Prospective Evaluation of the Outcome of Velopharyngeal Insufficiency Therapy after Simultaneous Double Z-Plasty and Sphincter Pharyngoplasty

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Key Words

Cleft palate · Velopharyngeal insufficiency · Furlow operation · Orticochea operation

Abstract

Objective: One of the main goals in the management of cleft palate is to achieve a good quality of speech. The aim of the prospective study was to evaluate the effectiveness of therapy in patients with velopharyngeal insufficiency treated by simultaneously performed Furlow and Orticochea operations. **Patients and Methods:** From May 2007 to May 2008 we treated 14 consecutive patients (6 males and 8 females, mean age 14 years). The indications for surgery were based on nasofiberscopic examination, evaluation of speech quality, nasometry and morphology of the palate. The velopharyngeal closure was below 80% in all the patients; they had pronounced nasality and limited intelligibility of speech. All the palates were short. **Results:** The final outcome of treatment was based on the combined evaluation of 4 parameters: closure, speech intelligibility, nasality and the nasalance index. Ten patients achieved full recovery (71%), the remaining 4 had improved recovery (29%). **Conclusion:** A 1-stage Furlow operation and sphincter pharyngoplasty are an effective modality in the therapy of velopharyngeal insufficiency. Indications for posterior pharyngeal flap pharyngo-

plasty should be limited to the cases in which a simultaneous Furlow operation and sphincter pharyngoplasty are not possible due to a deficit of the palatine tissue.

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Introduction

One of the main goals in the management of a cleft palate is to achieve a quality of speech that enables normal functioning in society. It is achievable in the majority of patients; however, about 10–40% of cleft patients complain of unsatisfactory speech quality [1]. In Poland, where the incidence of this defect is about 1.7 in 1,000 live births, 600–800 children are born annually with clefts [2]. This means that, even with the most optimistic assumptions, at least 100 patients a year require therapy for velopharyngeal insufficiency.

The most typical feature of speech in cleft patients, so-called open nasality, results from changes in nasal resonance. It may be due to the presence of fistulae and residual openings in the palate, immobility of the pharyngeal walls or anatomical disproportions referred to as ‘deep pharynx’. Most commonly, however, it is the effect of velopharyngeal sphincter insufficiency resulting mainly from insufficiency of the palate, i.e. the palate contains

scars, is too short and usually poorly mobile. Poor mobility of the palate may also result from abnormal, oblique placement of the muscles which were not anastomosed in their anatomical position during the primary cleft operation. Sometimes it may happen that palatal function remains undisturbed, or slightly weaker, and velopharyngeal insufficiency results from decreased mobility of the pharyngeal walls.

The development of velopharyngeal insufficiency in patients with clefts may be affected by numerous factors. According to various authors, a significant role is attributed to the kind and severity of cleft, choice of surgical modality [3–6], timing and sequencing of the therapy [7–9], and the experience of the surgeon and quality of the operation [10–15].

The severity of velopharyngeal insufficiency and the therapeutic possibilities also depend on hearing acuity, muscle performance, nerve supply, and – secondarily – on the development of compensatory mechanisms.

No specific criteria determining strict indications for the operation have been proposed yet, so the age at which the operation should be performed cannot be determined explicitly. The majority of specialists believe that the corrective procedure should not be delayed too much after speech therapy [16]; therefore, it should be performed at school age, or even pre-school age, if possible [17–19].

In the last 100 years, numerous surgical modalities used in the treatment of velopharyngeal insufficiency were described: pharyngoplasty with the use of a flap harvested from the posterior pharyngeal wall [20, 21], ‘push-back’ operations elongating the palate and moving it backwards [22], palatal sphincter pharyngoplasty [23, 24], operations augmenting the posterior pharyngeal wall by means of implants and tissue grafts [25, 26], and a double Z-plasty Furlow operation.

As shown in the literature and our own prospective studies [27, 28], the above methods are not 100% effective. For this reason, despite achieving significant improvement in 90% of the patients after pharyngeal flap surgery with the use of a flap from the posterior pharyngeal wall and Furlow and Orticochea operations, we decided to combine the 2 latter methods in 4 patients who had an unsatisfactory outcome. And thus, the Orticochea operation was followed by the Furlow method, and vice versa, in 4 patients, resulting in a very good outcome. Taking this excellent outcome into consideration, we decided to report on the altered management in this publication. It was believed that simultaneous Orticochea and Furlow operations should improve the general outcome for pa-

tients who had indications for lengthening the palate to improve its effectiveness.

The aim of the prospective study was to evaluate the effectiveness of therapy in patients with velopharyngeal insufficiency treated by simultaneously performed Orticochea and Furlow operations.

Material and Methods

From May 2007 to May 2008 there were 14 consecutive patients with velopharyngeal insufficiency who qualified for simultaneous Orticochea and Furlow operations (P.W.). The indications for surgery were based on nasofiberscopic examination, evaluation of speech quality, nasometry and morphology of the palate. The velopharyngeal closure was below 80% in all patients (below 50% in the majority of patients), and they had pronounced nasality and limited intelligibility of speech. All patients revealed an increased nasometric index which exceeded the norm (30%). All palates were short. The levator muscle fibers were positioned in a more or less oblique pattern. The study group included 6 males and 8 females aged 4–27 years (mean 14). Seven patients had bilateral clefts, 3 had a unilateral cleft, 2 had an isolated cleft of the palate and 2 had a submucosal cleft.

All patients were consulted by a speech therapist and a plastic surgeon 3–6 months after the operation (before 3 months for 2 patients). In order to compare the initial condition with the outcome of the surgical management, pictures of the palate, pharynx and velopharyngeal ring were taken from the side of nasal ducts (during nasofiberscopy) in all patients. Moreover, each patient's speech was recorded for listening analysis and nasometric recordings were performed. The anatomical conditions and the quality of speech were evaluated on the basis of direct examinations as well as nasofiberscopy and nasometry [29]. The evaluation included: the length of the soft palate, velopharyngeal closure, intelligibility of speech, open nasality and nasalance ratio.

The length of the soft palate was measured by means of a paper ruler. This was used to measure the distance between the posterior margin of the hard palate and the tip of the uvula with the patient on the operating table prior to and after the operation, as well as at the follow-up examination. In children, the palate was anesthetized with a local anesthetic in order to abolish reflexes [30, 31].

The evaluation findings of a group of 20 healthy subjects aged 15–20 years (mean 18) were taken as a reference; the length of the soft palate was found to range between 29 and 41 mm, with a mean of 35 mm.

Velopharyngeal closure, mobility of the soft palate and of the posterior and lateral pharyngeal walls, and the position of the muscles were evaluated during phonation by means of nasofiberscopy (Storz 1101 TP Nasofiberscope). The qualification criteria for individual procedures were based on morphological and functional evaluation of the palate, i.e. position and location of the palatine levator muscles and dynamics of the velopharyngeal ring.

The severity of velopharyngeal insufficiency was established on the basis of criteria presented by an international working group [32]. The size of the open velopharyngeal space was evalu-

ated in relation to the resting conditions [33] and classified on a 4-point scale: (1) 100% = full closure, (2) 80–100% = slight insufficiency, (3) 50–80% = marked insufficiency, and (4) <50% = extreme insufficiency.

In order to evaluate speech intelligibility, open nasality and the nasalance ratio, older children and adults were asked to read a text which did not contain any nasal sounds in Polish. The patients were asked to repeat the text several times, and the words pronounced unintelligibly were marked [34, 35]. Speech intelligibility was classified on a 1–5 point scale [36, 37]: (1) fully intelligible speech, (2) <10% not fully intelligible = good intelligibility; (3) 10–20% not fully intelligible = limited intelligibility; (4) 20–30% not fully intelligible = poor intelligibility and (5) >30% not fully intelligible = unintelligible speech.

The severity of open nasality was evaluated on the same 1–5 scale: (1) norm, (2) slight, (3) moderate, (4) marked and (5) severe.

The nasalance ratio was evaluated with the use of the System for Objective Automated Testing of Nasality manufactured by Unifon from Września. Computer software was used to plot a graphic record of the findings [38–40].

The physiological nasal resonance used as a reference was established on a check-up of 30 healthy subjects at about 15.1%. It was presented as a percentage and evaluated on a 5 point scale: (1) <20% = norm, (2) 21–25% = slight resonance, (3) 26–30% = moderate nasal resonance (requiring phoniatic therapy), (4) 31–35% = marked resonance (patients require phoniatic therapy and may qualify for surgical treatment) and (5) >35% = patients qualify for surgical therapy.

The Surgical Procedure

An anteriorly pedicled mucosal flap and a posteriorly pedicled mucomuscular flap were incised on both sides from the oral approach. Incision lines were marked according to Z-plasty principles. The flaps were incised at 70–80° angles. In the nasal layer, flaps positioned reversely than those situated from the side of the oral cavity were prepared. The scar tissue located between the upper pharyngeal sphincter and the palatine velum levator muscle was dissected. After double Z-plasty, the soft palate was elevated upwards and the level of the incision on the posterior pharyngeal wall was determined marking the closure plane [24]. A ring formed from the mucomuscular flaps elevated from the palatopharyngeal folds was located in the plane. The flaps were big enough to allow the soft palate to close the velopharyngeal space inferiorly, like a sphincter, in resting conditions.

Results

The final outcome of treatment was based on a combined evaluation of 4 parameters: closure, speech intelligibility, nasality and the nasalance index. It was assumed that patients whose parameter levels were at least at a good level, i.e. classified as '1' or '2', achieved 'very good' outcome of treatment. If none of the grades was below '3', the total outcome was considered as 'improved'; it was believed that the patients did not require

surgical treatment and could rely on speech therapy. 'Poor' treatment outcome was reserved for cases in which at least 1 of the grades was '4' or '5'.

On follow-up examination 6 months after the operation (after 3 months for 3 patients), all patients were asked to answer the question [41] 'How do you and your relatives evaluate the outcome of the operation?' with either 'very good', 'good' or 'poor'.

In the study group before the operation, 9 patients had velopharyngeal closure below 50%, while it ranged between 50 and 80% in 5 patients. After the operation, 4 patients had 80–100% closure and 10 patients achieved complete closure (100%). Before the operation, the mean length of the palate was about 26.7 mm, after the operation, it was 29.8 mm.

Prior to the therapy, 2 patients had unintelligible speech, 10 had poorly intelligible speech and 2 patients had limited intelligibility. After the operation, speech was fully intelligible in 4 patients, not fully intelligible in 9 patients and only 1 patient had limited intelligibility of speech. Nine patients had had hypernasality. After the operation 2 patients were found to be free from nasality, 8 had slight nasality and 4 had moderate nasality. Prior to the operation, all patients had an abnormal nasalance ratio, with the mean value above 44%. After the operation, only 3 patients revealed values ranking between 26 and 30%, and they were found to have moderate hypernasality, while the mean value decreased to 24% (table 1).

Ten patients achieved full recovery (71%), the remaining 4 had improved recovery (29%). These latter patients qualified for further speech therapy.

Discussion

The earliest surgical modality aiming at lengthening the palate was performed in 1937. It used the 'push-back' method proposed by Wardill [42] and Dorrance [22]. However, in many patients the range of elongation with this method is inadequate and the inevitable exposition of the palatine processes may lead to impairment of blood supply to the maxilla, resulting in disturbances in its growth. A method introduced by Sommerland et al. [43] is a way of palatal lengthening by anatomical microscopic reconstruction of the palatal structures.

In 1978, Leonard Furlow [44] proposed primary sewing of the soft palate by means of alternate, double Z-plasty. Subsequently, the Furlow operation was also used in the therapy of velopharyngeal insufficiency [33, 45–

Table 1. Data evaluation

Patient	Sex/age ¹	Clefts	Year of Orticochea + Furlow operation	Palate length		Velopharyngeal closure		Intelligibility		Nasality		Nasalance ratio		Result	Patient's opinion
				pre	post	pre	post	pre	post	pre	post	pre ²	post ³		
B.T.	M/21	CLP-B	07	29	31	<50	80–100	5	2	5	3	48	24	i	g
B.M.	M/20	CLP-B	07	28	31	<50	80–100	5	2	5	3	47	30	i	g
W.E.	F/8	CLP-B	07	25	29	50–80	100	4	1	5	2	44	24	vg	vg
O.D.	M/4	CLP-B	07	24	28	50–80	100	3	1	5	2	43	25	vg	vg
B.M.	M/17	CP	07	28	29	<50	80–100	4	2	4	2	40	25	vg	vg
G.A.	F/21	CP-SM	07	29	31	<50	80–100	4	3	4	3	43	28	i	vg
S.P.	M/27	CLP-B	07	27	30	<50	80–100	4	2	4	3	45	28	i	vg
G.J.	F/5	CLP-B	07	26	30	50–80	100	4	1	5	1	47	20	vg	vg
S.M.	F/4	CLP-L	08	25	29	<50	100	4	1	5	2	45	20	vg	vg
W.R.	M/14	CLP-B	08	28	30	<50	80–100	3	2	4	2	40	24	vg	vg
M.N.	F/4	CP-SM	08	25	29	<50	100	4	2	5	1	45	20	vg	vg
R.W.	F/8	CP	08	26	28	50–80	100	4	2	5	2	46	22	vg	vg
P.M.	F/16	CLP-L	08	27	31	50–80	100	4	2	4	2	42	24	vg	vg
K.O.	F/24	CLP-R	08	28	32	<50	100	4	2	5	2	44	25	vg	vg

vg = Very good; i = improvement; g = good; CP = cleft palate only; CLP = cleft lip and palate; L = left; R = right; B = bilateral; SM = submucosal.

¹ Mean = 14. ² Mean = 44.2. ³ Mean = 24.2.

51]. The operation consists of lengthening the palate and improving levator muscle function.

Sphincter surgery was proposed by Hynes in 1950 [52]. An original method, called 'pharyngoplasty by muscle transplantation' by the author, it consists of medial rotation of inferiorly based flaps harvested from lateral pharyngeal walls into a transverse incision on the posterior pharyngeal wall. In 1968, Orticochea [53] developed a modification of this method. He incised mucomuscular flaps well supplied in blood and nerve vessels alongside palatopharyngeal folds containing the palatopharyngeal muscle fibers.

Reduction of the velopharyngeal space is also achieved by operations augmenting the posterior pharyngeal wall. Initially this aim was achieved by paraffin injections. In 1963, Blocksma [54] applied silastic implants, 2 years later Lewy et al. [55] used teflon, and in 1989, Woplford et al. [56] implanted proplast into the posterior pharyngeal wall. In order to approximate the posterior pharyngeal wall to the soft palate, autogenic tissue grafts, e.g. fat [57], fascia and cartilage [25, 58], and corium [28], are also used. According to many authors, improvement or recovery is achieved in about 70–80% of cases, but as it may be assumed, these concern patients with less severe forms of velopharyngeal insufficiency [25, 26, 56].

In the opinion of some authors, candidates for the Furlow operation include mainly patients with mild velopharyngeal insufficiency [17, 45, 50], in whom the lack of full closure in the central part is due to a mobile soft palate which is short or weak. Double Z-plasty lengthens the soft palate and improves its function. A complete recovery or improvement was achieved in 56–97% of patients [33, 45–49].

The aim of sphincter pharyngoplasty is to form a dynamic sphincter by inseting flaps and creating a transverse fold on the posterior pharyngeal wall. The operation is especially recommended in children and teenagers. It is believed that better results are achieved in patients with circular central insufficiency and a velopharyngeal opening not bigger than 5 mm [23]. Also patients with weaker mobility of the lateral walls may qualify for the procedure, but the mobility of the soft palate must be maintained [17]. According to various authors, a complete or partial improvement is obtained in 70–85%, even 95%, of patients [23, 59–64].

In patients with a cleft palate, velopharyngeal insufficiency results mainly from poor mobility of the soft palate. This is associated with a shorter palate, oblique position of the levator muscles and the presence of scar contractures. Since May 2007, all patients with a severe form of the deformity and an abnormal 'oblique' malposition

of the levators (including those with an almost transverse position of the levator muscles) have been qualified for double Z-plasty and sphincter pharyngoplasty performed in 1 operation. In such cases, it was believed that there was an effective possibility to lengthen the palate and improve its function [27].

The Furlow operation improves mobility, but does not always enable satisfactory elongation of the soft palate [48]. It may happen that the palate contracts and shortens slightly with time. Although double Z-plasty alone would probably prove to be effective in some of the patients, possibly leading to a full recovery or at least improvement [65], in our opinion additional narrowing of the velopharyngeal space from the back and the sides improves anatomical conditions and almost enables physiological reconstruction of the articulatory mechanisms. This complex management seems to be much more effective than combining the Furlow operation with augmentation of the posterior pharyngeal wall as it allows it to maintain the sphincter function, which, in case of augmentation, may become limited [28]. Velopharyngeal closure depends on the cooperation of the soft palate and pharyngeal walls (which is included in the name itself). The surgeon can never be sure whether corrective surgery undertaken to improve the efficacy and length of the palate will produce the expected anatomic and, more importantly, functional effect. For these reasons, narrowing of the velopharyngeal space increases the probability of achieving the desired result [66].

During posterior pharyngeal wall flap pharyngoplasty, it is possible to narrow, or rather to cover, the nasopharynx in a free way. Complete or partial recovery from velopharyngeal insufficiency is obtained in 75% [67–69] (or about 90% according to other authors [20]) of cases. However, this procedure is not free from limitations. The postoperative course may be characterized by nocturnal apnea (up to 90% of patients [70–73]) and nasal congestion. The healing and scar formation processes occurring on the surface of the wound may cause tubulization of the flap leading to about a 50% narrowing of the initial space and to downward dislocation of the flap pedicle [74]. Thus, good early results of treatment may deteriorate with time. Some authors also point to disturbances in the growth of the maxilla [75, 76] and the mandible [77]. However, other authors have suggested that these unwanted consequences of velopharyngoplasty occur only in a small percent of the cases [78].

Although the short follow-up period and small number of the patients seems to be unsatisfactory for justifying the final results of the new method, in our material a complete

recovery was achieved in 10 patients (71%) and significant improvement was achieved in the 4 oldest patients (29%). It seems that consistent rehabilitation may improve the final outcome. Prospective evaluation proves the efficacy of the method. Nasal congestion observed in all patients in the early postoperative period was of a transient character and withdrew completely in the first 2 months.

As in the case of primary lip and plate cleft surgery, there is also no universal management scheme for patients with velopharyngeal insufficiency. This is associated with a lack of a universal, complete system of speech evaluation. There is, however, universal consent that the operation should be performed as soon as possible, preferably before the age of 6 years [17–19]. On the basis of prospective studies carried out at our center since 2001, a novel modality consisting of a simultaneous Furlow operation and sphincter pharyngoplasty was proposed in 2007. This management was based on the premise that all patients who do not need posterior wall flap pharyngoplasty (mainly due to tissue deficit) and have a real chance for palate performance improvement qualify for double Z-plasty. A simultaneously performed Orticochea operation improves the efficacy of treatment and is free from any additional risk.

Conclusions

In conclusion, a 1-stage Furlow operation and sphincter pharyngoplasty are effective modalities in the therapy of velopharyngeal insufficiency. Indications for posterior pharyngeal flap pharyngoplasty should be limited to the cases in which a simultaneous Furlow operation and sphincter pharyngoplasty are not possible due to a deficit of the palatine tissue.

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