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Technical note

# Reconstruction of the anterior wall of the frontal sinus by a custom-made titanium prosthesis after resection of a giant osteoma of the frontal sinus

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## ABSTRACT

Osteoma is a benign, usually asymptomatic bone tumour, frequently arising in the nose and paranasal sinuses. Surgical treatment is required when the patient becomes symptomatic or presents ophthalmological or neurological complications. Although an endoscopic approach is increasingly used, depending on the size and site of the osteoma, open surgery may be preferable and remains the standard treatment. This technical note describes a case of giant osteoma of the frontal sinus that required a bicoronal approach with reconstruction by a custom-made titanium prosthesis.

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## 1. Introduction

Osteoma is a benign bone tumour and represents the most common benign tumour of the paranasal sinuses, mainly arising in the frontal sinus. Osteoma, usually asymptomatic, occurs after the age of 20 to 30 years with a male predominance. The aetiology of osteoma remains unknown, although traumatic, infectious or embryological aetiologies have been proposed.

An osteoma is considered to be a giant osteoma when it measures more than 3 cm in diameter [1]. Osteomas must be treated surgically when they are symptomatic or in the presence of ophthalmological and/or neurological complications. Various surgical approaches are used, depending on the site and size of the osteoma and the surgeon's usual practice: open surgery, endonasal surgery or a mixed approach. Reconstruction may be necessary when resection of the tumour leaves a large defect. Various materials can be used: autologous bone graft, titanium mesh, Porex® Porous plastics [2], etc.

The purpose of this article is to report the surgical approach adopted by our team for frontal sinus reconstruction after resection of a giant osteoma of the frontal sinus, a rare disease, as only 34 cases have been reported in the literature from 1951 to 2017.

## 2. Technique

Mr. N., 45 years old, with no notable history, attended the emergency department of our institution in 2012 for right orbital cellulitis complicated by subperiosteal abscess, with a favourable outcome after open surgical drainage (medial canthus incision) and antibiotics.

Paranasal sinus CT scan showed a very large, calcified, multi-lobed lesion arising from the anterior wall of the right frontal sinus with extension to the superomedial part of the orbit, in contact with the right superior rectus muscle, with no posterior wall involvement, measuring 5 × 3.7 cm. (Fig. 1).

This lesion was considered to be a probable giant osteoma of the right frontal sinus with orbital extension, responsible for acute frontal sinusitis complicated by orbital cellulitis. MRI confirmed the absence of cerebral involvement and ophthalmological examination did not reveal any abnormality.

The patient was subsequently lost to follow-up, but returned to the emergency department in 2016 with another episode of right orbital cellulitis, with a favourable outcome in response to empirical intravenous antibiotics. He had not experienced any other episodes of infection between 2012 and 2016.

At the follow-up examination one month after this last episode, the patient presented minor right exophthalmos with palpebral oedema. Ophthalmological examination demonstrated preserved visual acuity, no papilloedema on fundoscopy, and Lancaster's test showed a deficit of upward gaze in adduction. Another imaging assessment showed a stable appearance of the probable osteoma.

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Fig. 1. CT scan, axial section, bone window setting.

Surgery was then considered to be indicated due to the recurrent infection and ophthalmological involvement and was performed three months after the second infectious episode.

The bone tumour of the anterior wall of the right frontal sinus was resected via a bicoronal incision. Precise osteotomies were performed by using a custom-made bone cutting guide designed in each plane of section on the preoperative CT scan by Materialise®, in order to avoid meningeal effraction. The anterior wall of the right frontal sinus was removed en bloc with the osteoma and its implantation base and resection was completed by curettage of the rest of the cavity. As the frontal sinus mucosa was destroyed during resection, a galea flap, maintained with fibrin sealant, was used to reline the cavity. Reconstruction of the defect of the anterior wall of the right frontal sinus and superomedial margin of the right orbit was performed during the same procedure by a custom-made porous titanium prosthesis manufactured by Materialise® from the mirror image of the CT scan of the contralateral sinus. (Figs. 2 and 3).

The postoperative course was uneventful. Histological examination of the operative specimen confirmed the diagnosis of benign osteoma. Follow-up CT scan at the fourth postoperative month showed no signs of recurrence or mucocele, with correct positioning of the prosthesis. At the 10-month postoperative follow-up visit, the scar was cosmetically satisfactory despite the patient's baldness. He did not present any frontal palsy, or levator palpebrae superioris deficit.

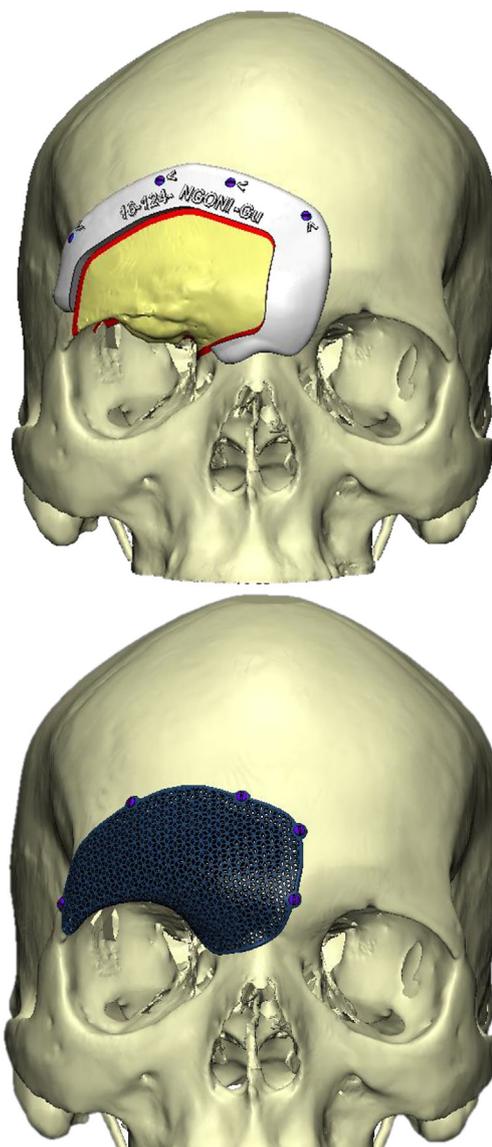


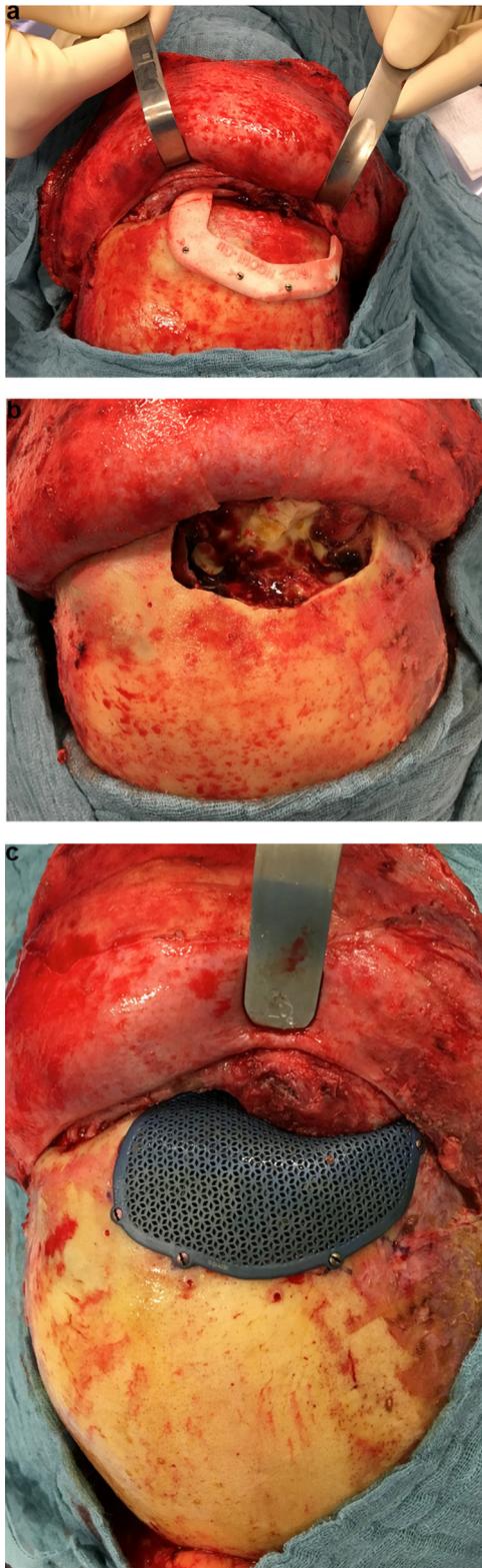
Fig. 2. 3D reconstruction, showing the bone cutting guide and the prosthesis.

### 3. Discussion

Osteoma, a benign osteoblastic tumour, is the most common benign tumour of the paranasal sinuses, mainly involving the frontal sinus, followed by the ethmoid sinus. It represents 1% of all bone tumours and 11% of all benign bone tumours [3]. These usually asymptomatic, slow growing, rounded, often irregular, sessile tumours arise from the subperiosteum or endosteum. The real prevalence of osteoma is difficult to estimate, but ranges from 0.1 to 3% in the general population [1]. Osteoma is a disease of young people, from the ages of 20 to 30 years, with a marked male predominance: sex ratio 2:1/3:1 [3].

The aetiology of osteoma remains unknown, but the various hypothesis include trauma during puberty, infection related to chronic sinusitis, or embryological causes (frontoethmoidal suture: junction between the embryonic ethmoid cartilage and the membranous frontal bone) [4]. Osteomas are considered to be giant osteomas when they measure more than 3 cm in diameter [4].

Patients are usually asymptomatic, but may subsequently present various signs due to local extension of the frontal osteoma: headache, recurrent sinusitis, mucocele. More rarely, osteoma may present at the stage of orbital complications: exophthalmos, ptosis,



**Fig. 3.** Intraoperative views. a: bicoronal approach; galea flap, bone cutting guide in place; b: view after en bloc resection of the anterior wall and the tumor; c: insertion of the titanium prosthesis.

loss of visual acuity, diplopia or even neurological complications, such as meningitis, brain abscess, pneumocephalus, cerebrospinal fluid leak, intracranial hypertension [5].

Osteomas may be part of Gardner's syndrome, an autosomal dominant genetic disease, comprising intestinal polyps, epidermal

cysts, subcutaneous desmoid tumours, fibromatosis, and multiple osteomas in the mandible, skull, and long bones.

Differential diagnoses in the paranasal sinuses include: periosteal osteosarcoma, osteoblastoma and osteochondroma [5]. The pathognomonic sign of osteochondroma is an excrescence of cortical bone and bone marrow continuous with the underlying healthy bone. On imaging, osteoblastoma and osteosarcoma are heterogeneous osteolytic lesions, and osteosarcoma is associated with an intense periosteal reaction.

Osteomas are treated surgically according to well-defined indications: symptomatic patient, or the presence of ophthalmological or neurological complications and/or active osteoma (growing by more than 1 mm/year) involving more than 50% of the sinus volume and/or presenting extension to the frontal recess or sphenoid sinus (risk of compression of the optic canal) [5].

Various surgical approaches are used, depending on the site and size of the osteoma and the surgeon's usual practice. An endonasal approach, increasingly used since the advent of endoscopic surgery, can be used for ethmoidal osteomas, but is technically more difficult for frontal osteomas due to problems of accessibility, especially in the presence of lateral extension. Open surgery remains the standard technique for frontal osteomas.

The first frontal bone flap was performed in Europe in 1894 [6]. Open surgery for resection of the frontal sinus can be performed via several incisions, such as the Lynch-Howarth incision or bicoronal incisions.

The Lynch-Howarth incision is essentially used for osteomas involving the anterior wall of the frontal sinus. The bilateral eyebrow incision is easy to perform and provides wide exposure, especially in patients with a large frontal sinus. The disadvantages of this technique are the visible facial scar and a risk of supraorbital nerve (V2) injury.

The bicoronal incision, designed for the resection of larger tumours involving the lateral or anterior wall of the frontal sinus, allows optimal exposure and leaves a barely visible scar in the patient's hairline. This approach also allows preservation of the supraorbital nerve and harvesting of a temporalis fascia or cranial bone graft during the same procedure. Its drawbacks are greater blood loss and a risk of frontal palsy (but the frontal branch of the facial nerve can be easily avoided by passing above the temporalis fascia). This approach is contraindicated in the presence of osteomyelitis of the frontal bone or a history of frontal surgery [3]. The bicoronal incision allows repair of the bone defect created during resection by the bone of the bone flap, titanium mesh, titanium prosthesis, etc.

Open surgery via a bicoronal incision was chosen in this case, as an endonasal or mixed approach was not indicated because of the size of the tumour, involving all of the frontal sinus, corresponding to Grade 4 of Chui's classification [7], and, due to the presence of the implantation base on the anterior wall of the right frontal sinus, it was decided that en bloc resection of the osteoma together with its implantation base would be preferable to avoid recurrence [8]. Other types of implantation base can be progressively resected by drilling. Reconstruction of the anterior wall of the right frontal sinus and superomedial margin of the orbit was required due to the inevitable bone defect resulting from resection.

Other reconstruction options include the use of autologous cranial bone graft, which remains standard practice at the present time, or the use of a titanium mesh conformed in situ, as titanium is a stable, biocompatible material [9]. An article published in 2016 reported the use of Porex® Porous plastics for reconstruction of a similar case with a satisfactory result [2].

We preferred to use a custom-made bone cutting guide and titanium prosthesis, based on collaboration between the Materialise® engineer and the surgeon using the patient's CT data. This technique allowed us to ensure safe section of the anterior wall of

the sinus, while avoiding meningeal effraction and reducing the reconstruction operating time of the large defect extending to the orbit.

Incomplete resection is associated with a risk of postoperative recurrence of osteoma [1]. This risk can be considered to be minimal in the present case as a result of optimal resection of the osteoma implantation base [6].

The use of this technique for reconstruction of a giant osteoma of the frontal sinus has never been previously reported in the literature. The time required for manufacture of the cutting guide and prosthesis was compatible with a non-urgent elective procedure. The cost of acquisition of the material was compensated by the gain in operating room occupation time.

#### 4. Conclusion

Reconstruction of a giant osteoma of the frontal sinus by a custom-made titanium prosthesis was cosmetically and functionally satisfactory at medium-term follow-up. The use of a bone-cutting guide allowed a reduction of operating time and surgical risks. This technique, not previously reported in the literature for reconstruction of a giant osteoma of the frontal sinus, was effective and financially acceptable.

#### Disclosure of interest

The authors declare that they have no competing interest.

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