Osteomata of the Paranasal Sinuses: What Are the Limits of the Endoscopic Approach?

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KEYWORDS

- Osteoma • Osteomata • Draf type 3 procedure
- Endoscopic procedures

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Osteoma is a benign, slow-growing bone tumor consisting primarily of well-differentiated mature, compact, or cancellous bone. Osteoma is the most common benign tumor of the paranasal sinuses with a point prevalence of 3%, as demonstrated in 2 computed tomography (CT) radiological studies of 1500[1] and 1889[2] patients respectively.

AGE AND SEX

Osteomas occur more often in men, with a variable male-to-female ratio of 1.3:1.0[1] to 1.5:1.0.[2,3] Their peak incidence is between the fourth and sixth decades, with an average age at presentation of 50 years.[1,2]

LOCATION

Most osteomas (58%[1] to 68%[3]) involve the frontal sinus (37% arise in the immediate vicinity of the nasofrontal duct and 21% above and lateral to the frontal ostium).[1] The ethmoid sinus is the second most common area to be involved, whereas maxillary sinuses are affected in about 20% of cases, and sphenoid sinuses are rarely involved.[1] Osteomas can occur in conjunction with Gardner syndrome (familial adenomatous polyposis) (**Fig. 1**), an autosomal dominant condition consisting of multiple osteomas, soft tissue tumors (including skin cysts and desmoid tumors), and colon polyps with

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Fig. 1. A 51-year-old patient with Gardner syndrome. Note the multiple osteomata of the facial skeleton occurring in unusual locations, including the orbita, maxillary sinus, and zygomatic bone.

a high propensity toward malignant transformation.[4] As osteomas tend to appear an average of 17 years before the colon polyps, early gastroenterology referral is strongly advised.[5]

ETIOLOGY OF OSTEOMA

There are 3 main pathogenetic theories regarding the etiology of osteomas: developmental, traumatic, and infective.[6,7] According to the developmental theory, as proposed by Cohnheim,[7] osteomas arise from stem cells of the junctional area Q9 between the frontal and ethmoid bone. This is supported by the fact that osteomas frequently occur at the fontoethmoid suture line where the frontal sinus (membranous bone) borders the ethmoid labyrinth (endochondral ossification). However, this theory does not explain osteomas found in other locations. The traumatic theory, as proposed by Gerber, suggests that osteomas arise as an abnormal proliferative Q10 response to trauma and is supported by both the higher incidence of osteomas in men and the development of osteomas during puberty, when the rate of skeletal development is at its peak.[8] However, most osteomas are detected later in life and the great majority of patients do not report any history of trauma, whereas an increased incidence of osteomata in patients undergoing multiple endoscopic sinus surgery procedures has never been documented. Alternatively, it has been suggested that osteomas may arise as a result of infection stimulating osteoblasts within the mucoperiosteal lining of the sinus, which in turn may become secondarily calcified. Although there is an association between osteoma and sinusitis, the cause-and-effect relationship is not clear, and in up to 63% of cases, osteomas arise in healthy sinuses.[2] Other less substantiated theories suggest that osteomas may be osteodysplastic lesions, osteogenic hamartomas, embryonic bone rests, or the result of ossification of sinus polyps. However, none of these hypotheses have been proven.[4]

HISTOLOGY OF OSTEOMA

Macroscopically, osteomas are round or oval, hard, ivory-white, bosselated, well-circumscribed lesions attached to the underlying bone by a broad base or occasionally by a small stalk and covered by a thin layer of fibrous periosteum.[9] Histologically, osteomas can be classified into 3 types: ivory or compact, mature or cancellous, or spongiotic and mixed.[6,10] Ivory osteomas usually have a sessile base and are characterized by hard bone with a thick matrix containing only a small amount of fibrous tissue and minimal marrow. Cancellous osteomas often have a pedunculated base and are composed of cancellous bone with intertrabecular hematopoietic bone marrow or fat, whereas mixed osteomas share characteristics from both types (**Fig. 2**).[9,10]

GROWTH

In a study of 13 osteomas with serial radiographs, the average growth rate was 1.61 mm per year, ranging from 0.44 to 6.00 mm per year.[11] It has been shown that most osteomas recur infrequently even after incomplete removal.[12] However, given enough time, osteomas can recur,[13,14] and indeed accelerated regrowth following incomplete removal has been documented.[15] Malignant transformation of an osteoma has never been described, and osteomas should not be considered neoplastic lesions.[10]

CLINICAL CHARACTERISTICS OF OSTEOMA

Most osteomata are asymptomatic, slow-growing lesions diagnosed incidentally in imaging studies. Only 4%[1] to 10%[16] of all osteomas produce clinical symptoms, with osteomas of the frontoethmoidal region tending to be associated with earlier symptoms. Such symptoms are most commonly frontal pressure or headache,[17,18] either directly resulting from the lesion or indirectly from impaired drainage of the frontal sinus with or without concomitant chronic rhinosinusitis. The incidence of headache in various osteoma series varies between 52%[19] and 100% (**Table 1**).[17]

Complete obstruction of a sinus ostium by an osteoma may lead to secondary formation of mucocele.[25,26] When an osteoma extends beyond the confines of the sinuses, it may produce an external deformity (**Fig. 3**).[27] Orbital extension may lead to proptosis and periorbital pain, as well as chemosis and diplopia if the oculomotor muscles are affected[28–30] or epiphora if the nasolacrimal duct is compressed (**Fig. 4**)[31,32] and rarely decreased visual acuity in cases of optic nerve compression.[33,34] Intracranial extension of the lesion can lead to intracranial mucocele with meningitis, cerebral abscess,[35–37] or even tension pneumocephalus (**Fig. 5**).[38] In our experience, headache is the sole presenting symptom of osteomas in the vast majority of cases, whereas the slow growth of an osteoma usually precludes eye symptoms, even in cases of significant orbital extension, unless a concomitant mucocele is present.

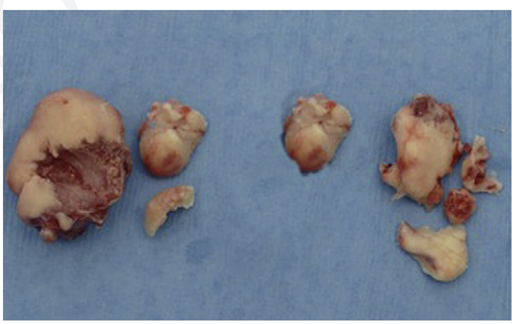

Fig. 2. Fragments of a mixed osteoma removed via an external osteoplastic flap approach. Note the thin mucosal layer overlying the osteoma.

Table 1
Osteomata case series

Study, Year, Journal	Cases	Presenting Symptoms	Location	Tumor Grade	Procedure	Outcome	Complications	Follow-Up (Months)
Brodish et al,[20] 1999, Am J Rhinol	9	Headache	9 frontoethmoidal	nr	9 end	0	2 CSF leaks	40
Schick et al,[17] 2001, Rhinology	34	Headache	23 frontal sinus 11 ethmoid	nr	23 end 11 open	3 residuals (end)	0	1–32
Chiu et al,[21] 2005, Am J Rhinol	9	Headache 88% Sinusitis 66%	9 frontal sinus	I: 1 II: 2 III: 4 IV: 2	3 end 5 combined 1 open	nr	0	7.4
Dubin and Kuhn,[22] 2006, Otolaryngol Head Neck Surg	12	Headache: 100%	12 frontal sinus	I: 3 III: 8 IV: 1	8 end 4 combined	2 residuals (open) 1 residuals (end)	1 frontal stenosis (open)	19.2
Bignami et al,[23] 2007, Rhinology	26	Headache: 63%: Nasal obstr: 38%	26 frontal sinus	nr	11 end 13 combined 2 open	0 recurrences	0	40
Castelnuovo et al,[19] 2008, J Craniofac Surg	48	Headache: 52%:	18 frontal sinus 13 frontoethmoid 9 ethmoid 8 other	nr	22 end 26 open	nr	0	53 (end) 35 (open)
Seiberling et al,[18] 2009, Am J Rhinol Allergy	23	Headache 62.5% Sinusitis 56.5%	18 frontal sinus 5 frontal recess	I: 5 II: 4 III: 6 IV: 8	2 combined 21 end	4 residuals	1 frontal stenosis (end)	33
Ledderose et al,[24] 2010, Eur Arch Otorhinolaryngol	24	Headache 83% Sinusitis 87%	7 frontal sinus 7 frontal recess	I: 3 II: 5 III: 10 IV: 6	12 combined 8 end 4 open	95% satisfied[a] 1 pain increase	1 bleeding (combined) 1 bleeding (open)	nr

Abbreviations: CSF, cerebrospinal fluid; nr, ■.
[a] SNOT 20 questionnaire.

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Q20
Q21

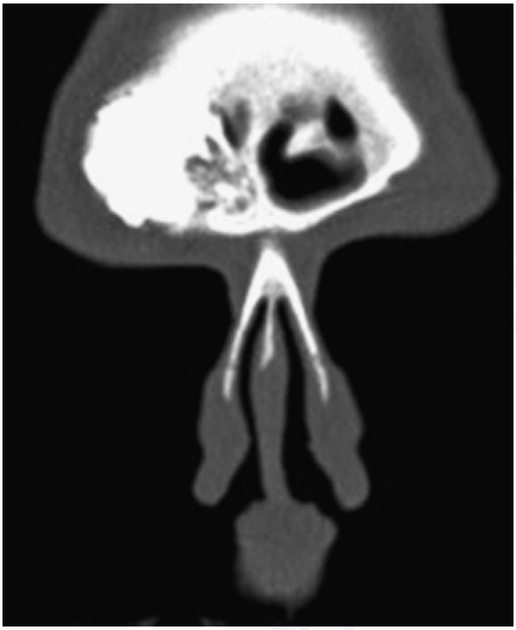

Fig. 3. Osteoma extending through the anterior frontal plate and associated with facial deformity.

IMAGING

Although osteomata can be seen in simple sinus radiographs, the imaging modality of Q11 choice is thin-slice CT. This allows the precise estimation of the size and the location of the osteoma, as well as concurrent sinus pathology. Osteomata appear as well-circumscribed masses of heterogeneous consistency on CT, with hyperostotic (high signal) and spongiotic (lower signal) components (**Fig. 6**). The lower signal components may be confused with associated mucoceles. In such patients, magnetic

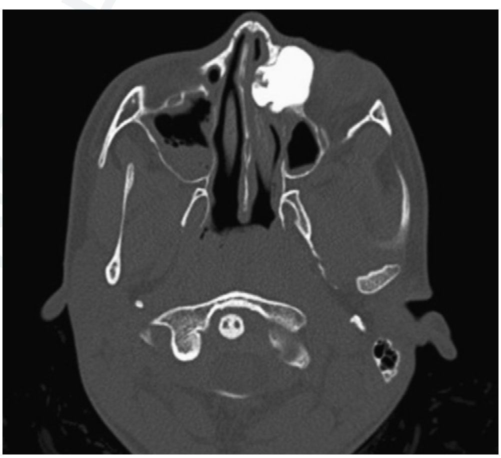

Fig. 4. A patient referred by the ophthalmologist where he attended with epiphora. Note the osteoma obstructing the nasolacrimal duct.

Fig. 5. Large osteoma of the frontal sinus in a patient presenting with headache. There was significant intracranial extension but the dura was intact and the patient had no neurologic complications.

resonance imaging is useful to assess the extent of the tumor as well as the presence of complications (mucoceles, orbital or intracranial extension).

INDICATIONS

Although it is generally agreed that symptomatic osteomas (unless there are serious contraindications) should be surgically excised, management of asymptomatic osteomata is controversial. In the case of small, uncomplicated osteomata, watchful

Fig. 6. Frontal sinus osteoma. Note the heterogeneous appearance on CT.

waiting with interval radiologic imaging is usually advised. Savić and Djerić[39] recommend surgical removal of enlarging frontal sinus osteomas, those extending beyond the boundaries of the sinus, localized adjacent to the nasofrontal duct, associated with chronic sinusitis, or in patients complaining of headaches when all other causes have been excluded, as well as osteomas in the ethmoid sinuses, irrespective of their size. Smith and Calcaterra recommend surgery if the osteoma occupies more than 50% of the frontal sinus.[40] Our policy is to treat the following:

- Osteomas associated with symptoms (usually headache) after all other explanations for the symptoms have been excluded
- Large (extending to more than 50% of the frontal sinus) or growing osteomas, as seen on serial CTs
- Osteomas associated with current (mucocele, orbital symptoms, neurologic symptoms, external deformity), imminent (complete obstruction of the frontal recess, intraorbital or intracranial extension) complications

We do not operate small ethmoid osteomas, which, more often than not, are incidental CT findings with no clinical significance.

Lynch Procedure

One of the first methods used to treat symptomatic frontal or frontoethmoid osteomas was the external frontoethmoidectomy approach (Lynch procedure).[22] This has been used for small, medially or inferiorly situated tumors. However, it can lead to an unsightly scar, does not provide adequate access laterally, and has a high rate of frontal recess stenosis.[41]

Osteoplastic Flap Procedures

The osteoplastic approach, as popularized by Goodale and Montgomery,[42] has been the most widely used technique for frontal sinus osteomas. It provides excellent visualization and wide access to the frontal sinus, including its superior, posterior, and lateral aspects, although the nasofrontal duct and ethmoids may not always be adequately visualized. The osteoplastic flap procedure is well established, being in use for more than 40 years, and is technically accessible to most otolaryngologists. Nevertheless, it is an invasive procedure, with significant morbidity, including blood loss, impaired cosmesis, postoperative frontal pain, paresthesia, or anesthesia from supraorbital nerve damage and (rarely) in the case of intracranial entry, potentially devastating complications including cerebrospinal fluid (CSF) leak and meningitis . If the frontal sinus is obliterated, then the added morbidity of an abdominal incision for fat harvesting is introduced, as well as the risk of late mucocele formation, which can be as high as 9% after 2 years.[43]

Endoscopic Procedures

Endoscopic approaches to the nose and paranasal sinuses were introduced in the 1980s, and by the early 1990s the first cases of endoscopic management of ethmoid osteoma were published.[44,45] The accumulation of experience with endoscopic sinus surgery, technological advances, including the development of dedicated instruments (malleable forceps; 40-degree, 55-degree, and 70-degree curved diamond and cutting drills; straight high-speed neurolosurgical drills; and dedicated bipolar intranasal diathermy forceps), improved endoscopes, and the introduction of CT navigation, expanded the limits of endoscopic approaches. On the other hand, the work of Draf, in systematizing the approaches to the frontal sinus,[46] laid the foundations of Q12 modern endoscopic frontal sinus surgery. Importantly, he described the type 3 ("Draf 3") procedure (endoscopic modified lothrop,[47] bilateral frontal sinus drillout,[48] median drainage procedure[49]) as a way to establish the widest possible transnasal access to the frontal sinus.

WHAT ARE THE LIMITS OF THE ENDOSCOPIC APPROACH?

As with most surgical techniques, Level 1 or 2 evidence is missing; however, Level 3 evidence can be collected using case series and retrospective cohorts. The evolution of these indications testifies to the progress affected in endoscopic surgery over the past decades.

Ethmoid Sinus

Endoscopic approaches to an ethmoid osteoma are relatively straightforward. The involvement of the cribriform plate is not a contraindication, as gentle drilling using a diamond burr until the osteoma is paper thin can help to remove the osteoma. Even extensive involvement of the orbit can usually be dealt with endoscopically; the limit being the anterior extension. Extension anteriorly to the nasolacrimal duct and under the skin usually requires a combined endoscopic/external (transconjunctival) approach in this case (see **Fig. 2**).

Frontal Sinus

Draf, in his seminal paper on the Fulda concept in 1991, suggested that any "large osteoma" was not amenable to an endoscopic approach and should be dealt with via an osteoplastic flap approach.[50]

Since then, 8 case series, including at least 5 osteomata each, have been published (see **Table 1**).

Brodish and colleagues[20] presented in 1999 a series of 8 osteomata treated endo- Q13 scopically. They were removed with osteotomes and curettes and there were 2 incidences of (anticipated) CSF leaks. No specific indications were described for the endoscopic approach.

The first large series of sinonasal osteomata treated endoscopically was published by Schick and colleagues[17] in 2001. They suggested, on the basis of 35 patients, that exclusion criteria for an endoscopic approach included

1. intracranial extension
2. large intraorbital involvement
3. anteroposterior diameter of the frontal sinus smaller than 10 mm
4. lateral extension over a virtual plane through the lamina papyracea
5. erosion of the posterior or anterior wall of the frontal sinus

However, the first systematic attempt to codify the limits of endoscopic resection was by Chiu and Kennedy in 2005.[21] Drawing from their experience with 9 osteomas Q14 between 1999 and 2003, they developed a grading system (**Table 2**) maintaining that only grades 1 and 2 osteomata can be removed endoscopically.

Essentially, their grading suggests that the 3 contraindications for endonasal removal of an osteoma are the following:

1. base of attachment anteriorly or superiorly within the frontal sinus
2. extension laterally to a virtual sagittal plane through the lamina papyracea
3. complete obliteration of entire frontal sinus

Table 2
Frontal sinus osteoma grading system

Grade I	Base of attachment is posterior–inferior along the frontal recess. Tumor is medial to a virtual sagittal plane through the lamina papyracea. Anterior–posterior diameter of the lesion is *less* than 75% of the anterior–posterior dimension of the frontal recess.
Grade II	Base of attachment is posterior–inferior along the frontal recess. Tumor is medial to a virtual sagittal plane through the lamina papyracea. Anterior–posterior diameter of the lesion is *greater* than 75% of the anterior–posterior dimension of the frontal recess.
Grade III	Base of attachment is anterior or superiorly located within the frontal sinus AND/OR tumor extends lateral to a virtual sagittal plane through the lamina papyracea.
Grade IV	Tumor fills the entire frontal sinus

Data from Chiu AG, Schipor I, Cohen NA, et al. Surgical decisions in the management of frontal sinus osteomas. Am J Rhinol 2005;19(2):191–7.

Castelnuovo and colleagues,[19] on the basis of 33 osteomata, suggested that an endoscopic approach was contraindicated in cases of

1. ateral extension to the sagittal plane passing through the lamina papyracea
2. intracranial extension
3. involvement of the posterior and anterior wall of the frontal sinus
4. anteroposterior frontal sinus diameter smaller than 1 cm

In 2007, Bignami and colleagues,[23] on the basis of 25 osteomata, supported Chiu/Kennedy's grading system and criteria for endoscopic removal. They stated that an endoscopic approach was not feasible in cases with

1. intracranial extension
2. large orbital involvement
3. anteroposterior diameter of the frontal sinus smaller than 10 mm
4. lateral extension behind a virtual plane through the lamina papyracea
5. erosion of the posterior or anterior wall of the frontal sinus

Endoscopic surgery has been evolving at a very fast pace and a number of surgeons have challenged these assumptions. Just a year after the publication of the Chiu/Kennedy classification, Dubin and Kuhn[22] published their results of successful endoscopic removal of 5 grade III tumors attached either superior-anteriorly in the frontal sinus or extending lateral to the plane of lamina papyracea. In this article, an osteoplastic flap was recommended only for removal of tumors with more than 2 cm of vertical extension into the frontal sinus or occupancy of 100% of the frontal sinus.

In 2009, Seiberling and colleagues[18] reported their results of 23 patients with varying sizes of frontal sinus osteomas treated endoscopically, which included 8 patients with a grade IV tumor and 6 patients with a grade III tumor. A Draf 3 procedure was used for 15 of these tumors (including all grade III and IV tumors). In 4 of 8 grade IV (filling the entire frontal sinus) tumors, a residual was left toward the posterior frontal plate, as it was felt that the risk of penetrating the dura was too high. In 2 cases, a second procedure was necessary for the complete removal of the tumor, whereas in one patient with extensive orbital extension, an external blepharoplasty incision was used and an extended trephine incision was used in another patient.

In 2010, Ledderose and colleagues[24] proposed that, in carefully selected individual cases, it is possible to remove grade III and even grade IV osteomas endonasally. They described the endoscopic removal of 8 osteomas, 3 of which would have been classified as nonresectable endoscopically according to the Chiu/Kennedy classification: specifically, 2 grade III tumors were removed via a Draf 2b approach and a grade IV tumor was removed via a Draf 3 approach.

What we know now is that, although there is no number of external approaches that can prove the limits of endoscopic surgery, a small number of endoscopic approaches (replicated in more than one center) can shatter the myth of "unresectability." We believe that it is not the anteroposterior diameter or the lateral extension of the osteoma that defines its resectability endoscopically, but rather the relation between the interorbital distance, the anteroposterior diameter of the frontal beak, and the lateral height of the frontal sinus. We have attempted to codify our experience with the endoscopic approach to osteomata as follows (Grade C recommendations):

1. Lateral extent
2. Large tumors attached to the posterior/superior frontal walls/more than 2 cm superiorly in the frontal sinus
3. Orbital extension
4. Intracranial extension
5. Anterior extension

Lateral extent

Using the wide access provided by a Draf 3 procedure and curved drills, it is possible to access the lateral supraorbital ridge well beyond the medial orbit. We maintain that it is not the plane of lamina papyracea or the 2 cm lateral to it that define the lateral limits of respectability, but rather the ratio of lateral tumor extension to *interorbital distance.* Following the removal of the superior septum and the drilling of the nasal beak, lateral access to the frontal sinus is restricted primarily by the orbital walls. In patients with relatively large intercanthal distance, the lateral access that can be gained is increased, whereas the opposite is true for narrow nasal inlet (**Fig. 7**). Lateral access to the floor of the frontal sinus (orbital roof) may, however, be limited, as a recent study[51] confirmed.

Large tumors attached to the posterior/superior frontal walls/more than 2 cm superiorly in the frontal sinus

Similarly, tumors extending superiorly, to the posterior frontal plate, or associated with complete opacification of the frontal sinus can also be removed endoscopically (**Figs. 8** and **9**).

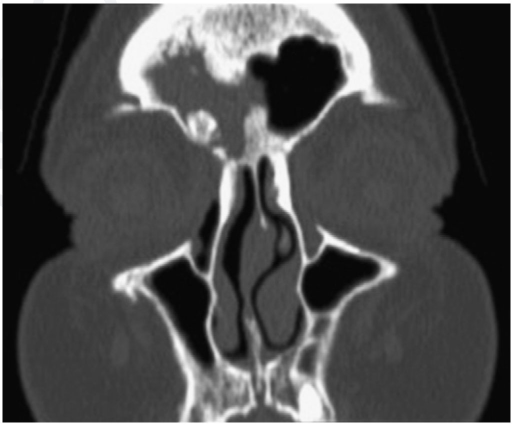

Fig. 7. Osteoma lateral to lamina papyracea removed endoscopically.

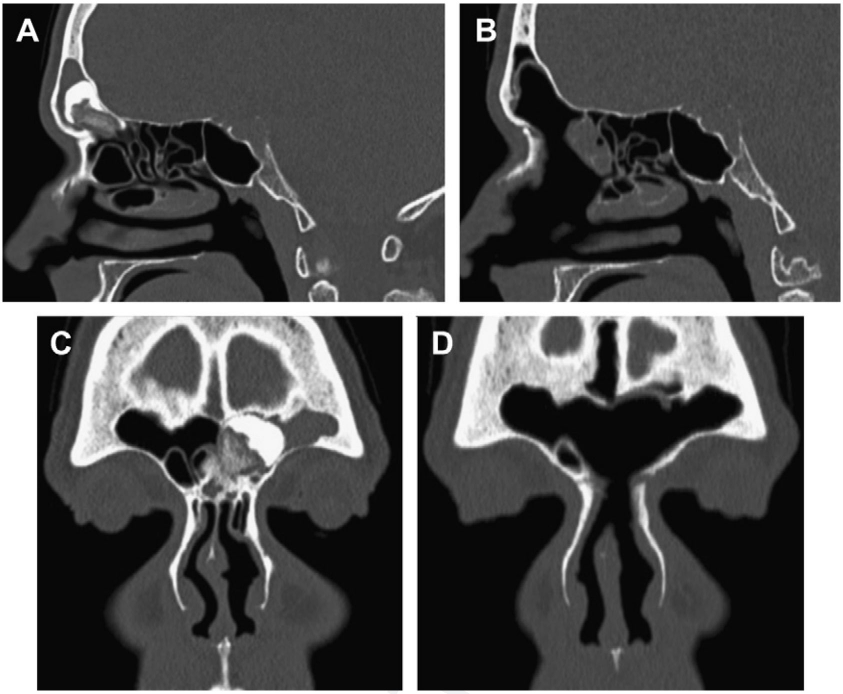

Fig. 8. (*A–D*) Preoperative and postoperative CT scans of a large osteoma attached to the posterior frontal sinus wall, extending more than 2 cm superiorly and completely obstructing the frontal sinus removed endoscopically.

In many cases, we saw that the approach of such tumors was time consuming, as the curved drills operating at 10,000 rpm (as opposed to the 80,000-rpm straight drills) would frequently fail and had to be changed. In one such case, our approach was staged, and the osteoma was removed completely in the second approach, and

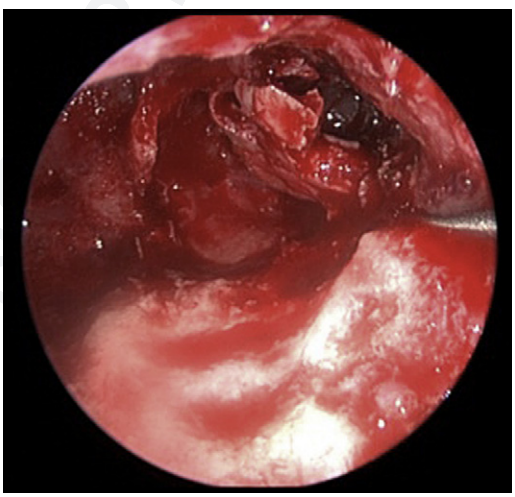

Fig. 9. Thinning out of the posterior attachment of the osteoma and removal with a curette: view through a Draf 3 procedure.

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Table 3
Evolution of contraindications of endoscopic approach

Anatomic Limitations	Schick	Chiu	Dubin	Bignami	Castelnuovo	Sieberling	Ledderose	AMC
Attachment anterior frontal plate		YES			YES			YES (when associated with large defect or very high attachment)
Attachment posterior frontal plate					YES	NO (may need to leave remnant)		NO
Attachment superior frontal sinus		YES				NO	NO	NO
Less than 1 cm frontal sinus diameter	YES			YES	YES			Relative
Extension more than 2 cm superiorly in frontal sinus			YES			NO	NO	NO
Lateral to lamina papyracea sagittal plane	YES			YES	YES	NO	NO	NO
2 cm lateral to orbit						NO	NO	NO
Erosion of anterior table	YES			YES	YES			YES
Complete obstruction of frontal recess			YES			NO	NO	NO
Complete opacification of frontal sinus		YES				NO		NO
Intracranial extension/erosion of posterior table	YES			YES	YES			NO
Extension anterior to nasolacrimal duct								YES
(Significant) orbital extension	YES			YES		NO (may require additional incision)		NO

with the use of a (much more effective) 80,000-rpm straight drill. The development in the future of high-speed curved drills may further facilitate the removal of such large laterally located osteomas.

Orbital extension

Orbital extension is not in itself a contraindication for an endonasal approach (see **Fig. 4**). However, as stated by others,[18] additional incisions may be required if the tumor extends *anteriorly*. We found that anterior extension (anteriorly to the nasolacrimal duct), rather than in the orbit per se, is an indication for an external incision. In most cases, the external approach can be performed via a subconjunctival incision, with no cosmetic consequences.

Intracranial extension

We maintain that limited endocranial extension does not always preclude the use of the endoscope. As we progress to manage intracranial/intradural tumors endoscopically, the limitation of posterior wall erosion/endocranial extension sounds irrelevant, with the proviso that the removal is done in combination with a endoscopically trained neurosurgeon.

Anterior extension

The one limitation to endonasal approaches that seems to withstand the test of time is anterior extension. Extension of the tumor through the anterior frontal plate is usually physically impossible to access endoscopically, whereas the associated bony defect and deformity necessitates an external approach for reconstruction (see **Fig. 3**).

The evolution of contraindications for the endoscopic approach is presented in **Table 3**.

SUMMARY

Advantages of the endoscopic approach include better close-up and 3-dimensional visualization of anatomic structures, absence of scars, smaller traumatic impact along the approach path, reduction of postoperative morbidity, preservation of the physiologic mucociliary drainage, less bleeding, and a shorter hospital stay. However, the endoscopic approach can make the management of potential intraoperative complications (massive bleeding, intracranial complications, CSF leak) more difficult and requires significant time commitment (for large osteomata, significantly more than an external approach) and highly sophisticated surgical tools.

We do not believe that the endonasal removal of osteomas is a procedure that should be undertaken lightly. Significant experience in all frontal sinus approaches, including Draf type 3 sinusotomy, is required, together with great facility in the use of the drill endonasally. Although temporal bone drilling is part of the curriculum in most residency programs, the development of similar skills for drilling in the anterior skull base is not required and is rarely acquired during training. As endoscopic sinus surgery comes of age, we expect that the skills required will be more widely shared. A new generation of surgeons will be moving forward the frontiers of endoscopic surgery, and we expect that what today are the "frontiers" of endonasal surgery will be standard procedures tomorrow.

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