakage on certain phonemes, notably with oral consonants [p,t,k,b,d] and oral vowels [a,e,o,u,i]. The phoniatric impact of velar insufficiency, or open rhinolalia, concerns breathing, certain phonemes, articulation, and hence intelligibility.

Anatomic causes are implicated in more than 50% of cases of VPI: velopalatine or submucosal cleft or short palate. Incidence is 1 live birth per 2000. VPI occurs in 10–40% of patients operated on for velar or velopalatine cleft. It is isolated in 50–70% of cases and syndromic in 30% [1].

VPI, whether or not related to velopalatine cleft sequelae, is found in many syndromes, of which 22q11 microdeletion is notably frequent, where VPI is found in 32–92% of cases [2]; 12.5–30% of cases of VPI involve 22q11 microdeletion [3].

Iatrogenic etiologies are frequent: adenoidectomy, uvulopharyngopalatoplasty or tonsillectomy, notably in case of overlooked submucosal cleft or short palate.

Following etiological and functional assessment, treatment systematically comprises early speech therapy, with surgery as necessary.

Surgery uses several possible techniques, and particularly passive or active pharyngoplasty or velopharyngoplasty.

In severe VPI with $<50\%$ sphincter closure on phonation and absence of soft palate contraction, velopharyngoplasty or active pharyngoplasty is indicated, with proven efficacy in selected patients but a significant risk of postoperative complications such as bleeding, respiratory disorder, obstructive sleep apnea

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syndrome, tubal dysfunction and pain, requiring a few days' hospital admission.

In mild to moderate VPI, with > 50% closure and contractile soft palate, passive pharyngoplasty by autologous fat injection in the posterior pharyngeal wall offers a good alternative, and can always be followed by heavier surgery if results are inadequate. It has the advantage of being relatively non-invasive, with simple postoperative course, and can be iterated if need be.

Passive pharyngoplasty by injection or implantation in the posterior pharyngeal wall was first described in the early 20th century: in 1900, Gersuny [4] reported Vaseline injection; subsequently, other materials were used: teflon [5], calcium hydroxyapatite [6], cartilage [7], and collagen [8]. These were abandoned, due to complications such as implant extrusion, migration or infection or loss of efficacy due to resorption or atrophy.

In 1926, Von Gaza described implantation of autologous fat in the posterior pharyngeal wall via a cervicotomy. Only in the 1990s, however, with the work of Sydney Coleman in plastic surgery [9], did autologous fat graft become widespread, thanks to the lipostructure technique. In 2001, Dejonckere and van Wijngaarden [10] used this minimally invasive technique of autologous fat injection in the posterior pharyngeal wall via a transoral approach.

Injection concerns only the posterior pharyngeal wall, as extra sites, such as the posterior pillars or soft palate, would increase the risk of postoperative complications. Moreover, according to Croft et al. [11], physiological VPS closure is predominantly coronal, in 55% of subjects.

The aim of the present study was to confirm the efficacy of lipostructure and update our management of VPI in terms of treatment and pre- and postoperative assessment.

2. Assessment

Pre- and 3-month post-operative assessment comprised clinical ENT examination, nasal endoscopy, voice recording, speech therapy examination with assisted vocal evaluation (EVA®), and MRI. A clinical check-up was performed in consultation at 1 month.

The clinical examination comprised resting assessment of velar symmetry, tonsillar pillars, VPS proportions, vegetation volume, and phonatory analysis of an occlusive vowel and of velar course, pharyngeal wall contraction, VPS closure and presence of Passavant's ridge. Nasal endoscopy, under local anesthesia using a cotton bud soaked in naphazoline xylocaine in either nostril for 5 minutes before examination, screened for submucosal cleft by transillumination of the soft palate and quantified vegetation; during phonation, it analyzed uvular muscle contraction, Passavant's ridge size and degree and type of VPS closure. After examination, an assessment form was filled out.

Speech therapy examination provided perceptual analysis of the voice: nasality, intelligibility, and compensatory mechanisms in pronouncing various phonemes, in semi-directed speech and free conversation. Nasal air loss was quantified using a Glatzel mirror. After examination, a phonation grade was attributed following Borel-Maisonny [12] (Table 1).

EVA® is a device developed by the Parole et Langage laboratory (URA CNRS 261, 1 Rue Guy Fabre, 13001 Marseille, France), described by Giovanni et al. [13]; it provides an objective quantitative nasality score, calculated automatically by the computer during pronunciation of a sustained "a" vowel and three standardized sentences of increasing nasality: "*Le Pape salue la foule*", "*La nappe touche le parquet*", and "*Lance ton ballon dans les airs*". The device comprises a computer connected up to three sensors: a microphone to record sounds, an air-flow detector (mask) in front of the moth, and an air-flow detector (two independent probes) in front of the nose. The system records and analyzes acoustic, aerodynamic and electrophysiological phenomena associated with speech production during in-breath and out-breath.

MRI provides objective quantitative analysis of VPS closure [14], using the SSFSE sequence, with sagittal slices centered on the mid-line and axial slices centered on the palatine bone, in neutral position then in occlusive vowel ("a", "o") phonation sustained for 10 s. As well as analyzing VPS anatomy, it measures the distance (mm) from the posterior part of the palate to the anterior part of the pharyngeal wall, and the open area (mm^2) behind the soft palate at rest and in phonation. Coronal slices measure laryngeal width and assess lateral pharyngeal wall involvement in VPS closure.

MRI also screens for vascular deformities liable to contraindicate velopharyngeal surgery. Abnormal carotid course in medial position is found in 27% of patients bearing the 22q11 deletion [15].

Postoperatively, as well as the preoperative parameters examination screened for postoperative complications: pain, infection, healing defect at the harvesting site and VPS, and nasopharyngeal dysfunction: nasal obstruction, pharyngeal obstruction, pharyngeal discomfort, snoring, obstructive sleep apnea syndrome, and closed rhinolalia.

The patient and family's satisfaction was assessed on a questionnaire.

3. Surgical technique

Static pharyngoplasty by retropharyngeal lipostructure was performed under general anesthesia (orotracheal intubation) by a single surgeon using a single technique.

The patient was positioned supine, with the neck in extension, and the posterior pharyngeal wall was exposed using a Kilner mouth gag.

The first step consisted in harvesting adipose tissue by lipoaspiration following Coleman. A peri-umblical abdominal site is to be preferred to the medial side of the knee, although this provided an alternative in thin patients. After physiological saline injection, a 10 cm^3 sample was taken using a 10 cm^3 Luer Lock syringe and classical aspiration cannula, and centrifuged for 10 minutes at 3000 rpm to extract serum-blood and oily phases so as to maximize healthy adipocyte concentration.

The second step was the injection. Under endoscopic control, using a 21-gauge epicranial needle, 3 injections points were used, submucosally in the posterior pharyngeal wall. The first was above the projection point of the soft palate in the relief of the anterior arch of the atlas or away from any persistent vegetation. Two paramedian injections were then made on a horizontal line through the first point, so as to achieve a homogeneous enlargement of the whole posterior pharyngeal wall (Fig. 1), forming a Passavant neo-ridge [16].

Admission was on an outpatient basis. Postoperative course included step-1 analgesia, progressive resumption of feeding with lukewarm soft foods for the first days, and a consultation on day 8 to remove sutures.

4. Results

Six patients (4 boys, 2 girls; age 6–12 years) were consecutively included from May 2012 to May 2014 in the University Hospital of Rouen, France. All underwent retropharyngeal lipostructure on the same technique, with complete pre- and post-operative assessment. In 4 cases, VPI was part of a 22q11 microdeletion syndrome; in the other 2, VPI was primary, with atonic soft palate and deep nasopharynx. There were no cases of submucosal cleft.

Peri-umbilical fat and fat from the medial side of the knee were used in 3 of the 6 cases each, with a mean 6.8 cm^3 (range, 6–8 cm^3). All patients had same-day discharge. There were no

Table 1
Nasality according to Borel-Maisonny (BM) classification.

Patient	Age (years)	Etiology	Preoperative BM	BM at 3 months
1	9	22q11, adenoidectomy, deep nasopharynx	2m	2m
2	12	22q11, atonic soft palate, deep nasopharynx	2m	2b
3	9	22q11, operated velopalatine cleft, atonic soft palate	2m	2m
4	6	Atonic soft palate	2m	2b
5	8	22q11, atonic soft palate	2b	1/2
6	10	Atonic soft palate, deep nasopharynx	2b	1/2

Borel-Maisonny classification: Type 1: good phonation; Type 1/2: variable nasal air loss; Type 2b: Constant nasal air loss without impaired intelligibility; Type 2m: Constant nasal air loss with impaired intelligibility; Type 3: Compensatory mechanisms: glottal stops and/or hoarse breathing.

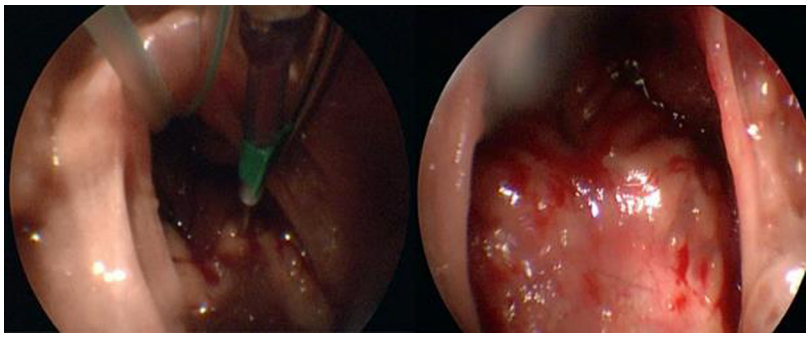

Fig. 1. Left: posterior pharyngeal wall before injection; right: after procedure (image from Rouen University Hospital).

cases of postoperative nasopharyngeal dysfunction. One patient had postoperative neck pain requiring step-2 analgesia, with resolution by the day-8 check-up. Another had contact pain at the harvesting site (medial side of the knee), lasting several weeks. There were no major complications.

Borel-Maisonny type improved by 1-grade at 3 months in 4 patients. Two of the 4 patients with type 2 m showed no improvement. The 2 patients with type 2b recovered satisfactory phonation with occasional nasality (Table 1).

VPS closure on nasal endoscopy improved in 5 of the 6 patients, with complete closure for 2.

All patients showed improvement in nasality, although for 1 patient this was only for the 3 sentences. Mean nasality on sustained "a" phonation was $14.5 \pm 11.6\%$ (range, 5.2–37.5) preoperatively, and $10.5 \pm 6.1\%$ (range, 2.7–20) at 3 months: i.e., decrease of 32%. For phonation of the 3 sentences, the mean values were $50 \pm 18.6\%$ (range, 27.5–70.5) and $35 \pm 18.8\%$ (range, 9.9–60), respectively (Table 2): i.e., decrease of 30%.

Three patients showed complete VPS closure on occlusive vowel phonation on axial and sagittal MRI slices (Table 3 and Fig. 2).

All families were satisfied with the treatment. At 3 months, 2 families reported improved vocal intensity, 1 family reported better breathing, and 1 family reported resolution of oral reparation.

5. Discussion

Static pharyngoplasty by retropharyngeal lipostructure is indicated for mild to moderate VPI or to supplement velopharyngoplasty showing inadequate results. Precise analysis of VPS anatomy and function is essential for making the indication: a contractile soft palate with partial VPS closure is required. Several criteria have been described:

- < 3–4 mm VPS closure [10];
- 0.5–2 cm^2 VPS area [17];
- VP closure < 1.5 cm [18];
- or "relatively small VP gap" [19].

In the present study, as in those by Leuchter et al. [20] and Cantarella et al. [21], > 50% closure appeared to be the most reliable and reproducible criterion.

The main drawback of lipostructure is fat resorption, with rates of 250–40% in facial plastic surgery [22]. Leuchter [20] reported that, in the particular case of retropharyngeal lipostructure, resorption occurred before month 2 in 30–50% of cases. According to Bardot and Dejonckere [10,23], resorption occurs within 3 months.

The present patient no. 3 is a good illustration of this. At the 1-month check-up, he showed intermittent nasality, with restored intelligibility. Family and friends reported better understanding and ease of communication. This clear improvement was not to be found at 3 months: the family described a pattern of gradual deterioration, with recurrence of intelligibility problems at around 2 months. To alleviate the problem of resorption, Leuchter recommended around 30% hypercorrection.

In terms of durability, results in plastic surgery and ENT are encouraging [22]. In laryngology, MRI showed that grafted cells remained alive in the receiver site [24]. In retropharyngeal lipostructure, Dejonckere et van Wijngaarden [10] reported stable results over more than 2 years. Once the graft has taken, cells should stay alive for as long as the receiving tissue does. On the other hand, grafted adipose cells are affected by patient weight variation: in case of overweight at surgery, weight-loss may lead to resorption. Leuchter et al. [20] reported a trend for decreasing nasality several months after lipostructure; this may be because the advanced posterior pharyngeal wall enables the VPS to develop compensatory mechanisms.

A major strong-point of lipostructure is its reproducibility, limited only by the amount of adipose tissue available at the donor site. Resorption often necessitates iterative procedures [16,20,23] to achieve an optimal result. Reintervention may be suggested after 3 months. The fixed fat is revascularized, so gain at each step, however slight, is definitive.

The results in the present series of 6 patients tend to confirm efficacy at 3 months, both objectively and subjectively. Subjective analysis found a 1-grade gain in Borel-Maisonny type in 4 of the 6 patients. Borel-Maisonny type, however, is observer-dependent, and not specific to velar function. Intelligibility can be impaired by

Table 2
Nasality (%) on EVA®.

Patient no.	Preoperative	Postoperative	Preoperative	Postoperative
	Sustained "a"	Sustained "a"	3 sentences	3 sentences
1	7.6	2.7	43.8	26.9
2	14.9	10.9	63.1	46
3	20	5.2	65	60
4	37.2	15	70.5	46.8
5	10.5	7.6	27.5	21.4
6	11.7	7.7	30.9	9.9

Table 3
VPS closure on MRI. Distance between soft palate and anterior pharyngeal wall (mm).

patient n°	VP distance in neutral position		VP distance in phonation		VPS area in neutral position		VPS area in phonation	
	Preop	Postop	Preop	Postop	Preop	Postop	Preop	Postop
1	12	7	7	<1	250	25	76	0
2	11.8	10	6.8	<1	141	60	106	57
3	9.8	11	9	7	164	143	96	75
4	12	10	9	3	148	121	168	136
5	12	7	6	0	250	115	64	0
6	11.1	11	4.7	<1	167	113	50	0

VPS: velopharyngeal sphincter.

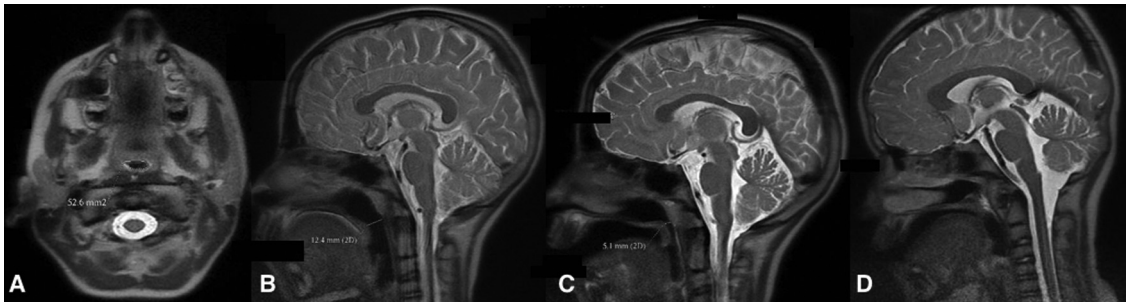

Fig. 2. MRI SSFSE sequence. Preoperative axial and sagittal slices. Velopharyngeal sphincter in neutral position (A and B) and in occlusive vowel phonation (C): soft palate not in contact with posterior pharyngeal wall. Postoperatively (D): soft palate in contact with posterior pharyngeal wall. Hypersignal at contact point, corresponding to adipose tissue graft (image from Rouen University Hospital).

praxis disorder, which is frequent in children with VPI, especially in case of 22q11 microdeletion, probably making it difficult for these children to get from grade 2 m to grade 2b.

Objective analysis found a mean 30% decrease in nasality on EVA® at 3 months. Comparably, Leuchter et al. [20] reported decrease from 36.5% to 23.3%: i.e., 36% decrease; and Lau et al. [17] reported decrease from 43.2% to 29.7%: i.e., 31%. Nasality is a simple, reliable and reproducible parameter, and should be assessed routinely in managing VPI, enabling objective classification. There are, however, no normal reference values or classification of VPI according to nasality.

MRI provides useful data for pre- and post-operative assessment. It is non-invasive and radiation-free, and was performed without problems of compliance or need for sedation in all 6 children, aged between 6 and 12 years. Like nasal endoscopy, it explores VPS anatomy and function, quantifying closure. Exploration was restricted to 2 planes, so as not to prolong examination. No abnormal carotid course was associated with 22q11 microdeletion. Hypersignal behind the posterior pharyngeal wall demonstrated adipocyte survival at 3 months. VPS closure improved in all cases, with complete closure on phonation in 3. MRI and nasal endoscopy results for preoperative and 3-month closure agreed, but MRI measurement were more precise, and assessed vertical closure. Both examinations require compliance, making them difficult to perform in under-5 year-olds. Interestingly, MRI and EVA® findings were concordant. In 2011, Filip et al. [19] reported MRI findings in 12 patients with multiple injection (soft palate, posterior pillars and posterior pharyngeal wall), and found significantly reduced VP distance on sagittal slices during phonation, and no significant difference in VPS area.

The present study confirms previous reports on the advantages of posterior pharyngeal wall lipostructure [10,17,20], with only 2 cases of mild complications, such as spontaneously resolving pain. All patients were able to be treated on an outpatient basis, which is not recommended for dynamic pharyngoplasty. Only 1 major complication has been reported elsewhere: onset of severe sleep apnea syndrome, after 2 injections which were effective against rhinolalia in an 8-year-old boy [25]. This rare complication should be systematically screened for during the first postoperative nights, by observation of respiratory breaks, of which the parents need to be informed in the preoperative consultations. In case of the slightest doubt, the child should be admitted for polysomnography (there have been no such cases in our department).

Long-term clinical follow-up was possible in only 4 patients; patients 1 and 6 were lost to follow-up. Patient 5 received a second lipostructure at 2 years, with stable improvement and a Borel-Maisonny type 1/2. In patient 2, initial benefit was maintained. In patients 3 and 4, second injection provided only transient benefit, and secondary velopharyngoplasty was required.

6. Conclusion

Retropharyngeal lipostructure is a feasible first-line attitude in mild to moderate VPI, being much less aggressive than surgery

such as velopharyngoplasty. Second injection at 3 months may be necessary in case of insufficient results. In some cases, however, velopharyngoplasty is the only solution for iterative recurrence.

EVA® and MRI provide precise objective functional and anatomic analysis of results.

Disclosure of interest

The authors declare that they have no competing interest.

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