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*Case Report*

# Ossifying fibroma of the ethmoid and sphenoid sinuses: A report of a rare case and literature review

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## ABSTRACT

**Background:** Fibro-osseous lesions include a variety of bone lesions with different clinical and histopathological features.

**Case Description:** We report a case of cemento-ossifying lesion involving the left ethmoid and sphenoid sinus in a 17-year-old male patient. Computed tomography showed an expansile hyperdense lesion with sclerosed peripheral mantle epicentered on the left ethmoid and sphenoid sinus and extending into the left nasal cavity. Magnetic resonance imaging was done for preoperative planning. Transnasal endoscopic resection was performed and histopathological examination confirmed the diagnosis of ossifying fibroma (OF).

**Conclusion:** Involvement of the ethmoid sinus with OF is a rare condition; therefore, we examined the literature for similar cases to highlight the possible clinical presentation and management. Endoscopic management is a safe, effective approach with a low rate of complications.

**Keywords:** Endoscopic management, Ethmoid, Fibro-osseous, Ossifying fibroma, Proptosis

## INTRODUCTION

Fibro-osseous lesions represent a variety of proliferative bone diseases, with each type representing a specific biological and morphological pattern. In these lesions, the normal bone is replaced by fibroblasts and they can be classified into dysplasia or bone tumors.[29] The most commonly seen pathologies include fibrous dysplasia, osteoma, aneurysmal bone cyst, and juvenile ossifying fibroma (OF).[25] In young adults, juvenile OF is a rare type of OF that may involve the skull and facial bones.[5] They have aggressive behavior and a high risk of recurrence if not totally resected.[29] It can be differentiated from fibrous dysplasia by its well-demarcated borders, rapid growth, and the considerable disfigurement that it can cause.[28] In rare cases, it may affect the sinonasal tract.[11]

In this article, we report a case of juvenile OF involving the sphenoid and ethmoid sinuses and review the literature to expose the clinical and radiologic features of this lesion. It is crucial for a skull base surgeon to differentiate between OF and the other osseous lesions.

### Methods

This review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) statement and PRISMA checklist [Figure 1]. The U.S. National Library of Medicine (PubMed), Google Scholar, and Science Direct databases were searched. The search terms included the following: OF, cemento-OF (COF), juvenile trabecular OF (JTOF), psammomatous OF, involving ethmoid sinus, involving sphenoid sinus, ethmoid sinus OF, and sphenoethmoidal OF. All age groups and both genders were included in the study. This review includes only articles describing OFs involving the ethmoid and sphenoid sinuses. Studies reporting OFs in other anatomical locations were excluded. Articles that are not written in English and articles reporting other different pathologies were excluded from the study. The abstracts were examined by two separate researchers independently for inclusion. The inter-reviewer analysis was assessed using Cohen's Kappa scores, and articles were categorized accordingly. We used Statistical Package for the Social Sciences software to conduct the statistical description.

## CASE DESCRIPTION

A 17-year-old male patient was referred to the otorhinolaryngology outpatient clinic with symptoms suggestive of recurrent nasal obstruction over eight months. He also complained of 1 year of progressive left eye proptosis. There was no history of epistaxis, anosmia, or trauma to the face. On examination, a polypoid irregular mass filling the left posterior nasal cavity was observed. It was a whitish mass, and when palpated by the forceps, it had a semisolid consistency. There were no other remarkable findings on general examination. Computed tomography (CT), in Figure 2, showed an expansile hyperdense lesion with a sclerosed peripheral mantle epicentered on the left sphenoid sinus and extending into the left nasal cavity. Magnetic resonance imaging (MRI)-T1-weighted image (T1WI), in Figure 3, showed a well-defined hyperintense lesion filling the left ethmoid, sphenoid sinuses, and the left nasal cavity. The lesion displaces the lamina papyracea, medial, and inferior recti.

The optic nerve displayed a normal signal intensity. The patient was prepared for surgery. The patient was operated on in a supine position under general hypotensive anesthesia. We performed a left ethmoidosphenoidectomy under general anesthesia using an endoscope. The nasal cavity was decongested with 1:10,000 adrenaline-soaked cotton pledgets. The 30° endoscope was introduced in the left nasal cavity. The bulla ethmoidalis is encountered medially and penetrated using the powered debrider. The lamina papyracea was identified laterally, and we tried to preserve its mucosa. We continued dissection posteriorly until we identified the basal lamella and the posterior ethmoid cells. Complete dissection of the ethmoid cells was done under the guidance of a 45° endoscope. The lesion was excised in a piecemeal fashion. The mucosa in the sphenoid sinus was polypoidal and congested due to recurrent inflammation and obstruction. Routine postoperative endoscopic examination of the left sinonasal cavity was performed, and no evidence of cerebrospinal fluid leaks was present. Histopathological examination revealed the proliferation of stromal spindle fibroblasts presence with woven and lamellar bone deposits composed of scattered osteoblasts and osteocytes [Figure 4]. There was no necrosis or features of malignancy. The diagnosis was compatible with OF. The patient was discharged uneventfully. Six months later, the symptoms completely disappeared. Rhinoscopic examination showed no residual lesion. A follow-up CT was done on an outpatient basis five months after surgery [Figure 5].

## RESULTS

A total of 200 articles were identified from different databases. About 197 records were screened for different inclusion and exclusion criteria**.** About 50 reports were excluded due to different reasons such as non-English language, different pathologies, or OF involving locations other than the ethmoid and sphenoid sinus. The clinical presentations, pathological types, and surgical management approaches were reviewed. The PRISMA flow chart for screening and selection of articles is shown in Figure 1. Cohen Kappa's inter-rater coefficient was 0.85.

The age of the patients ranged from 3 months to 65 years. Males were more affected than females, and they tended to be younger at presentation. About 60% of male patients presented with OF under the age of 30 years. The patients presented with symptoms related to the expansion of the mass, namely, proptosis, nasal obstruction, headache, facial masses, and deformities. Proptosis was the most common presentation in all types of OF involving the ethmoid and sphenoid sinuses. The asymptomatic presentation was reported only in four cases [Table 1]. Open surgical approaches were the most common treatment offered. Table 2 shows some of the numbers of cases reviewed in the selected articles.

## DISCUSSION

Manes, in 1872, was the first to identify OF as a rare benign osseous lesion.[16] It commonly affects the facial bones, especially the mandible, and, to a lesser extent, the sinonasal tract. OF commonly affects the maxillary sinus and ethmoid sinus, but the frontal and sphenoid sinus are rarely affected.[16]

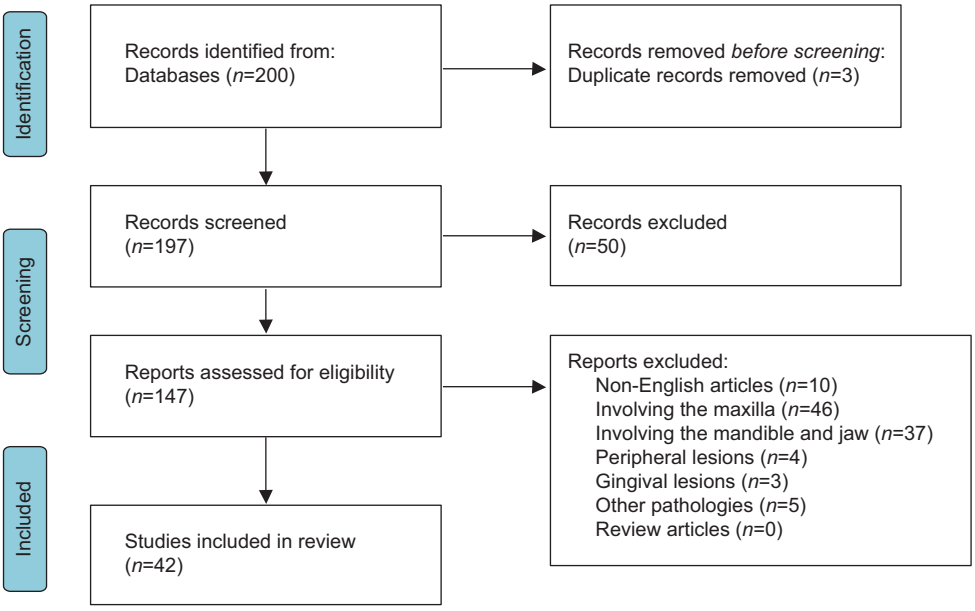


**Figure 1:** Preferred Reporting Items for Systematic Reviews and Meta-analyses flowchart of article selection.

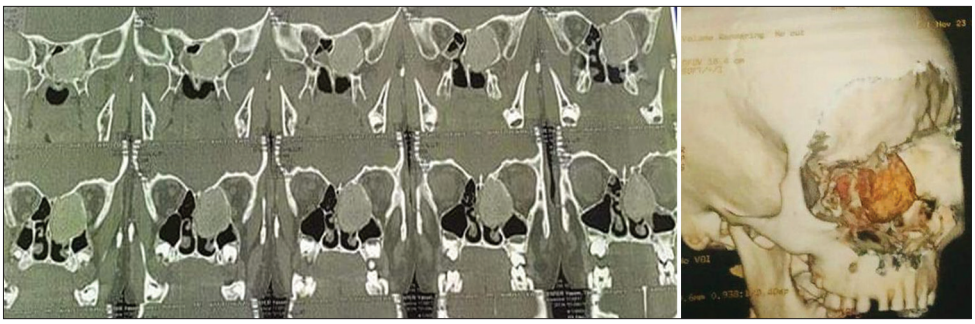

**Figure 2:** Coronal computed tomography scan and 3D reconstruction of the paranasal sinuses showing an expansile hyperdense lesion with a sclerosed peripheral mantle epicentered on the left sphenoid sinus and extending into the left nasal cavity.

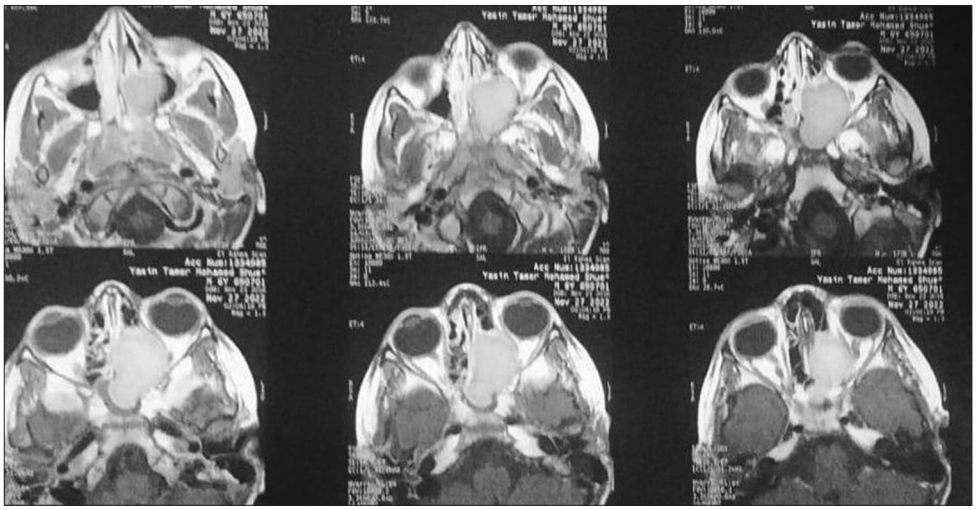

**Figure 3:** Magnetic resonance imaging-T1-weighted image of paranasal sinuses and the brain (axial cuts) showing a well-defined hyperintense lesion filling the left ethmoid, sphenoid sinuses, and the left nasal cavity.

**Table 1:** A list of studies of OF involving the ethmoid and the sphenoid sinuses.

| Study | Symptoms | Age at presentation | Tumor extensions | Tumor dimensions (mm) | Surgical approach | Surgical complications | Pathological type | Follow up | Reference |
|---|---|---|---|---|---|---|---|---|---|
| Appiani *et al.* | Headache | 32 | Ethmoid sinus | 9*10*10 | Endoscopic anterior and posterior ethmoidectomy and sphenoidotomy | NM | Juvenile Psammomatoid | NM | [4] |
| | Follow up of previously resected lesion | 13 | Anterior and posterior ethmoid and maxillary sinus with the medial orbital wall. | 38*16*18 | Endoscopic anterior and posterior ethmoidectomy and medial maxillectomy | NM | Juvenile trabecular type | NM | |
| | Asymptomatic | 46 | Posterior ethmoid, sphenoid sinus, and the middle cranial fossa | 33*24*28 | Endoscopic anterior and posterior ethmoidectomy and sphenoidotomy | NM | COF | NM | |
| | Proptosis and nasal obstruction | 27 | Anterior and posterior ethmoid, sphenoid sinus, and the medial orbital wall | 65*50*49 | Endoscopic anterior and posterior ethmoidectomy, medial maxillectomy, and sphenoidotomy | NM | Juvenile Psammomatoid | Recurrence after 6 months | |
| | Proptosis and nasal obstruction | 12 | Posterior ethmoid, sphenoid, and middle cranial fossa. | 43*24*44 | Endoscopic anterior and posterior ethmoidectomy and sphenoidotomy | NM | Juvenile Psammomatoid | Recurrence after 6 months | |
| | Nasal obstruction | 20 | Posterior ethmoid, sphenoid, and middle cranial fossa. | 72*58*60 | Endoscopic anterior and posterior ethmoidectomy and sphenoidotomy | NM | Juvenile Psammomatoid | Recurrence after 6 months | |
| Choi *et al.* | Epistaxis | 14 | posterior ethmoid sinus | NM | Endoscopic ethmoidosphenoidectomy of the right side, middle turbinectomy, and middle meatal antrostomy | No complications reported | Juvenile Psammomatoid | No Recurrence after 1 year follow-up | [3] |
| Liu *et al.* | NM | 11 | Right ethmoid sinus | NM | Endoscopic anterior and posterior ethmoidectomy | No complications reported | NM | No recurrence | [13] |
| Salina *et al.* | Face deformity and nasal congestion | 23 | Right ethmoid and frontal sinuses | NM | NM | NM | COF | No Recurrence for 9 months follow-up | [23] |
| Sterling *et al.* | Headache, decreased vision, galactorrhea, and proptosis | 26 | The sphenoid sinus and the right greater and lesser wings of the sphenoid | NM | Right fronto-pterional craniotomy with zygomatic osteotomy | NM | NM | NM | [24] |
| Nakajima *et al.* | Decreased vision | 19 | Right anterior sphenoid sinus | 15*20*12 | NM | NM | Juvenile Psammomatoid | NM | [22] |
| Margo *et al.* | Proptosis | 8 | Left ethmoid and sphenoid sinuses | NM | combined transnasal and intracranial approach | NM | Juvenile Psammomatoid | No Recurrence for 9 months follow-up | [18] |
| | Proptosis and decreased vision | 8 | The left ethmoid sinus, sphenoid sinus, both sphenoid | NM | combined transnasal and intracranial approach | NM | COF | No Recurrence for 1-year follow-up | |

*(Contd...)*

**Table 1:** *(Continued).*

| Study | Symptoms | Age at presentation | Tumor extensions | Tumor dimensions (mm) | Surgical approach | Surgical complications | Pathological type | Follow up | Reference |
|---|---|---|---|---|---|---|---|---|---|
| | | | bones, left maxillary sinus and left nasal cavity and both pterygoid fossa, and the clivus | | | | | | |
| Cheng *et al.* | Facial swelling | 28 | Right maxillary and sphenoid sinuses | NM | Caldwell–Luc approach | NM | COF | NM | [2] |
| Wang *et al.* | NM | 6 | Ethmoid and sphenoid sinuses and the orbit | NM | Endoscopic and infraorbital incision approach | No complications reported | NM | Recurrence over 32 months | [27] |
| | NM | 13 | The ethmoid sinus and the anterior cranial fossa | NM | Endoscopic | No complications reported | NM | No recurrence over 8 months | |
| | NM | 14 | Ethmoid and sphenoid sinuses and the orbit and the anterior cranial fossa | NM | Endoscopic | No complications reported | NM | Recurrence over 55 months follow-up | |
| | NM | 14 | The ethmoid sinus and the orbit | NM | Endoscopic | Yes | NM | No recurrence over 6 months | |
| | NM | 14 | Ethmoid, maxillary, and sphenoid sinus | NM | Endoscopic | No complications reported | NM | Recurrence over 49 months | |
| | NM | 14 | Ethmoid and sphenoid sinus and the orbit | NM | Endoscopic | No complications reported | NM | No recurrence over 8 months | |
| | NM | 15 | Ethmoid and sphenoid sinuses and the orbit and the anterior cranial fossa | NM | Endoscopic | Yes | NM | No recurrence over 23 months | |
| | NM | 15 | Ethmoid, maxillary sinuses, and the orbit | NM | Endoscopic | Yes | NM | No recurrence over 12 months | |
| | NM | 15 | Ethmoid, maxillary, sphenoid sinuses, and the orbit | NM | Endoscopic and sublabial approach | Yes | NM | No recurrence over 12 months | |
| | NM | 17 | Ethmoid sinus and anterior cranial fossa | NM | Endoscopic | Yes | NM | No recurrence over 11 months | |
| | NM | 17 | Ethmoid and sphenoid sinuses and the orbit | NM | Endoscopic | No complications reported | NM | Recurrence over 23 months | |
| | NM | 17 | The ethmoid sinus and the orbit | NM | Endoscopic | No complications reported | NM | No recurrence over 5 months | |
| | NM | 20 | The ethmoid sinus, the orbit, and the anterior cranial fossa | NM | Endoscopic and frontal sinus trephination | Yes | NM | No recurrence over 25 months | |

*(Contd...)*

**Table 1:** *(Continued).*

| Study | Symptoms | Age at presentation | Tumor extensions | Tumor dimensions (mm) | Surgical approach | Surgical complications | Pathological type | Follow up | Reference |
|---|---|---|---|---|---|---|---|---|---|
| | NM | 23 | Ethmoid, sphenoid sinus, and the anterior cranial fossa | NM | Endoscopic | Yes | NM | No recurrence over 13 months | |
| | NM | 26 | Ethmoid and sphenoid sinus | NM | Endoscopic | No complications reported | NM | No recurrence over 8 months | |
| | NM | 26 | Ethmoid and orbit | NM | Endoscopic | Yes | NM | No recurrence over 45 months | |
| Malcomson | Left eye proptosis | 24 | Anterior, posterior ethmoid and anterior cranial fossa | NM | external ethmoid approach | No complications reported | NM | NM | [15] |

OF: Ossifying fibroma, COF: Cemento-ossifying fibroma, NM: Not mentioned

There are three histological variants with different biological behaviors, including COF, juvenile psammomatoid OF (JPOF), and JTOF.[6] COF commonly affects females in the third to fourth decades of life, while JTOF commonly affects children between 8 and 12 years.[10] JPOF commonly affects the craniofacial bones as the maxillary and ethmoid sinuses.[17]

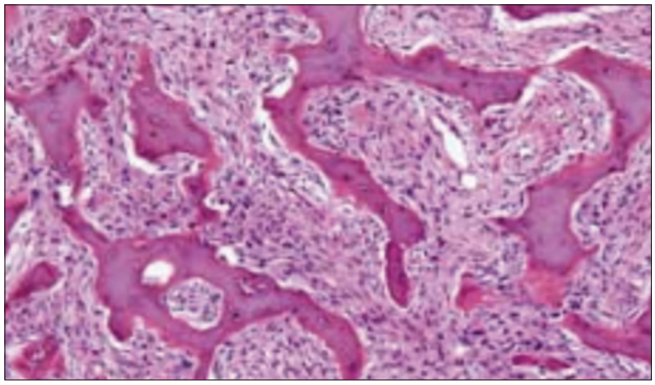

**Figure 4:** Microscopic picture (Hematoxylin and Eosin) staining shows features of juvenile trabecular ossifying fibroma with proliferation of stromal spindle fibroblasts, presence with woven and lamellar bone deposits composed of scattered osteoblasts and osteocytes.

Clinical presentations of OF are variable, and they differ according to the location. OF may be asymptomatic for a long time. Patients may present with a painless facial swelling, which may reside for several years.[30] Symptoms of sinus obstruction are a common presentation of OF and include headache, rhinorrhea, epistaxis, and disturbed sense of smell.[26] Vision problems, proptosis, epiphora, and ophthalmoplegia may be evident when the tumor invades the orbit. Large lesions may invade the anterior skull base; however, cerebral complications are not frequently reported.[9] In a case series of 31 lesions, Wang *et al*. did not report any cerebral symptoms.[27]

These lesions are diagnosed with CT, and MRI is used for preoperative surgical planning. OF usually appears as a well-defined lesion. The density of fibrous and osseous tissues is the major determinant of the radiological appearance of these lesions. In the advanced stages of OF, the center is filled with mature bone tissues, while in the earlier stages, the center

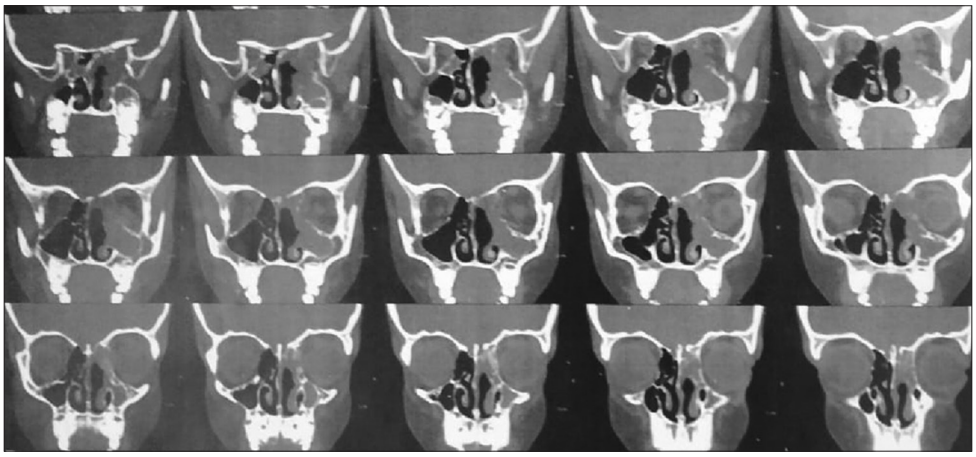

**Figure 5:** Postoperative computed tomography image shows a small residual tumor in the left compartment of sphenoid sinus and the left medial orbital wall and postoperative changes in the maxillary sinus.

**Table 2:** Summary of the articles selected showing descriptive variables of the cases.

| | Trabecular | Psammomatoid | Cemento-ossifying |
|---|---|---|---|
| Age | 7–34 Y | 3–65 Y | 3 W–41 Y |
| Involving the ethmoid sinus | 7 | 14 | 6 |
| Involving both sphenoid and ethmoid sinuses | 9 | 5 | 1 |
| Asymptomatic | 2 | 2 | 0 |
| Presented with a facial deformity | 0 | 7 | 1 |
| Presented with proptosis | 9 | 8 | 5 |
| Other presentations | 5 | 3 | 3 |
| Treated endoscopically | 5 | 6 | 4 |
| Treated with open surgery | 8 | 9 | 4 |
| Treated with a combined approach | 2 | 1 | 3 |

is usually soft and fibrous.[20] Associated remodeling of the involved sinuses may be present. On MRI, T1WI, OF usually has a low to intermediate signal, while on T2-weighted image (T2WI), the peripheral ossified part usually has a lower signal than the central part.[21] Calcifications also can happen in these lesions and usually have hypointense signals on T2WI.[21] The post-contrast T1WI usually shows enhancement in cases of OF.[8,21] Our CT and MRI findings were consistent with these findings.

There is no general consensus about the management strategy for OF, and it differs according to the location of the lesion and the rate of growth. The treatment of choice is complete surgical resection, while conservative management is tried in asymptomatic cases.[12,25] For lesions involving the mandible, simple curettage is sufficient.[19] However, radical resection is the treatment modality in lesions involving the sinonasal tract, orbit, and skull base.[16,19] External approaches allow for wider exposure and more tumor control than endoscopic approaches. Lateral rhinotomy, sublabial approach, and craniofacial resection are the most widely used external approaches.[7] The external approaches have some disadvantages, including the resulting facial scarring, which may affect proper facial growth and symmetry.[7] In certain cases, the use of a sinonasal endoscope is a safer and more convenient method with a lower hospitalization rate.[1,12] Ma *et al*. reported endoscopic management with image-guided navigation in treating OF.[14] Wang *et al*. also documented the safety and effectiveness of the endoscopic approach in the management of OF.[27] However, in cases with intracranial extension, they found that the endoscopic approach was not sufficient in totally resecting these lesions.[27] In this article, we tried to expose different studies with different approaches for the management of ethmoidal and sphenoidal OF [Tables 1 and 2].

## CONCLUSION

OF is an uncommon benign tumor that should be considered in the differential diagnosis of sinonasal obstructive lesions. In the present study, we report a rare case of OF involving the sphenoid and ethmoid sinuses that were treated endoscopically with complete resolution of the symptoms. Endoscopic management is a safe, effective approach with a low rate of complications.

### Ethical approval

The research/study was approved by the Institutional Review Board at Cairo University Hospital, number CU1245, dated January 10, 2023.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

### Financial support and sponsorship

Nil.

### Conflicts of interest

There are no conflicts of interest.

### Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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