



## Case Report

# Long-term persistent discomfort due to a giant frontoethmoidal osteoma despite complete surgical removal - A case report

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## ABSTRACT

**Introduction:** Giant frontoethmoidal osteomas are rare, slow-growing, benign osseous tumours, frequently causing severe life impairing symptoms due to their proximity to noble structures. Initially, osteomas are often diagnosed on radiographs by chance. Their aetiology can be considered ambiguous. They may either be treated by active observation, medical therapy, radio and thermal therapy, or surgery.

**Case presentation:** We report the case of a 56-year-old female patient with a giant osteoma spreading from the nasal cavity to the entire frontoethmoidal sinus, leading to headaches, respiratory problems, and nausea for several years. For a period of 20 years, a watch and wait approach was applied. Finally, the osteoma was removed using a combined open and endoscopic approach. One year after the operation, a secondary mucocele developed, accompanying headaches and facial pressure due to its continuous expansion. Despite numerous consultations, she refused surgical intervention until today.

**Discussion:** Early detection and removal of frontoethmoidal osteomas improves the prognosis for a favourable treatment outcome. The smaller the osteoma, the easier it can be removed endoscopically. The decision to perform surgery was made when the condition drastically affected the patient's quality of life. To date, there is still no strong consent regarding the best surgical approach and the best time to do it.

**Conclusion:** The combination of open and endoscopic surgery remains a safe and straightforward procedure for the removal of giant frontoethmoidal osteomas. Early detection and intervention are crucial for a predictable minimally invasive treatment with a favourable outcome for the patient.

## 1. Introduction

Osteomas are benign osteogenic tumours, primarily unilateral, but also bilateral in less than 10% of cases [1]. Depending on their location, osteomas can be divided into peripheral, central, and extra-skeletal types. The peripheral type develops from the periosteum, the central type from the endosteum, and the extra-skeletal type from soft tissue [2]. Osteomas can be classified in accordance with their location and are frequently discovered in the skull, located mainly in the frontal or ethmoid sinus [3], with involvement of the maxillary sinus in less than 2% of cases [2]. The size of an osteoma is usually between 2 and 30 mm. Any osteoma larger in size is rare and referred to as a large or giant

osteoma [1].

They can occur at any age but tend to be more common in the second to fifth decade [4]. Some studies claim no gender difference in their prevalence, whereas others indicate a slight predominance in men [5]. Their aetiology can be considered ambiguous [6]. Its presumed causes include embryologic development (e.g. Gardner's syndrome), trauma, and infection [6,7]. Frequently detected accidentally in the early phase, osteomas may cause symptoms such as proptosis, facial pain, headache, and infection due to obstruction of the nasofrontal duct [6]. Other symptoms include local pain due to a growing facial deformity, nasal obstruction causing breathing difficulties, and headache. The eyes may also be affected, leading to reduced visual acuity and diplopia.

**Abbreviations:** Computed tomography, CT; ear nose throat, ENT; panoramic radiograph, OPG; non-steroidal anti-inflammatory drug, NSAID.

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Inflammations such as sinusitis, mucocele, pneumocephalus or abscesses may occur [6]. The vast majority of osteomas are diagnosed at the onset of initial symptoms. As the entities grow slowly, it takes time for early impairing symptoms such as breathing difficulties to develop into a life-threatening condition. The symptoms of the tumour depend on its location, typically severe breathing difficulties and headache when the tumour grew within parts of the nasal cavity and the frontal sinus [6]. A computed tomography (CT) and a panoramic radiograph (OPG) are essential investigations to diagnose an osteoma and differentiate it from other diseases such as osteosarcoma [8]. Another differential diagnosis is Paget's disease, which is characterised by high bone turnover rates [9]. The odontoma, a tumour originating from the teeth, must also be excluded [10]. The ossifying fibroma, a slow-growing tumour of fibrous tissue, may lead to a false diagnosis of osteoma [11]. Radiologically, an osteoma is seen as a radiopaque mass with clearly defined margins. Sometimes it is incorporated into the cortical portion of bone, which gives rise to a mushroom-like entity on the radiological image [12]. Depending on the size and location of the osteoma, it may be treated by one of the following four methods: active observation, medical therapy, radio and thermal therapy, and surgery [3]. Surgical treatment is recommended when the osteoma affects the bone, causes destruction of the nasal cavity, or dyspnoea [1]. The development of a secondary mucocele, which is generally known as a common benign condition after sinus obstruction [13], is documented in up to 50% of cases [4].

This case report is a long-term clinical and radiological documentation of a giant osteoma spreading bilaterally from the nasal cavity to the entire frontoethmoidal sinus, from initial symptoms to a 10-year follow-up. We present this case report in accordance with the SCARE criteria [14].

## 2. Case presentation

We report the case of a 56-year-old Caucasian woman with a giant osteoma of the frontoethmoidal sinus, diagnosed 20 years prior to surgery. As a result, the patient had been suffering from progressively worsening headaches, respiratory problems, and nausea. In 2007, she presented herself to the department of oral and maxillofacial surgery at the University Hospital of St. Pölten, Austria, describing the symptoms as mentioned earlier. She had unremarkable family and psychosocial

histories and was a non-smoker. The clinical examination showed a deviated nasal septum due to tumour mass and radiological examination (CT) confirmed bilateral spread of the osteoma from the paranasal sinuses over the frontal sinus to the nasofrontal duct and the frontal recess. Due to the expansion of the tumour, it may be classified as a stage III osteoma according to the updated classification by Giotakis et al. [15]. The size of the osteoma has been measured from 4 cm to 2.5 cm [Fig. 1].

For more than 20 years a watch-and-wait approach was applied due to the benign nature of the tumour and the patient's fear of surgery. Her symptoms continued to worsen over time and significantly started to restrain her daily life. Finally in 2009, after two years of observation and the patient's consent, surgery appeared to be the best option. The following procedure was conducted under general anaesthesia and divided into two parts, performed by two different board-certified surgeons. The first part was performed by an ear, nose, and throat (ENT) surgeon (Klaus Böheim, MD), who operated endoscopically with an endonasal approach. The second part was performed by a maxillofacial surgeon (Dritan Turhani, MD), who approached the tumour through a coronal incision. The patient received perioperative antibiotics and

For the first part of the procedure, a speculum was used initially to access the surgical area in the left nasal cavity. The frontal portion of the medial nasal cavity was entirely worn out, and the rudiment was medialised. The frontal part of the nasal cavity and the medial nasal duct were filled with a yellow osseous tumour that had fused into the surrounding tissue. The tumour was separated from healthy tissue with a Blakesley forceps, starting at the caudal end. The bone around the caudal portion was too compact, but the surgeon was able to remove the tumour from the interwoven bone around the agger nasi by performing a core needle biopsy. Once the tumour mass had been mobilised, the medial nasal concha was found to be fully damaged, and the septum was highly perforated. The tumour was fully mobilised using Piezosurgery® (Mectron, Germany) and a punch biopsy. Due to its size, the tumour had to be fragmented and removed in smaller segments [Fig. 2].

The second part of the operation was then performed by the maxillofacial surgeon. The patient's hair was parted to avoid shaving the head and facilitate the healing process. Subsequently, an osteoplastic flap was raised. The tumour was removed in segments using Piezosurgery® (Mectron, Germany) and sent for histological examination [Fig. 2]. As

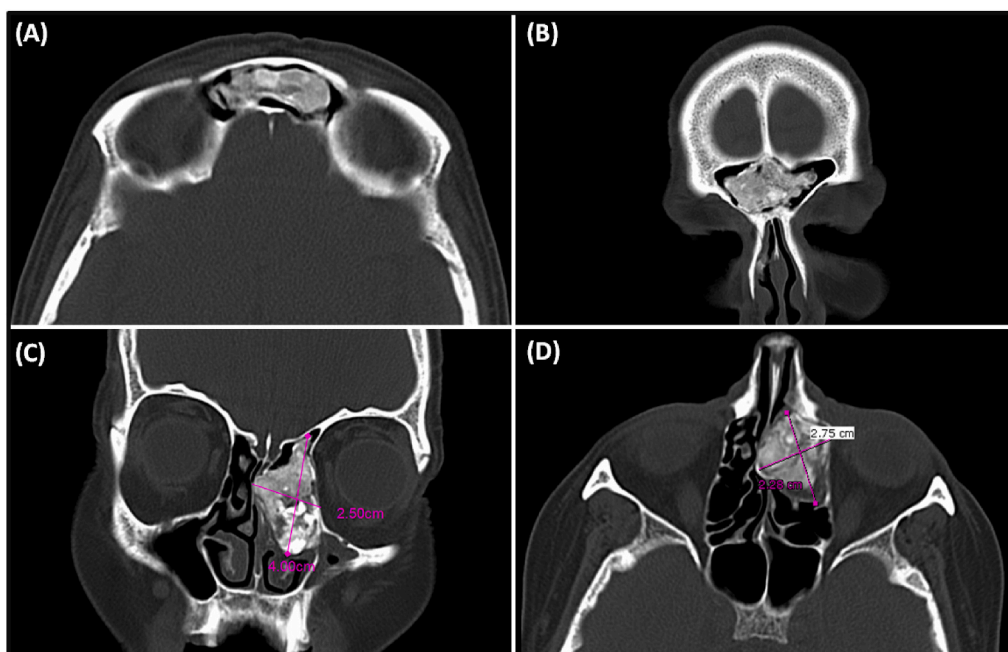
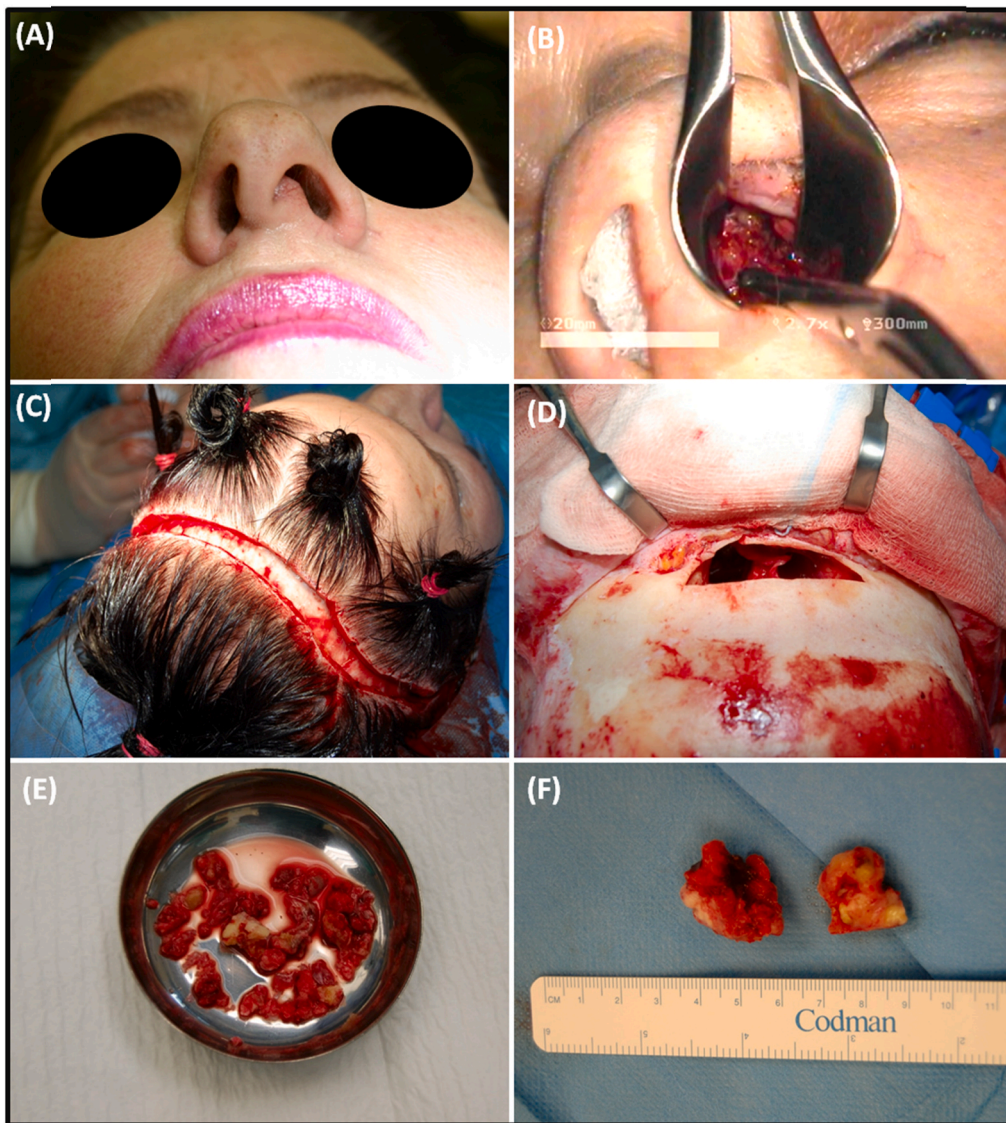


Fig. 1. (A),(B) Preoperative CT in 2009: Tumour in frontoethmoid; (C), (D) Preoperative CT in 2009: Tumour in the ductus nasalis.



**Fig. 2.** (A) Deformation of the septum nasalis (preoperative); (B) Removal of the tumour through the ductus nasalis; (C) Coronal incision; (D) Opening of the os frontale; (E) Removed tumour from the nose; (F) Removed tumour from the os frontale.

the posterior sinus wall was preserved, a neurosurgeon was not consulted. After the tumour had been removed, a direct connection was noted between the frontal sinus and the nose. Thus, no drainage was needed. Bleeding was controlled, and the septum was fixed with a splint. Both nasal cavities were tamponised. The osseous lid was repositioned and fixed with two X-plates and a reconstruction plate (Synthes, USA). Two drains (10.0) were fixed behind the ears with 2–0 Prolene™ sutures (Ethicon, Johnson & Johnson, USA). The periosteal flap was fixed with 4–0 Vicryl™ sutures (Ethicon, Johnson & Johnson, USA). Wound closure was performed using 4–0 Vicryl™ (Ethicon, Johnson & Johnson, USA) and 5–0 Prolene™ sutures (Ethicon, Johnson & Johnson, USA). The patient was extubated without difficulties and transferred to the recovery room. The postoperative CT showed that the entire osteoma had been removed. Histological examination confirmed that the lesion was an osteoma. The wound healed without any complications. The patient remained under supervision and was given postoperative antibiotics, a non-steroidal anti-inflammatory drug (NSAID) and a proton pump inhibitor and was discharged in stable condition.

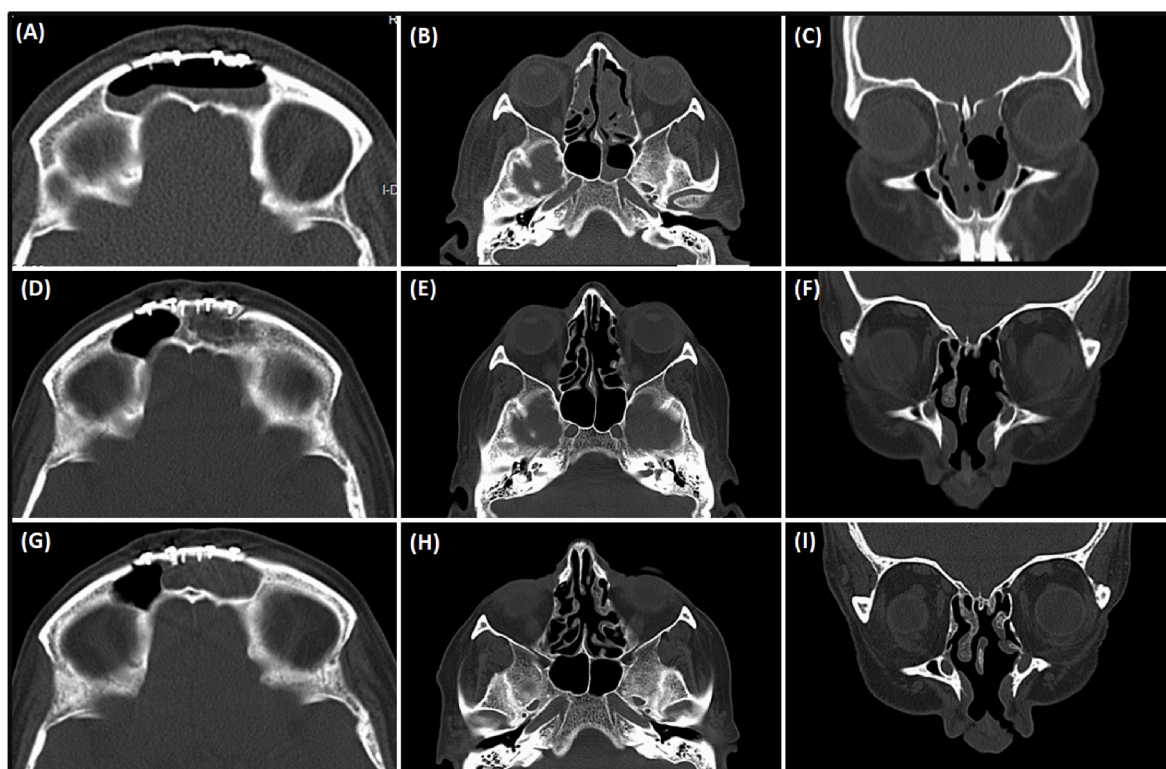
A routine CT examination taken one year after surgery revealed a beginning development of a mucocele. As the symptoms of a mucocele depend on its location, the patient reported about suffering from

headaches again. The CT scans taken during the next follow up examinations showed a continuous expansion of the mucocele, and the patient started to complain about headaches due to its presence. Therefore, surgical removal was discussed and recommended. However, despite numerous consultations, she refused surgical intervention until today. Although her symptoms are worsening again and the total removal of the osteoma has been uneventful, she is still very afraid of surgery. At the most recent appointment, no differentials were noted. The patient reported continuous headaches and pressure in the forehead area [Fig. 3]. She was given a NSAID and endoscopic removal was again strongly recommended.

### 3. Discussion

The aetiology of osteomas is largely unknown. Suggested causes include trauma, infection, and reactive mechanisms [16]. They are usually diagnosed accidentally when they reach a certain size [1]. As osteomas tend to grow slowly, they usually cause no pain or symptoms as long as they have no contact with nerves or other sensitive structures [7]. To diagnose an osteoma, a CT can be considered the most suitable method for diagnosing an osteoma because of its ability to show vascular





**Fig. 3.** (A),(B),(C) Postoperative Computer tomography – directly after surgery; (D),(E),(F) Postoperative CT in 2010–12 months after surgery; (G),(H),(I) Postoperative CT – 10 years after surgery.

contrast in the nidus. Small vascular grooves, a typical feature of osteoma, are seen clearly on CT [16].

A variety of methods are used for the treatment of osteoma, extending from minimally invasive procedures to open surgery [17]. Osteomas are initially managed conservatively with salicylates or NSAIDs. Some tumours resolve spontaneously over 2–6 years [18,19]. Surgery is indicated when the patient is unresponsive to painkillers or medical treatment or is unable to tolerate prolonged NSAID therapy due to its side effects.

Surgical osteoma removal may be performed by the endoscopic technique, the open approach, or a combination of the two. The choice of the procedure depends on several factors, including the location, extent, and size of the tumour, the presence or absence of favourable anatomy, and the surgeon's experience [20]. Further considerations include the patient's safety, minimising morbidity, and reducing aesthetic alterations. A literature review, including 37 cases of giant frontoethmoidal osteomas, concludes that the applied surgical method is irrelevant to the size of the osteoma but rather strongly dependent on the surgeon's experience and availability of equipment's [21].

Endoscopic treatment of frontoethmoidal osteomas was first published in 1992. The procedure involved transnasal removal of an osteoma of the frontal sinus, supplemented with mini-trepanation [22]. However, complete removal may be hindered in patients with a narrow frontal ostium, a tumour widely attached to the orbital roof, or filling the narrow supraorbital recess [23–25].

Regarding the open approach, the osteoplastic flap is a commonly used technique for osteomas of the frontal sinus [26,27]. In cases of grade III or grade IV osteomas, an osteoplastic flap was advised as the method of choice [28]. The procedure has very high success rates and provides excellent access to all regions of the frontal sinus. However, the technique of an open approach also has several drawbacks [26,29]. Therefore, the endonasal endoscopic approach should be used whenever possible, as open surgery involves higher morbidity rates and a longer period of recovery from the operation [24,30]. In the recent published

literature, the most mentioned criteria for an exclusive endoscopic approach are the location, size, and extent of the tumour. The last fifteen years have witnessed efforts on the part of surgeons to extend the boundaries of endoscopic management. In a series of publications from 2007 to 2009, including two comparative studies with a follow up duration of 6 years, and a retrospective study, researchers elaborated on these concepts [20,30,31]. Indications for the endonasal approach were expanded considerably. However, a medial location with the sagittal plane passing through the lamina papyracea remained a critical criterion [31]. Widespread intraorbital involvement was also regarded as contraindication, with a 'footnote' saying that if the pedicle of the lesion was favourably located, it might be possible to perform stepwise caviations, mobilisation, and excision [31]. Dubin and Kuhn noted that five of eight tumours treated exclusively by the endoscopic technique could be classified as grade III osteomas. This signified a further extension of the endoscopic approach [32]. In the following years, other authors also reported the use of endoscopic surgery for grade III or grade IV osteomas [25,33,34].

Although minimally invasive surgery is the most gentle and preferred surgical technique, open surgery may be needed when the tumour is large or its location inaccessible to the endoscopic approach [17]. Despite the advantages of endoscopic surgery, lesions in the frontal sinus are frequently treated by open surgery or in combination with an endoscopic procedure, as in some cases endoscopic surgery alone may be ineffective or impossible [15]. One literature review addressed all 45 reported cases of patients with an osteoma in the frontal sinus from 1975 to 2011 and concluded that the combination of open surgery and endoscopy is the treatment of choice [1]. A retrospective single centre study carried out from 2001 to 2015 also concluded that the combined approach can be successfully applied for resection of complex osteomas [35].

One year after the operation, the patient developed a secondary mucocele, which is reported with an incidence up to 50% after osteoma removal [5]. Mucoceles are benign, slow-growing lesions, consisting of

accumulated mucus unable to flow out due to an obstruction [36]. They are known to develop over a period of two years [36], but in the present case it appeared as early as one year post-surgery. The pressure caused by the mucocele may result in expansion of the maxillary sinus, thinning of the bony wall, further swelling of the orbit, and finally expansion into the cranial cavity and orbit through the weakest part [37]. Surgical excision is recommended [36], but was not conducted since the patient has not consented to further surgery.

Finally, the early detection and removal of the osteoma improves the prognosis for a favourable treatment outcome [1]. The smaller the osteoma, the easier it can be removed exclusively by endoscopy. A watch-and-wait approach for more than 20 years was understandably used in the present case because of the location of the osteoma and decision to perform surgery was made ultimately when the condition affected the patient's quality of life. In principle, the surgical procedure for treating an osteoma should be decided individually after careful consideration of all the above-mentioned factors, since to date there is still no strong consent regarding the best surgical approach.

#### 4. Conclusion

The combination of open and endoscopic surgery remains a safe and straightforward procedure for more complex cases, permitting access and complete removal of giant osteomas in the frontal sinus. Early detection and intervention are crucial for a predictable minimally invasive treatment with a favourable outcome for the patient. Finally, an untreated secondary frontoethmoidal mucocele may lead to headache, facial pressure, orbital pain, and other life impairing symptoms.

#### Ethical approval

The ethical approval has been exempted by our institution.

#### Author contribution

Sarmad Aburas and Benedikt Schneider: study concept and design, writing the paper. Florian Pfaffeneder-Mantai, Oliver Meller and Arne Balensiefer: data collection, analysis, and discussion of data. Dritan Turhani: final approval of the version to be published.

#### Registration of research studies

Not applicable.

#### Guarantor

Dritan Turhani.

#### Provenance and peer review

Not commissioned, externally peer reviewed.

#### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Declaration of competing interest

The authors declare that there is no conflict of interest.

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#### Appendix A. Supplementary data

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