Impact of pharyngofixation in cleft palate repair surgery on the development of chronic adhesive otitis media

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Abstract

Background: A significant percentage of children with cleft palate suffer from otitis media with effusion and its consequences, such as deafness, chronic adhesive otitis and cholesteatoma. This study aimed to determine whether these effects can be minimised by selecting pharyngofixation as the surgical technique for cleft palate correction.

Methods: A retrospective study was performed of 155 patients (308 ears) who underwent surgery from age 5 months to 8 years and were followed up for 36–84 months.

Results: In all, 125 ears (41 per cent) had epitympanic retraction, 45 ears (14 per cent) had sinus tympani retraction and 5 patients (3 per cent) had cholesteatoma. Use of the pharyngofixation technique did not significantly correlate with (1) the severity of otological findings or (2) the incidence of retraction pockets in the epitympanum and sinus tympani (p = 0.53).

Conclusion: Pharyngofixation did not significantly alter the severity of long-term otological findings.

Key words: Cholesteatoma; Cleft Palate; Eustachian Tube; Middle Ear Ventilation; Otitis Media with Effusion

Introduction

Cleft palate patients have multiple handicaps affecting their appearance and pronunciation, including occlusion disorder and impaired hearing (due to otitis media with effusion (OME) and its sequelae). They are also at risk of cholesteatoma and its complications. Malformation of the epipharyngeal region causes an enlarged epipharyngeal space, localised postero-inferior narrowing of the pharyngeal tubal ostium, tube cartilage hypoplasia, and shortening and general deformity of the Eustachian tube. Additionally, the angle between the tubal cartilage and the tensor veli palatini muscle is wider and the muscle inserts into a different location. 1-4 Malformation of the tensor and levator veli palatini muscles, especially in the hamulus area, prevent normal opening of the pharyngeal tubal ostium during swallowing and yawning.1,2 Pneumatisation of the temporal bone pyramid is reduced by thinning in the mediolateral and anteroposterior directions, resulting in a significantly decreased volume, similar to in sclerotised bones. 1,2 However, this causes OME to develop due to poor long-term middle-ear ventilation. The consequences are conductive hearing loss, defective and delayed speech

development, consecutive episodes of chronic adhesive otitis (retraction pockets), and acquired cholesteatoma. The incidence of acquired cholesteatoma in these children is 100–200 times higher than in unaffected children, ranging between 1 and 6 per cent.^{3,5–8} Different studies have documented the relationship between the type of corrective cleft palate surgery and the development of hearing loss or OME, ^{5,7,9–13} compared different ventilation tube induction protocols ^{3,14,15} and explored the relationship between cleft type and cholesteatoma development.⁶ After pharyngofixation using the inferiorly based pharyngeal flap, the incidence of OME is reported to decrease significantly. ¹²

Various procedures can be used to correct the palate defect: the most common are Veau-Wardill-Kilner palatoplasty with two lobes, von Langenbeck palatoplasty with two or three lobes and Furlow double-opposing Z-plasty. ^{9,11} The first two can be supplemented by pharyngofixation using an inferiorly or superiorly based pharyngeal flap to minimise velopharyngeal insufficiency; the latter two are used to minimise maxillary growth restriction.

This study aimed to determine whether the use of pharyngofixation in corrective cleft palate surgery was associated with the occurrence of OME sequelae such as chronic adhesive otitis or cholesteatoma.

Materials and methods

Patients

A retrospective study was performed of patients who underwent cleft palate repair at the Cleft Palate Care Centre of Královské Vinohrady University Teaching Hospital between 2001 and 2006. Patients were divided into four groups according to age at the time of surgery (range, 5 months to 8 years; mean \pm SD, 36 ± 19.45 months): 0–29, 30–39, 40–59 and at least 60 months.

Surgery

Patients underwent Veau-Wardill-Kilner palatoplasty with two lobes or von Langenbeck palatoplasty with two or three lobes, with optional pharyngofixation using an inferiorly based pharyngeal flap. The use of pharyngofixation was not randomised: the decision was made by the surgeon. In general, pharyngofixation was used more often in surgery for complicated clefts with significant velopharyngeal insufficiency and in revision surgery.

In pharyngofixation, the inferiorly based flap containing the mucosal and muscle layer on the posterior wall of epipharynx was elevated. The flap was then tubulised, tumed anteriorly and sutured in the midline to the nasal surface of the repaired soft palate close to the uvula. The defect was covered by primary suture of the mobilised mucosal layer. Complete healing took approximately six weeks.

Outcome

Patients were followed up for 36-84 months to evaluate the development of adhesive and/or retraction changes, especially retraction pockets and cholesteatoma. As Eustachian tube function gradually improves after the age of six or seven years, the most important examinations were those performed after patients had reached seven years of age. The degree of retraction, an important prognostic factor for cholesteatoma development, was assessed using the Charachon scale for the superior posterior quadrant and the Tos scale for epitympanic retractions. After otological assessment, patients were divided into four groups according to their need for further surgical intervention: normal group (normal otology findings); no surgical intervention group (Tos grade I-II, Charachon grade I; surgical treatment not needed); possible surgical intervention group (Tos grade III, Charachon grade II; surgical intervention may be needed); and definite surgical intervention group (Tos grade IV, Charachon grade III; requiring retraction pocket elevation or resection and tympanoplasty plus eventual scutum reconstruction).

Ethical standards

All procedures complied with the ethical standards of the relevant national and institutional guidelines on human experimentation and with the Helsinki Declaration of 1975, as revised in 2008.

Statistical analysis

Ordinal logistic regression was used because the use of pharyngofixation was unequally distributed among age groups. This statistical method allowed the influence of age at the time of surgery to be disregarded. The test hypothesis was that a relationship exists between the severity of otological findings (as expressed by surgical group, i.e. Tos and Charachon grade) and use of the pharyngofixation technique.

Results

Of the 180 patients (359 ears) examined, 22 were lost to follow up and 3 had a cleft as part of a syndrome defect. A total of 155 patients (85 boys and 70 girls; 308 ears) for whom complete surgical and follow-up data were available were included in the final analysis. They were aged 7–11 years at the time of significant investigation.

At the final follow-up, 72 patients had a complete cleft (46 per cent); of these, 52 (72 per cent) had undergone pharyngo fixation. In all, 83 patients had a palate cleft (54 per cent); of these, 32 (39 per cent) had undergone pharyngo fixation.

Assessment of malformations identified one external auditory canal atresia, one microtia connected with hypoplasia of half of the face, two middle-ear malformations (one of which was associated with a third mobile window – the large internal auditory meatus), three auricle malformations, four preauricular appendices and four external auditory meatus stenoses were identified. A total of 56 patients had otitis media with effusion (OME).

Tympanostomy was performed once in 18 patients, twice in 8 patients, 3 times in 10 patients, 4 times in 4 patients, 5 times in 1 patient and 6 times in 1 patient.

At the final follow up, 14 patients had bilateral OME and 25 had unilateral OME. Five patients had tympanostomy tubes at presentation, four bilaterally. The parents of five patients with OME lasting for at least three months refused the recommended tympanostomy. Two patients had a perforated eardrum; one of these had undergone three tympanostomies. Retraction pocket elevation or resection and myringoplasty or tympanoplasty were indicated for all 23 ears in the definite surgical intervention group and 23 ears in the possible surgical intervention group (53 per cent). In all, five patients (3 per cent) developed cholesteatoma, two developed epitympanic cholesteatoma, two developed sinus cholesteatoma and one developed extensive cholesteatoma of the middle ear, mastoid and epitympanum with supralabyrinthine propagation. Cholesteatoma was unilateral in three patients and bilateral in two; four of these patients

TABLE I SURGERY WITHOUT PHARYNGOFIXATION: OTOLOGICAL OUTCOME BY AGE AT TIME OF SURGERY

Age group (mon)	Otological outcome*			
	Group 0	Group 1	Group 2	Group 3
0-29 30-39 40-59 ≥60	38 (51.4) 10 (50.0) 12 (70.6) 13 (59.1)	14 (18.9) 8 (40.0) 2 (11.8) 6 (27.3)	18 (24.3) 1 (5.0) 2 (11.8) 1 (4.5)	4 (5.4) 1 (5.0) 1 (5.9) 2 (9.1)

Data are n (%). *Group 0, normal otological findings; group 1, Tos grades I–II or Charachon grade I; group 2, Tos grade III or Charachon grade II; Group 3 Tos grade IV or Charachon grade III. Mon = months

TABLE II SURGERY WITH PHARYNGOFIXATION: OTOLOGICAL OUTCOME BY AGE AT TIME OF SURGERY

Age group (mon)	Otological outcome*				
	Group 0	Group 1	Group 2	Group 3	
0-29	2 (50.0)	0 (0.0)	2 (50.0)	0 (0.0)	
30-39	62 (59.0)	25 (23.8)	14 (13.3)	4 (3.8)	
40-59	20 (47.6)	14 (33.3)	4 (9.5)	4 (9.5)	
≥60	15 (62.5)	3 (12.5)	2 (8.3)	4 (16.7)	

Data are n (%). *Group 0, normal otological findings; group 1, Tos grades I–II or Charachon grade I; group 2, Tos grade III or Charachon grade II; Group 3 Tos grade IV or Charachon grade III. Mon = months

had previously undergone tympanostomy (once for one patient, twice for one and three times for two). Otological findings in patients who underwent surgery without pharyngofixation are shown in Table I and in patients who underwent pharyngofixation otological findings are shown in Table II. Figure 1 shows the distribution of otological findings with respect to pharyngofixation and age at the time of surgery.

As there was an unequal distribution of patient age with respect to pharyngofixation, the statistical analysis could not evaluate the influence of each factor separately. Ordinal logistic regression is a multivariate statistical method that can separately assess the effects of several independent factors on an observed endpoint. Pharyngofixation and age at the time of surgery were used as independent factors (regressors) in the logistic regression model. This analysis showed that neither pharyngofixation (p=0.53) nor age (p=0.48) had a significant effect on post-operative otological findings.

Discussion

Cleft palate patients are at a higher risk of cholesteatoma, among other disorders, compared with healthy children. 3,6-8,10 Most have otitis media with effusion (OME) from early childhood, 1,3,5,16,17 which can develop into chronic adhesive otitis and cholesteatoma. There has been considerable discussion on how to improve OME progression and its sequelae with conservative treatments or different strategies using venti-Treatment varies from obligatory lation tubes. grommet insertion for all cleft palate patients to a watch-and-wait strategy. 1,9,14,15 Sheahan et al. suggested a conservative approach because of the higher incidence of cholesteatoma and retraction pockets in patients who undergo multiple tympanostomies.3 However, whether these problems are the cause or sequelae of multiple tympanostomies is open to debate. In contrast, Spilsbury et al. advocate early tympanostomy in these patients because their tubal function is very poor.

In the cleft palate centre in the present study, patients undergo tympanostomy and re-tympanostomy after three months of persistent middle-ear secretion, and retraction pockets are managed by elevation or resection and myringoplasty or tympanoplasty. These procedures should be timed to take place when an assessment

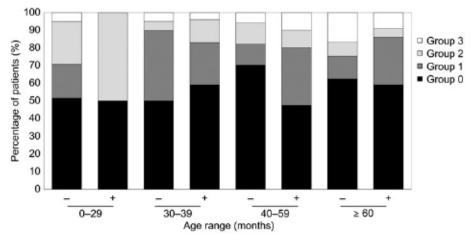


FIG. 1

Distribution of ear-findings in groups 0-3 with respect to pharyngofixation stratified by age at the time of surgery. Group 0, normal otological findings; group 1, Tos grades I-II or Charachon grade I; group 2, Tos grade III or Charachon grade II; Group 3 Tos grade IV or Charachon grade III.

of Charachon grade II in the middle ear or Tos grade III in the epitympanum has been made.

The impact of cleft palate reconstruction techniques on hearing and tubal function improvement has been investigated. 9,13,18 The surgical technique usually has no significant influence on these parameters, although Smith *et al.* demonstrated a significantly lower frequency of tympanostomy when using Furlow double-opposing Z-plasty. 11 So far, no studies have reported an association between the surgical technique used for cleft palate correction and post-operative otological findings.

- Few studies have investigated whether the surgical cleft palate correction technique affects otological outcome
- Cholesteatoma incidence is 100–200 times more common in children with a cleft palate than in unaffected children
- Cholesteatoma increases the risk of intracranial complications and hearing loss, and causes considerable discomfort
- Pharyngofixation is reported to decrease the incidence of otitis media with effusion, a common premorbidity to adhesive otitis and cholesteatoma
- This study found no significant effect of pharyngofixation on the long-term incidence of retraction pockets and cholesteatoma

Spauwen et al. reported a significant 60 per cent reduction in the incidence of OME at 3 months after pharyngofixation in a group of 51 patients. These results were explained by an apparent reduction in oronasal air leakage and retro-positioning of the levator veli palatini muscle hinge. Consistent with these findings, the present study found no relationship between surgery type, the sidedness of the cleft and the occurrence of bilateral OME. There was no positive long-term impact of pharyngofixation on otological findings. However, it is possible that the use of a randomised selection procedure might have produced different results. Alternatively, pharyngofixation may have only a short-term effect on the otological outcome.

Further evidence showing that pharyngofixation has no effect on otological outcome was provided by the theoretical work of Sheer and colleagues using a three-dimensional mathematical model based on the finite element method. They showed that in children, particularly in those with a cleft palate, tubal function depends much more on the function of the peri-luminal mucosal tissue than of the tensor veli palatini muscle.⁴

Conclusion

This retrospective study found that the type of cleft palate surgery had no impact on otological outcome. However, the study had the same limitation of all retrospective studies: lack of randomisation. Therefore, it cannot be definitively stated that pharyngofixation has no effect on the otological outcome. A prospective study using randomised selection of the surgical procedure is necessary to demonstrate the influence of the surgical technique used for cleft palate correction on otological outcomes. This should exclude cases in which tubal and nasopharyngeal malformations are so extensive that no type of corrective cleft palate surgery (including pharyngofixation) would be expected to have a positive long-term otological outcome.⁴

References

- 1 Kuo CL, Lien CF, Chia HC, Shiao AC. Otitis media with effusion in children with cleft lip and palate: A narrative review. Int J Pediatr Otorhinolaryngol 2013;77:1403–9
- 2 Rajion ZA, Al-Khatib AR, Netherway DJ, Townsend GC, Anderson PJ, McLean NR et al. The nasopharynx in infants with cleft lip and palate. Int J Pediatr Otorhinolaryngol 2012; 76:227-34
- 3 Sheahan P, Blayney AW, Sheahan JN, Earley MJ. Sequelae of otitis media with effusion among children with cleft lip and/ or cleft palate. Clin Otolaryngol 2002;27:494-500
- 4 Sheer FJ, Swarts JD, Ghadiali SN. Three-dimensional finite element analysis of Eustachian tube function under normal and pathological conditions. *Med Eng Phys* 2011;34:605–16
- 5 Alper M, Losee JE, Mandel EM, Seroky JT, Swarts JD, Doyle WJ. Pre- and post-palatoplasty Eustachian tube function in infants with cleft palate. Int J Pediatr Otorhinolaryngol 2012; 76:388-91
- 6 Harris L, Cushing SL, Hubbard B, Fisher D, Papsin BC, James AL. Impact of cleft palate type on the incidence of acquired cholesteatoma. Int J Pediatr Otorhinolaryngol 2013;77:695–8
- 7 Spilsbury K, Miller I, Semmens JB, Lannigan FJ. Factors associated with developing cholesteatoma: a study of 45,980 children with middle ear disease. *Laryngoscope* 2010;120:625–30
- 8 Vincenti V, Marra F, Bertoldi B, Tonni D, Saccardi MS, Bacciu S et al. Acquired middle ear cholesteatoma in children with cleft palate: experience from 18 surgical cases. Int J Pediatr Otorhinolaryngol 2014;78:918–22
- 9 Carroll DJ, Padgitt NR, Liu M, Lander TA, Tibesar RJ, Sidman JD. The effect of cleft palate repair technique on hearing outcomes in children. Int J Pediatr Otorhinolaryngol 2013;77: 1518–22
- 10 Reiter R, Haase S, Brosch S. Repaired cleft palate and ventilation tubes and their associations with cholesteatoma in children and adults. Cleft Palate Craniofac J 2009;46:598–602
- Smith LK, Gubbels SP, MacArthur CJ, Milczuk HA. The effect of the palatoplasty method on the frequency of ear tube placement. Arch Otolaryngol Head Neck Surg 2008;134:1085–9
 Spauwen PH, Ritsma RJ, Huffstadt BJ, Schutte HK, Brown IF.
- 12 Spauwen PH, Ritsma RJ, Huffstadt BJ, Schutte HK, Brown IF. The inferiorly based pharyngoplasty: effects on chronic otitis media with effusion. Cleft Palate Craniofac J 1998;25:26-32
- 13 Tiwari R, Shama RK, Panda NK, Munjal S, Makkar S. Tensor tenopexy: a clinical study to assess its efectiveness in improving Eustachian tube function and preventing hearing loss in patients with cleft palate. J Plast Reconstr Aesthet Surg 2013;66: e239–45
- 14 Klockars T, Rautio J. Early placement of ventilation tubes in cleft lip and palate patients: does palatal closure affect tube occlusion and short-term outcome? Int J Pediatr Otorhinolaryngol 2012;76:1481-4
- 15 Szabo C, Langevin K, Schoem S, Mabry K. Treatment of persistent middle ear effusion in cleft palate patients. Int J Pediatr Otorhinolaryngol 2010;74:874–7
- 16 Bluestone CD. Anatomy and physiology of the eustachian tube. In: Cummings CW, Fredrickson JM, Harker LE, eds. Otolaryngology Head and Neck Surgery, 3rd edn. St. Louis: Mosby, 1998;3003–22
- 17 Phua YS, Salkeld LJ, de Chalain TMB. Middle ear disease in children with cleft palate: protocols for management. Int J Pediatr Otorhinolaryngol 2009;73:307–13

18 Guneren E, Ozsoy Z, Ulay M, Eryilmaz E, Ozkul H, Geary PM. A comparison of the effects of Veau-Wardill-Kilner palatoplasty and Furlow double-opposing Z-plasty operations on eustachian tube function. Cleft Palate Craniofac J 2000;37:266-70

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RHINOLOGY

Mucosal maxillary cysts: long-term subjective outcomes after surgical treatment

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Abstract Mucosal maxillary cysts (MMCs) are usually asymptomatic and are often diagnosed as an incidental finding. The aim of this study is to assess clinical significance of MMCs and the long-term effect of surgical treatment on the symptoms initially addressed to MMCs. The study included a retrospective analysis of 64 patients who had undergone surgery for MMC using a questionnaire focused mainly on the effect surgery had on symptoms. Mean time of follow-up was 79 months. Patients were also divided and compared according to the presence of rhinitic symptoms. Twenty-six patients (63.4 %) reported complete disappearance of symptoms, 8 (19.5 %) reported improvement, 4 (9.7 %) reported no change in symptoms following surgery and 3 (7.3 %) reported that symptoms reappeared. Significantly (p = 0.0365) better results were achieved in patients without preexisting rhinitic symptoms. This study supports the opinion that in some cases, MMCs are involved in the development of sinonasal symptoms. Surgical treatment leads, in most patients, to disappearance or improvement of symptoms and the effect is better in patients without rhinitic symptoms.

Keywords Maxillary sinus cysts · Endonasal surgery · Long term outcomes · Retrospective study

Introduction

Mucosal cysts of the maxillary sinus (MMCs) are among the most frequent findings from imaging of the paranasal

P. Schalek (☑) · L. Otruba · Z. Hornáčková · A. Hahn ENT Department of 3rd Medical Faculty of Charles University, Šrobárova 50, 100 34 Prague 10, Czech Republic e-mail: schalekp@hotmail.com sinuses. MMCs are usually asymptomatic and are often diagnosed as an incidental finding on imaging done for other reasons. Prevalence in the general population is reported to range from 3.2 to 35.6 % [1–5]. A recent study conducted on 6,831 subjects with paranasal MRIs reported the prevalence of paranasal sinus cysts to be 7.4 %, with most (93 %) being located in the maxillary sinus [1]. The pathogenesis of MMCs has not been fully elucidated; however, an origin based on an obstruction of the duct of seromucinous mucosal gland is considered likely [6].

Some authors have reported a link between the presence of MMCs and clinical symptoms (facial pain, nasal obstruction, rhinorrhea, cephalea) [7, 8], the clinical significance of MMCs, if any, and the effectiveness of surgical treatment has not yet been fully documented.

The aim of this study was to assess the long-term effectiveness of surgical treatment on the symptoms initially associated with MMCs.

Materials and methods

A retrospective analysis of 64 patients who had undergone surgery for MMC between February 2001 and December 2009 was added by distribution of a questionnaire focused on: (a) effectiveness of surgery relative to symptoms, (b) seeking other medical care for the same symptoms, (c) undergoing further sinonasal surgery and (d) current status of nasal symptoms. The time between surgery and completion of the questionnaire (May 2012) ranged from 29 to 136 months (mean 79 months).

The study included only symptomatic patients older than 18 years with a typical presentation of MMCs on their CT scan. Patients with sinonasal symptoms (facial pressure/pain, headache, nasal obstruction and rhinorrhea) located ipsilaterally to the affected maxillary sinus were considered as symptomatic.

Patients with odontogenic cysts and patients who had undergone concomitant sinonasal surgery in the same procedure with the removal of the cyst (e.g. septoplasty, septorhinoplasty, functional endonasal sinus surgery, reduction of volume of an inferior turbinate) were not enrolled in the study. In all patients with facial pain and cephalalgia, neurological cause had been excluded. All patients were indicated for the surgery after failure of medical treatment which included mainly intranasal steroids, antihistaminics, nasal salines and analgesics.

All patients signed consent to the publication of their anonymized data and the study was approved by local ethic committee.

Subjective evaluation

Patients evaluated the effectiveness of surgery as: (a) complete disappearance of symptoms, (b) improvement of symptoms, or (c) no change in symptoms or return of symptoms after an asymptomatic period.

Patients evaluated their current sinonasal status as: (a) no sinonasal symptoms, (b) mild symptoms but without treatment or (c) symptoms requiring treatment.

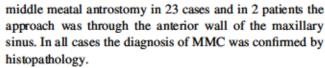
Patients were also divided into two groups and compared based on symptoms. The first group (25 patients) consisted of patients without rhinitic symptoms (i.e. nasal obstruction, discharge, postnasal drip) but experiencing facial pain/pressure or headache. The second group (16 patients) consisted of patients with rhinitic symptoms, regardless of facial pain.

Statistical analysis

The Chi square test was used for the comparison of (a) nonrhinitic and rhinitic patients and (b) surgical approaches (supra- vs. infra-turbinal antrostomy), relative to complete disappearance of symptoms. p value less than 0.05 was considered statistically significant.

Results

Of the 64 patients who met the criteria for study inclusion 36 (56.25 %) were men and 28 (43.75 %) were women, mean age was 40.4 years (range 18-70 years). In 34 patients the cyst was located in the right maxillary sinus, in 28 patients in the left maxillary sinus and in two cases the cysts were bilateral. All cysts were removed endoscopically. Infraturbinal antrostomy was performed in 41 cases,



During the follow-up period one patient died and contact was lost with five patients. In two patients an antrochoanal polyp developed on the 'procedure' side and the patients underwent a second procedure and therefore were excluded. Of the remaining 56 patients, 41 (73.2 %) responded to the questionnaire. Of those patients 26 (63.4 %) reported complete disappearance of symptoms, 8 (19.5 %) reported improvement, 4 (9.7 %) reported no change in symptoms following surgery and 3 (7.3 %) reported that after a symptom-free interval, symptoms reappeared (Fig. 1).

After surgery and during the follow-up period only nine patients sought medical care for the same reasons reported prior to participating in this study. Seven patients received further treatment by an otorhinolaryngologist, three by a neurologist and one by a dentist.

Apart from two patients treated for antrochoanal polyps, none had to undergo any further sinonasal surgical procedures.

At the time of completing the questionnaire 19 (46.3 %) patients had no sinonasal problems, 13 (31.7 %) patients had mild symptoms without necessity of treatment and 9 (21.9 %) patients continued medical treatment (Fig. 2).

When comparing the effectiveness of surgical treatment, relative to complete resolution of symptoms and with regard to the preexistence of rhinitic symptoms prior to surgery, 76 % patients (19 patients out of 26) without rhinitic symptoms reported complete resolution versus 43.7 % of non-rhinitic patients (7 patients out of 16). This difference was statistically significant (p = 0.0365) (Fig. 3). Comparison of surgical approaches (supraturbinal vs. infraturbinal antrostomy) demonstrated slightly better results for supraturbinal antrostomy (66 % patients without symptoms postoperatively vs. 58 % for infraturbinal

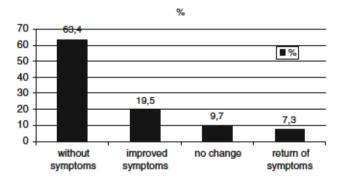


Fig. 1 Effect of surgery on the symptoms: 26 patients (63.4 %) reported complete disappearance of symptoms, 8 (19.5 %) reported improvement, 4 (9.7 %) reported no change in symptoms following surgery and 3 (7.3 %) reported that after a symptom-free interval, symptoms reappeared



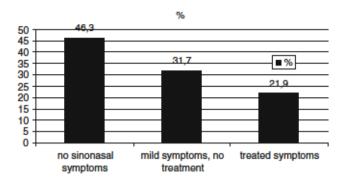


Fig. 2 Presence of sinonasal symptoms at the time of completing the questionnaire: 19 (46.3 %) patients had no sinonasal problems, 13 (31.7 %) patients had mild symptoms without necessity of treatment and 9 (21.9 %) patients continued medical treatment

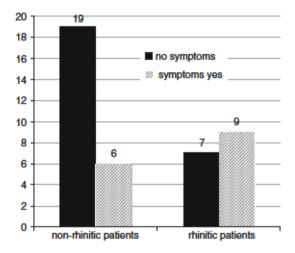


Fig. 3 Comparison of the effectiveness of surgical treatment, relative to complete resolution of symptoms and with regard to the preexistence of rhinitic symptoms prior to surgery: 76 % patients (19 patients out of 26) without rhinitic symptoms reported complete resolution versus 43.7 % of non-rhinitic patients (7 patients out of 16). This difference was statistically significant (p = 0.0365)

antrostomy). However, the difference was not statistically significant (p = 0.6048) (Fig. 4). Both patients with antrochoanal polyp were included in this analysis as a surgical failure.

Discussion

The etiology of MMCs is not entirely clear. A possible relationship between rhinosinusitis and MMC has been discussed in the literature. Harar et al. [5] in a study involving 110 patients with MMC concluded that chronic rhinosinusitis played an important role in the etiology of MMCs. Other studies have suggested that the presence of MMC does not reflect an obstruction of the ostiomeatal complex, nor does it reflect subjective or objective evidence of sinus disease [2, 4].

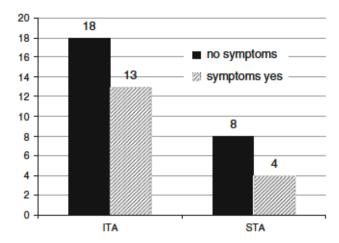


Fig. 4 Comparison of the effectiveness of surgical treatment, relative to complete resolution of symptoms and with regard to the surgical approach—supraturbinal antrostomy (STA) versus infraturbinal antrostomy (ITA) demonstrated slightly better results for STA: 66% (8 patients out of 12) with STA reported complete resolution versus. 58% (18 patients of 31) with ITA. However, the difference was not statistically significant (p=0.6048)

Moon et al. [1] demonstrated that smoking was associated with a higher risk of developing MMCs.

Most MMCs are asymptomatic and their mere presence is not an indication for surgery [1, 2]. Moreover, in studies with repeated MRI or radiography it was shown that most MMCs do not progress in size and in some cases MMCs undergo spontaneous shrinkage or completely disappear [9, 10].

Some authors have reported a link between the presence of MMC and clinical symptoms such as headache, facial pain and pressure, nasal obstruction and anterior or posterior rhinorrhea [7, 8]. In his study, Moon et al. [1] showed that sinonasal symptoms (nasal obstruction, discharge, sneezing) occurred in association with paranasal sinus cysts in only 2.6 % of the cases and in association with other airway symptoms (postnasal drip, wheezing, cough) in only 1.6 % of the cases; however, facial pain or pressure and headache were not mentioned in the study. The same authors, in another study, reported nasal symptoms in 4.5 % of patients with MMCs [10].

In our study, MMCs were the only established and surgically treated sinonasal pathology. From a long-term perspective, improvement or complete resolution of the original symptoms was achieved in almost 83 % of cases and at the time of completing the questionnaire only nine patients were receiving treatment for continued sinonasal symptoms. These outcomes are consistent with the position that some MMCs are associated with clinical symptoms. Significantly (p = 0.0365) better results were reported in the group of patients with facial pain/pressure or cephalea but without rhinitic symptoms (nasal obstruction, posterior or anterior rhinorrhea) compared to those with rhinitis.

Albu [11], in a recent study, concluded that symptoms in most patients with MMCs are not caused by the cyst itself and therefore during surgical treatment it is more important to restore ventilation and drainage of the affected sinus than to simply remove the MMC. In our study, approximately one-third of patients had a middle meatal antrostomy. Improved ventilation and drainage thorough the natural ostium can be expected with this procedure. The remaining two-thirds of patients had an osteoplastic antrostomy in the inferior meatus with intention to preserve intact ostiomeatal unit. Therefore we believe that the 83 % improvement or disappearance of symptoms, which we saw in our study, was not associated only with restoration of sinus ventilation. Comparing both approaches we did not prove significant difference with respect to complete resolution. To clarify this issue, a prospective, rhinoendoscopy and MRI controlled study should be conducted.

Two of our patients developed antrochoanal polyps (ACP) on the 'procedure' side. Although the etiology of ACPs is not completely clear, the sinus component of an ACP is histologically identical to a MMC [12] and it is possible that ACPs form from MMCs [13, 14]. However, the difference between the prevalence of ACPs, which account for only 4-6 % of all nasal polyps and the prevalence of MMCs (3.2-35.6 % in the general population), suggests that ACPs arise from only a small percentage of MMCs. Although the aim of surgery was complete removal of MMC (not only marsupialization), mere supra- or infraturbinal antrostomy (without extended endonasal or external approaches) cannot guarantee a radical procedure in all sublocalities of maxillary sinus. Question remains whether the ACP arises from small remnants of MMC or from a new cyst in patients showing an increased tendency to MMC formation.

Conclusion

This study supports the opinion that in some cases, MMCs are involved in the development of sinonasal symptoms. Surgical treatment is usually successful in most patients and leads to improvement or disappearance of symptoms, with even better results in those patients who present without rhinitic symptoms.

The choice of surgical approach should be the subject of further prospective study.

Conflict of interest None.

References

- Moon IJ, Lee JE, Kim ST, Han DH, Rhee CS, Lee CH, Min YG (2011) Characteristics and risk factors of mucosal cysts in the paranasal sinuses. Rhinology 49:309–314
- Kanagalingam J, Bhatia K, Georgalas C, Fokkens W, Miszkiel K, Lund VJ (2009) Maxillary mucosal cyst is not a manifestation of rhinosinusitis: results of a prospective three-dimensional CT study of ophtalmic patients. Laryngoscope 119:8–12
- Rodrigues CD, Freire GF, Silva LB, Fonseca da Silveira MM, Estrela C (2009) Prevalence and risk factors of mucous retention cysts in a Brazilian population. Dentomaxillofac Radiol 38:480–483
- Bhattacharyya N (2000) Do maxillary sinus retention cysts reflect obstructive sinus phenomena? Arch Otolaryngol Head Neck Surg 126:1369–1371
- Harar RP, Chadha NK, Rogers G (2007) Are the maxillary mucosal cyst a manifestation of inflammatory diseases? J Laryngol Otol 121:751–754
- Kaffe I, Littner MM, Mosksona D (2008) Mucosal antral cysts: radiographic appearance and differential diagnosis. Clin Prev Dent 10:3–6
- Hadar T, Shvero J, Nageris BI, Yaniv E (2000) Mucus retention cyst of the maxillary sinus: the endoscopic approach. Br J Oral Maxillofac Surg 38:227–229
- Rhodus NL (1990) The prevalence and clinical significance of maxillary mucous retention cysts in a general clinic population. Ear Nose Throat J 69:82–87
- Wang JH, Jang YJ, Lee BJ (2007) Natural course of retention cysts of the maxillary sinus: long term follow-up results. Laryngoscope 117:341–344
- Moon IJ, Kim SW, Han DH, Shin JM, Rhee CS, Lee CH, Min YG (2011) Mucosal cyst in the paranasal sinuses: long-term follow-up and clinical implications. Am J Rhinol Allergy 25:98–102
- Albu S (2010) Symptomatic maxillary sinus retention cysts: should they be removed? Laryngoscope 120:1904–1909
- Frosini P, Picarella G, De Campora E (2009) Antrochoanal polyp: analysis of 200 cases. Acta Otorhinolaryngol Ital 29:21–26
- Yaman H, Yilmaz S, Karali E, Guclu E, Ozturk O (2010) Evaluation and management of antrochoanal polyps. Clin Exp Otorhinolaryngol 3:110–114
- Sousa DW, Pinheiro SD, Silva VC, Bastos JP (2011) Bilateral antrochoanal polyps in an adult. Braz J Otorhinolaryngol 4:539



LETTER TO THE EDITOR

Quality of life in patients with chronic rhinosinusitis: a validation of the Czech version of SNOT-22 questionnaire

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Dear Sir,

Quality of life (QoL) is defined as the difference between expectations and experience [1]. A part of the overall QoL is health-related quality of life (HRQoL), which is influenced by the health of patients and can be changed through treatment. Since the 1990s, there have been increased efforts to evaluate QoL of patients in all medical fields. Tools used to evaluate QoL are either generic health instruments for assessing general conditions or disease-specific questionnaires focused on symptoms of a disease. Chronic rhinosinusitis (CRS) is a disease characterized by a high prevalence and significant reduction in QoL. Although today there are many diagnostic methods for evaluating sinonasal disease, it cannot be said that the results clearly correlate with QoL, as it is perceived by patients [2].

Until now, there has been no validated Czech version of the questionnaire for evaluating QoL in patients with CRS. We used sinonasal outcome test (SNOT-22) questionnaire [3], which is based on the SNOT-20 questionnaire [4]. The difference between the two assessment tools is the addition of two questions relating to the evaluation of (i) nasal obstruction and (ii) smell and taste, which can be regarded as important indicators of QoL. SNOT-22 is recommended as one of the best tools for assessing QoL in patients with CRS [5].

A group of 52 patients (31 men, 21 women; mean age 50.5 years) with CRS, either with or without nasal polyps (NP) who were scheduled to undergo endoscopic sinus surgery (ESS) were enrolled in the study. All the patients filled a SNOT-22 questionnaire (Czech version) preoperatively (T0), 3 months (T1), and 6 months (T2) after surgery.

There were two control groups: group 1 consisted of 50 patients admitted to the ENT clinic with non-sinonasal disease (24 men, 26 women; mean age 44.9 years); group 2 consisted of 50 healthy students of the Faculty of Medicine (22 men, 28 women; mean age 24.1 years). Both control groups also completed the SNOT-22 questionnaire.

The study was approved by local Ethics Committee and each patient signed an informed consent. Properties of the questionnaire were tested by determining reliability, validity, and sensitivity. Reliability (internal consistency) was tested using Cronbach's α and test-retest reliability determination. For the latter (determination of the stability of the questionnaire over time), 10 patients completed the questionnaire for a second time—3 weeks after completing the questionnaire in time T1.

Discriminate validity (the ability of the questionnaire to distinguish between those who suffer from disease and other populations) was tested by comparing the patients with the two control groups. The groups were then compared with analysis of variance (ANOVA) and Tukey-Kramer HSD test. Sensitivity (responsiveness) was assessed using the standardized mean response (SRM) coefficient.

Results of Cronbach's α were 0.852, 0.904, and 0.877 in T0, T1, and T2, respectively. Values approaching 1.0 indicate excellent internal consistency. Also, the value of test-retest reliability, expressed as Pearson's coefficient, of 0.86 indicates a high correlation between the two questionnaires completed by the same patient 3 weeks apart.

ANOVA and the Tukey-Kramer HSD test showed significant differences between patients with CRS and the two control groups (Table 1).

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Table 1 Comparison study of patients with two control groups for all questions (1-22)

ANOVA							
Question no.	p	Tukey-Kramer HSD test					
		Patients versus controls 1	Patients versus controls 2	Controls 1 versus controls 2			
1	< 0.0001	+	+	-			
2	< 0.0001	+	+	-			
3	< 0.0001	+	+	-			
4	< 0.0001	+	+	-			
5	< 0.0001	+	+	-			
6	< 0.0001	+	+	-			
7	< 0.0001	+	+	-			
8	< 0.0001	+	+	-			
9	0.0022	+	+	-			
10	0.0141	-	+	+			
11	0.1707	-	-	-			
12	< 0.0001	+	+	-			
13	0.0001	+	+	-			
14	< 0.0001	+	+	+			
15	< 0.0001	+	+	-			
16	0.0007	+	+	-			
17	0.0278	+	_	_			
18	0.0313	+	-	-			
19	0.2011	-	-	-			
20	0.0641	-	-	-			
21	0.4180	-	-	-			
22	0.1579	-	-	-			
TS	< 0.0001	+	+	_			

Means of the total score of the questionnaire for CRS patients, non-sinonasal patients, and healthy controls were 38.52, 13.68, and 10.22, respectively. In terms of sensitivity, expressed by the SRM coefficient, the questionnaire recorded a significant improvement in the postoperative QoL, which would be expected in a correctly compiled health-related QoL questionnaire. SRM coefficients for difference in total score between T0 and T1 and between T0 and T2 were 1.806 and 1.566, respectively. QoL is a standard part of the algorithms used to evaluate disease severity, treatment efficacy, or different treatment modalities. QoL assessment is a unique instrument, which is particularly important from the patient's point of view.

Of interest is the fact that QoL, as reported by patients, does not correlate with objective findings of other examinations. A weak, clinically insignificant association was demonstrated between SNOT-22 and the commonly used Lund-Mackay CT score [6].

Also no correlation was demonstrated between mucociliary clearance or eosinophilia and severity of symptoms of CRS [7, 8]. Hopkins et al. [3] in a level of evidence IIc study, investigated the effect of surgical treatment of CRS (with or without NP) on QoL using SNOT-22. In this study, 3128 patients with CRS were evaluated and the authors demonstrated significant improvement in the SNOT-22 score at 3, 12, and 36 months after surgery. Also, our patients showed statistically significant improvements in QoL at 3 and 6 months after surgery (expressed as a SRM coefficient) confirming a desirable level of sensitivity (responsiveness) for the Czech version of the questionnaire.

In our study, Cronbach's α for the total SNOT-22 score (TS) at T0 was 0.852. This result shows good internal consistency for the Czech version of the questionnaire, and approaches the results of the study by Hopkins et al. [9] (Cronbach's $\alpha = 0.91$). Also, the values of the test–retest coefficients are comparable (0.86 vs. 0.93) and confirm good reliability between repeated measurements.

The ability of the questionnaire to distinguish the disease-affected group was tested by comparison with healthy subjects and with a group of patients with non-sinonasal disease. Means of the total scores are comparable with the



TS total score

+ significant difference; - without significant difference English-validated version of SNOT-22 (38.5–40.8 in CRS patients and 10.2–9.3 in healthy controls) [9].

Besides the expected difference between patients with CRS and healthy individuals, we demonstrated the ability of the questionnaire to distinguish CRS patients from a group of patients with non-sinonasal disease. While it is clear that SNOT-22 is not intended to be a diagnostic instrument, this finding again demonstrates good internal consistency and balance between the disease specific and non-specific items in the questionnaire.

In conclusion, the study showed that the Czech version of SNOT-22 QoL questionnaire is a valid tool for assessing QoL of patients with CRS and the effectiveness of surgical treatment.

Conflict of interest statement None.

References

 Calman KC (1984) Quality of life in cancer patients: an hypothesis. J Med Ethics 10:124–127

- Lund VJ (2001) Health related quality of life in sinonasal disease. Rhinology 39:182–186
- Hopkins C, Browne JP, Slack R, Lund VJ, Topham J, Reeves B, Copley L, Brown P, van der Meulen J (2006) The national comparative audit of surgery for nasal polyposis and chronic rhinosinusitis. Clin Otolaryngol Allied Sci 31:390–399
- Piccirilo JF, Merritt MG, Richards ML (2002) Psychometric and clinimetric validity of the 20-item Sino-Nasal Outcome Test (SNOT-20). Otolaryngol Head Neck Surg 126:41–47
- Morley AD, Sharp HR (2006) A review of sinonasal outcome scoring systems: which is the best? Clin Otolaryngol 31:103–109
- Hopkins C, Browne JP, Slack R, Lund VJ, Brown P (2007) The Lund-Mackay staging system for chronic rhinosinusitis: how is it used and what does it predict? Otolaryngol Head Neck Surg 137:555-561
- Soler ZM, Sauer DA, Mace J, Smith TL (2009) Relationship between clinical measures and histopathologic findings in chronic rhinosinusitis. Otolaryngol Head Neck Surg 141:454

 461
- Boatsman JE, Calhoun KH, Ryan MW (2006) Relationship between rhinosinusitis symptoms and mucociliary clearance time. Otolaryngol Head Neck Surg 134:491

 –493
- Hopkins C, Gillett S, Slack R, Lund VJ, Browne JP (2009) Psychometric validity of 22-item sinonasal outcome test. Clin Otolaryngol 34:447–454