Chevreux M, et al., J Clin Stud Med Case Rep 2023, 10: 194
DOI: 10.24966/CSMC-8801/1000194

HSOA Journal of Clinical Studies and Medical Case Reports

Case Series

Ossifying Fibroma of the Paranasal Sinuses: About Three Cases and Review of the Literature

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Abstract

Benign bony tumours of the paranasal sinus, orbit and skull base are uncommon entities with a higher incidence in males. Among them are ossifying fibromas (OF). The nomenclature and terminology have changed a lot in recent years.

We report herein three cases of (OF) of the paranasal sinuses successfully managed in the ENT department of the CHU UCL Namur on the site of Godinne between October 2012 and August 2023. The first case was a juvenile aggressive trabecular ossifying fibroma affecting a 14 year-old girl. The second was a case of trabecular ossifying fibroma in a 56 year-old man infiltrated with a squamous cell carcinoma. The third case was a psammatoid ossifying fibroma in a 30 year-old woman. All these cases were operated endoscopically with a navigation system.

Patient 2 received a preoperative chemotherapy and a postoperative radiotherapy. The first case required surgical revision one year after the first due to local recurrence.

Patient 1 and 3 have small residual tumour that did not grow with time. All the patients are asymptomatic with a minimum follow-up of 2 years (range: 2 to 11 years/mean 4.6 y).

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Citation: Chevreux M, De Dorlodot Cl, Eloy Ph, Riahi I, Nollevaux Mc (2023) Ossifying Fibroma of the Paranasal Sinuses: About Three Cases and Review of the Literature. J Clin Stud Med Case Rep 10:194.

Received: September 12, 2023; **Accepted:** September 22, 2023; **Published:** September 29, 2023

We report clinical data, imaging, operative data, and follow-up data and review the relevant literature.

Introduction

Benign bony tumours of the paranasal sinuses, orbit and skull base are uncommon entities [1]. Among them, we find the fibro-osseous lesions usually divided into three different types, including osteoma, fibrous dysplasia and ossifying fibroma [2]. Ossifying fibromas (OF) are benign fibro-osseous neoplasms affecting the jaws and the craniofacial skeleton? They are usually slow-growing lesions formed by normal bone being replaced by a fibrous cellular stroma containing foci of mineralization or ossification.

The nomenclature and classification of the ossifying fibromas have changed a lot in recent years. In the past, they were exclusively classified as odontogenic tumours but more recently, they are classified as odontogenic tumours when they involve the jaws or mandible and as non-odontogenic tumours when they involve the paranasal sinus cavities or the skull base [3-5].

Methods

We report herein three cases of ossifying fibroma involving the paranasal sinus cavities and the skull base managed between 2012 and 2023 in the Head and Neck department of the CHU UCL Namur, on the site of Godinne.

Medical records were reviewed for age, sex, clinical presentation, pre- and postoperative imaging, treatment approach, and outcomes. All pathology slides were studied by a head and neck experienced pathologist.

The clinical characteristics, imaging findings, surgical approaches, histological findings and prognosis were discussed with a review of the literature.

Patient 1

A 14-year-old Vietnamese girl living in Belgium was referred in October 2012 for a significant right sided nasal obstruction and right exophthalmos evolving since several months. There were no reports of epistaxis or pain. Clinical examination revealed a significant left septal deviation and an expansive process of firm consistency obstructing the right nasal cavity.

The sinuses CT scan showed a tumoral process obliterating the right nasal cavity, originating from the right ethmoidal sinus, with ipsilateral orbital extension and no invasion of the orbital periosteum. The septum was deviated towards the left nasal cavity (Figures 1a,b).

The patient underwent a surgical resection of the mass under endoscopic control. Surgery was difficult due to the presence of intratumoral vascular lakes that caused significant intraoperative bleeding. The histopathological examination of the removed specimen confirmed the diagnosis of a juvenile ossifying fibroma in its trabecular form also called aggressive juvenile ossifying fibroma (Figure 2).

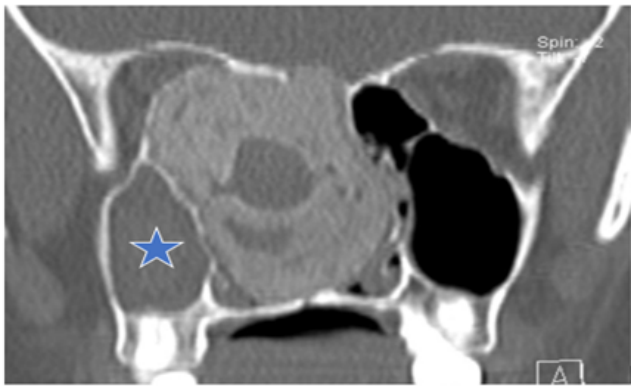

Figure 1a: Bone window sinus CT scan/Coronal cut; huge tumor originating from the ethmoid sinus extended towards the orbit and nasal cavity. Within the tumor some hypodense areas are seen. There is also an opacification of the right maxillary sinus (★).

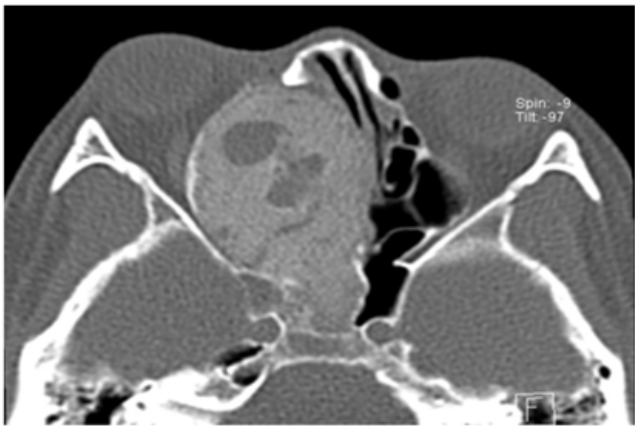

Figure 1b: Bone window sinus CT scan/axial cut; huge tumor with some hypodensedense areas displacing the orbit laterally.

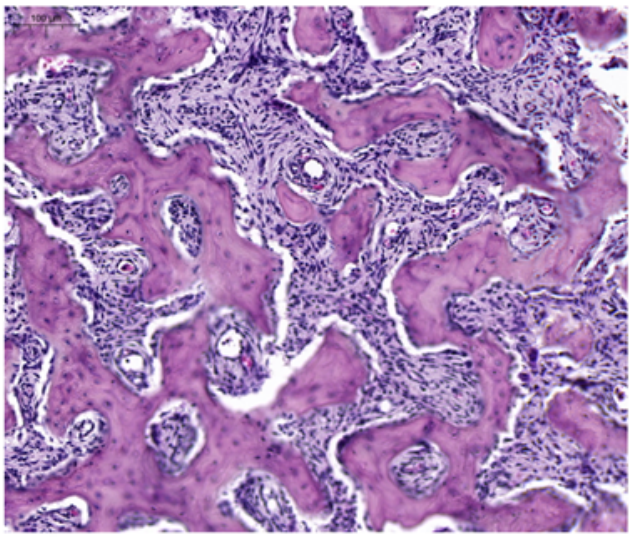

Figure 2: Cellular fibrous stroma composed of spindled fibroblastic cells with bony trabeculae surrounded by plump osteoblasts. Trabeculae show an anastomosing pattern.

The early postoperative follow-up was uneventful and the postoperative sinus CT scan was rather reassuring. One year later a CT scan showed a recurrence of the tumor. A revision surgery was therefore carried out, allowing subtotal exeresis of the mass. Postoperative imaging (2015, 2020, 2023) confirmed the persistence of small tumoral remnants in the skull base. (Figures 3a & b) CT in 2020.

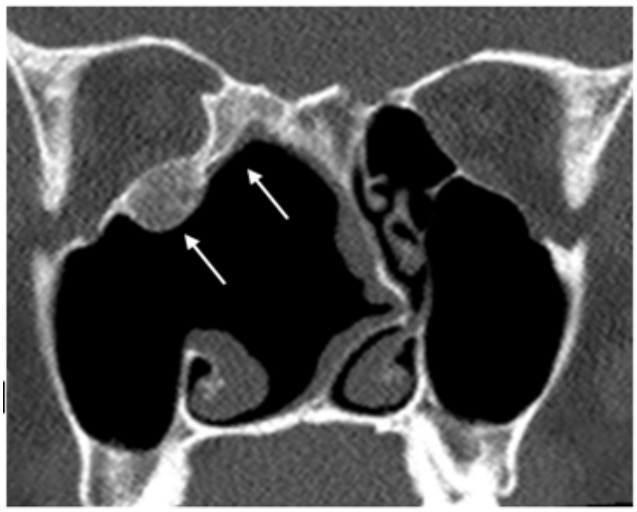

Figure 3a: Bone window sinus CT scan / coronal cut; postoperative result: persistence of a left septal deviation and small residual tumor (arrows).

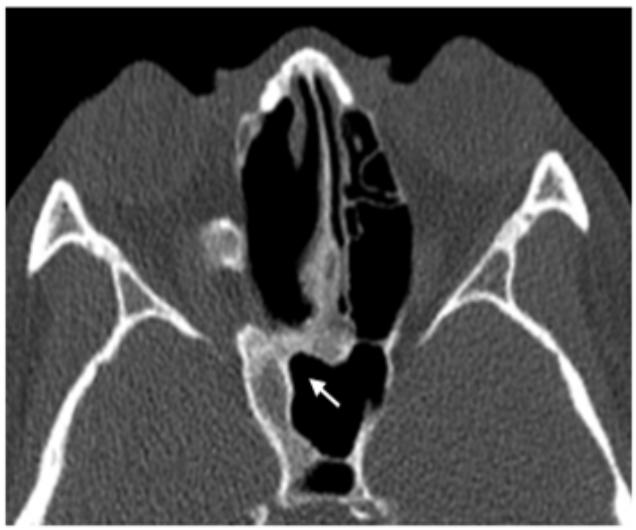

Figure 3b: Bone window sinus CT scan/axial cut: persistence of some residual tumor on the skull base (arrows).

In 2023, the patient is still asymptomatic despite the persistence of the septal deformity. A septoplasty has not yet been performed and the sinus CT scan confirms the stability of the residual lesions. (Figure 4a) CT in 2023.

Patient 2

A 54-year-old man with no significant past medical history was referred in January 2020 to the ENT department for chronic bilateral nasal obstruction and bilateral dacryocystitis.

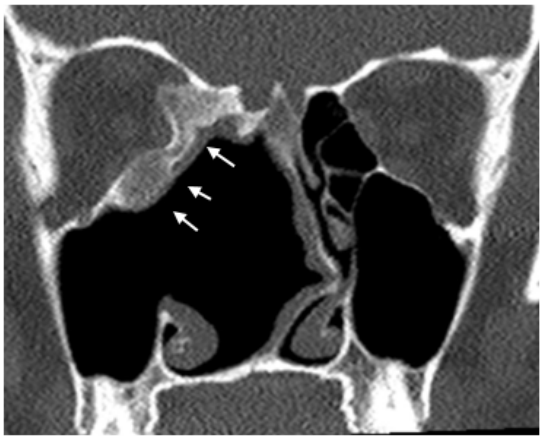

Figure 4a: Sinus CT scan/coronal cut/persistence of some residual tumour, which do not evolve.

The clinical examination revealed a bulging of the septum obstructing the nasal cavities. Imaging (CT and MRI) showed a heterogeneous mass centered on the nasal septum and obstructing both nasal cavities (Figures 5a-c).

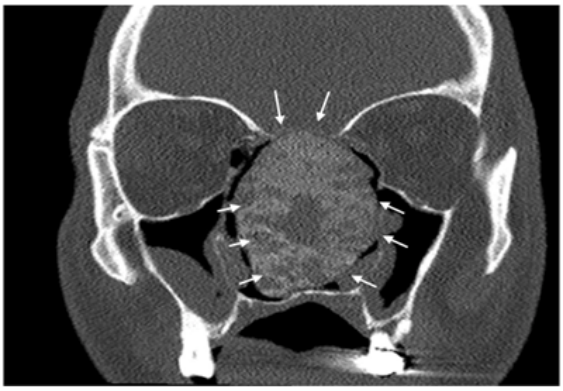

Figure 5a: Coronal bone window pre-operative CT scan showing a tumor developed from the nasal septum, extending to the skull base without intracranial extension (white arrows).

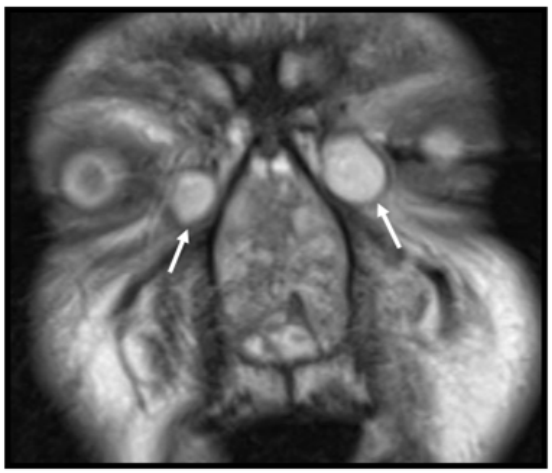

Figure 5b: MRI bilateral dilation of the lacrimal sacs (white arrows) due to the compression of both naso lacrimal ducts by a tumor.

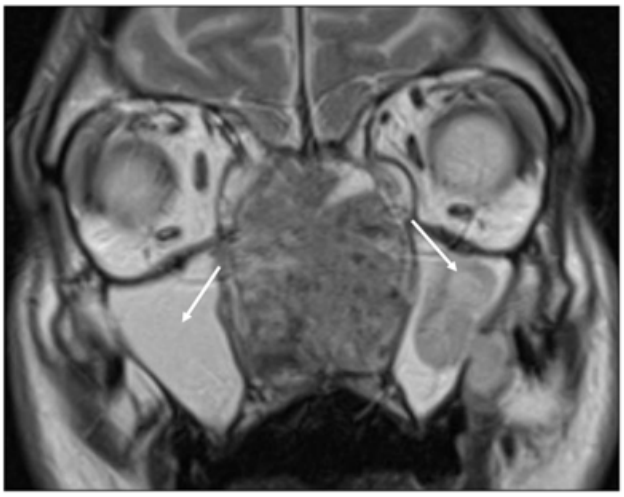

Figure 5c: Coronal MRI images showing bilateral retention maxillary sinusitis (white arrows).

A large biopsy was performed under general anesthesia. The histological diagnosis was that of an ossifying fibroma in its trabecular form with areas infiltrated by a squamous cell carcinoma expressing the P16 protein (Figure 6). The tumor was classified as cT3 cN0 cM0. Induction chemotherapy was then performed including three courses of Docetaxel, Cisplatin and Fluorouracil. The patient underwent, then, endoscopic surgical resection of the mass, followed by radiotherapy.

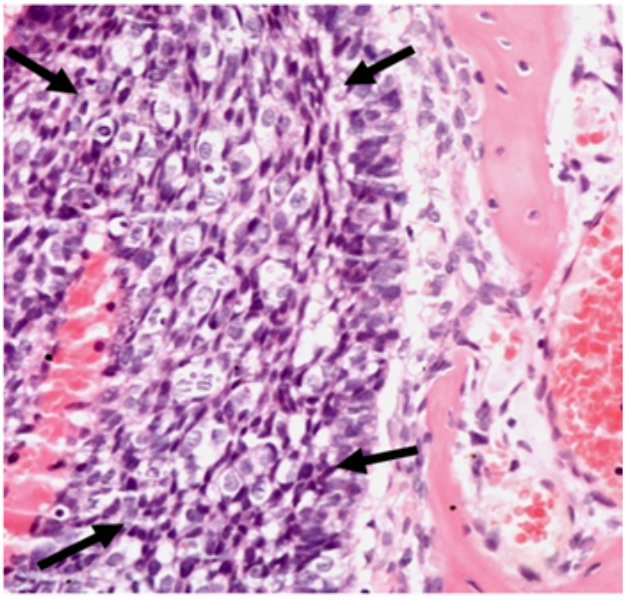

Figure 6: Large cluster of squamous cell carcinoma (*on the left/black arrows*) closely intermigling with osteoid trabeculae (*on the right*).

During the early follow-up period a CT scan revealed opacity of both frontal sinuses. A Draf III frontotomy was then performed. One year later a sinus CT scan performed for the follow-up confirmed the absence of residual tumor or any sinusitis signs (Figures 7a,b).

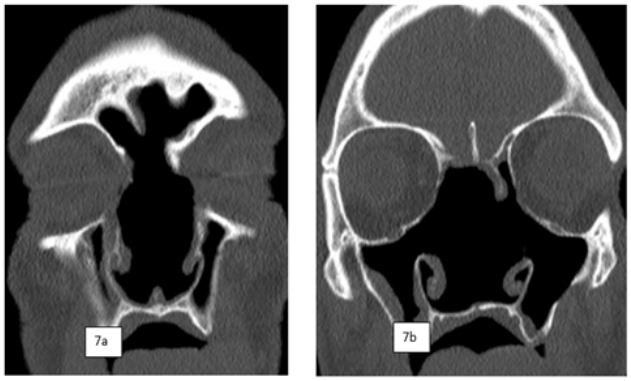

Figure 7a,b: Bone window Sinus CT scan/coronal cuts/visualization of wide opened paranasal cavities without sinusitis or tumor recurrence.

With three years of follow-up, the patient does not present clinical or radiological signs of recurrence.

Patient 3

A 30-year-old patient with no significant medical history was referred to the ENT department in June 2021 following the fortuitous discovery on a sinus CT scan of an extensive bony tumor inserted on the skull base. The radiologists raised the possibility of a chondrosarcoma. There was no purulent rhinorrhea or epistaxis. Clinical examination showed a bony tumor without necrotic or hemorrhagic areas occupying the right nasal cavity. The physical examination did not reveal any lymphadenopathy. The sinus CT scan showed an expansive bony process inserted on the skull base in close relation to the sphenoid sinus (Figures 8a-d).

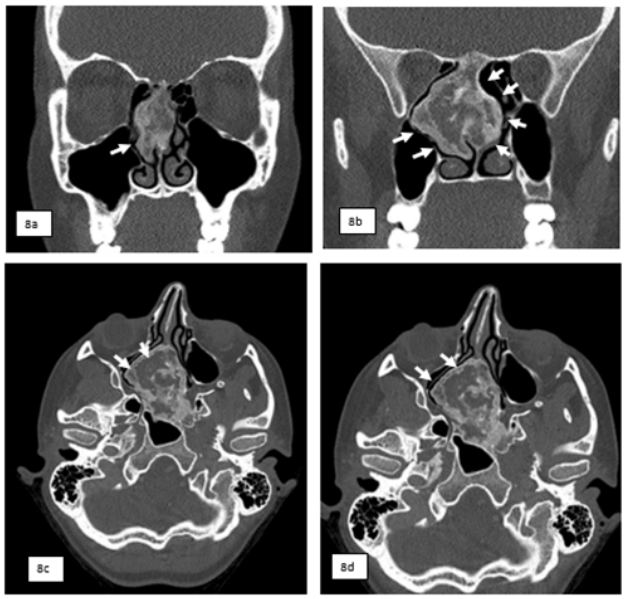

Figure 8a-d: Sinus CT scan coronal and axial cuts showing a bony tumour implanted on the skull base, obstructing the nasal cavity (white arrows).

A large biopsy excision was performed under general anesthesia with endoscopic control. The mass was of hard consistency with intratumoral vascular lakes. The histological diagnosis was that of a psammatoid ossifying fibroma (Figure 9).

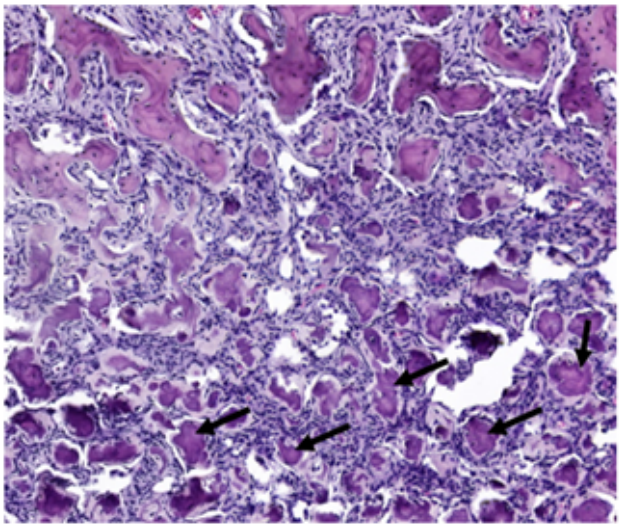

Figure 9: Histological examination of the lesion revealed hyper cellular fibroblastic stroma containing small uniform round basophilic ossicles (psammomatoid bodies), bearing some resemblance to dental cementum.

Postoperatively, the patient was asymptomatic. A CT scan of the sinuses performed a year later revealed a significant residual tumor. A surgical revision was then carried out helped by the navigation system (Brainlab system colibri) and drills use. The bone was very hard and surgeons encountered difficulties to identify a dissection plan between the tumor and the skull base. The postoperative period was uneventful. The postoperative scan carried out in 2023 was satisfactory (Figures 10a-d).

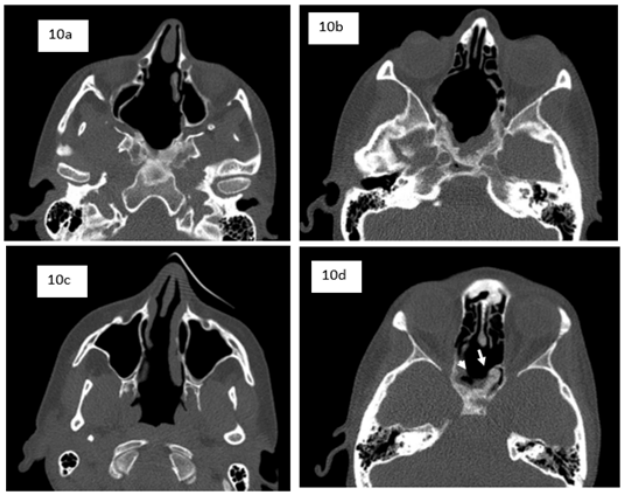

Figure 10a-d: Bone window Sinus CT scan/Axial cuts/Persistence of small residual tumor close to the posterior wall of the sphenoid sinus and the left internal carotid artery.

A wait-and-see attitude was preferred since the patient was asymptomatic. A sinus CT scan is scheduled every year for follow-up.

Discussion

"*Ossifying fibromas*" are benign slow growing tumors that affect adults and young patients.

First described by Menzel in 1872 as a form of osteoma and then by Montgomery in 1927 as a separate entity. The classification and nomenclature have changed considerably in recent years. A distinction is made between Cemento-ossifying fibromas and ossifying fibromas. Cemento-ossifying fibromas (COF) are considered as benign non-cancerous neoplasms of mesenchymal origin occurring exclusively in the tooth-bearing region of the gnathic bones (jaws, maxilla, premolar or molar areas of the mandible). In the fifth edition of the WHO classification (2022), they are classified as odontogenic tumors. They are thought to originate from an ectopic periodontal tissue. Mesenchymal blast cells are capable of producing bone, cementum and fibrous tissue. They are well known by the stomatologists and maxillofacial surgeons. They commonly affect adult women between the third and fourth decade of life. The behavior of these tumors is extremely variable. Most of them are well demarcated, slowly growing mass and characteristically behave in a benign form [6-8]. However occasionally some of them may present as an aggressive gigantiform lesion or be locally destructive [9-10]. Surgical resection is the option of treatment. They are insensitive to radiotherapy and recurrences are rare. On the other hand ossifying fibromas (OF) are located in the paranasal cavities or the skull base. They can take their origin in the ethmoid sinus, the frontal sinus, the sphenoid sinus or can be implanted in the skull base as reported in our cases. These tumors are classified as non-odontogenic lesion (Who classification) [11-14]. The etiopathogenesis is unknown. Histologically there are two forms: the psammatoid and the trabecular ones [12-16].

The first one is characterized by small uniform spherical ossicles resembling psammoma bodies (psammomatoid ossifying fibroma). The other is distinguished by trabeculae of fibrillary osteoid and woven bone (trabecular ossifying fibroma). They can affect adults and young patients. In our series, we observed two trabecular forms, one in a teenager and the other in an adult, and one psammatoid form in an adult. The behavior can be different in adults and teenagers as it was observed in our series. In adults, the tumour is usually well defined and grows slowly. In young patients, the tumor is locally more aggressive, with a trend of recurrence, which was the case of Patient1 who needed a revision surgery one year after the first operation. That is why it is used to call them juvenile aggressive ossifying fibromas.

The symptoms depend on the location and extension of the tumor. The tumor can be asymptomatic and constitute an accidental discovery on CT scan as it was the case with our 3rd patient. When the tumor is large, nasal obstruction, epistaxis, pain, hypertelorism, exophthalmos and dacryocystitis can be the presenting symptoms as it was the case with patients 1 and 2. Among the loco regional complications, there are ophthalmic involvement and fronto-ethmoidal mucocele in addition to the skull base extension. On the CT scan, the tumor is usually well demarcated. A mass effect can be observed with subsequent compression and displacement of the neighboring structures. In our 1st case, we noted a septal deviation and a lateralization of the lamina papyracea without infiltration of the orbital content but exophthalmos. In the 2nd case, the septum was largely infiltrated by the tumor, obstructing both nasal cavities and compressing the lachrymal pathways. In the 3rd case, the nasal cavity was fully occupied by the tumor.

An MRI can be a complementary investigation to evaluate the impact of the tumor on the paranasal sinuses. In our second case, MRI clearly identified bilateral dacryoceles and retention sinusitis in both maxillary sinuses. The definitive diagnosis of this fibrooseeous tumor needs of course a histological evaluation. This evaluation can identify a trabecular or a psammatoid form. That is what we have observed in our cases.

This tumor is considered as benign. Unexpectedly, the pathologist isolated an area infiltrated by a sqaumous cell carcinoma expressing the P16 protein in the second case. Was it a malignant transformation of the OF? In the literature, we found one reference reporting a Low-grade osteosarcoma arising from Cemento-ossifying fibroma. However, to our knowledge infiltration of an (OF) with a sqaumous cell carcinoma was never published before. Is it the result of a malignant transformation of the fibrous tumor or is it more likely an association of two different diseases. The differential diagnosis of the ossifying fibroma includes the osteoma and fibrous dysplasia. There may be some overlap between this different entities.

Paranasal sinus osteoma is classically a rare, benign, slow growing bony tumor that affects the adults in the 2nd and 5th decades. Usually the lesion is monostotic, located in the frontal sinus, followed by the ethmoid sinus, the maxillary sinus or the sphenoid sinus. Classically osteomas are asymptomatic found on an imaging of the sinus performed for Sino nasal symptoms or unrelated complaints. It can be symptomatic when it blocks the normal sinus drainage or has a direct mass effect. Osteomas can present in a polyostotic form, particularly in the Gardner syndrome [17].

Surgery is the treatment of choice in case of symptomatic tumor or in case of complication such as mucocele. Fibrous dysplasia is another differential diagnosis [18-21]. Fibrous dysplasia is uncommon and can affect all ages. Its incidence is estimated to be less than 5% of benign bone lesions. There is no gender predilection. It can affect any bone. In the majority of the cases, it occurs in a monostotic form involving only one bone like the maxillary, frontal or sphenoid bone. A polyostotic form involving multiple bones is mainly seen in young patients. A particular form is when there is a severe circumferential shape around the orbital apex, which may affect the visual prognosis [22]. In young patients, the lesion is still active and characterized by a high vascularity. After the puberty the lesion can be inactive and a wait and see attitude is the rule. Fibrous dysplasia can be associated with endocrinopathy. The follow-up includes serial CT scan to rule out significant mechanical complications and assure biological inactivity. The management of the ossifying fibroma is surgical when the tumor is large, progressive, and symptomatic or when there is a complication due to a mass effect. In absence of these criteria a wait and see attitude may be recommended associated to an accurate follow-up [23]. An endoscopic approach is better when it allows complete resection of the lesion. This depends on the experience of the surgeon. The use of an image-guided system is recommended. Complete tumoral resection is the theoretical goal. However even if the tumors seems well demarcated on the CT scan, it is not always that easy to find a well defined dissection plan intraoperatively. This difficulty was encountered in two of our cases. However, a subtotal resection does not mean recurrence in all the cases as we have observed in our series. In fact, only the young woman required a revision surgery one year after the first surgery and despite persistence of small residual tumour there is no recurrence even after seven years [24].

Previously, the external approach was the recommended one and probably the only way to do. We published a case in 1993 operated via an external approach consisting in a lateral rhinotomy associated to a neurosurgical approach [25]. With the advances of the

endoscopic transnasal techniques, the development of new instrumentations (drills, shaver) and the navigation system, endoscopic endonasal surgery became the modality of choice for an experienced surgeon giving less postoperative morbidity [26-27]. Our second case, similar to the case published in 1993, was successfully operated via a pure endonasal endoscopic approach assisted with a navigation system.

However, the surgery is not that easy as the bone can be very hard and difficult to drill out. Moreover, the tumors frequently have intratumoral vascular lakes that bleed significantly during the surgery. These tumours are classically insensitive to radiotherapy. Our second patient had radiotherapy because of the lesion infiltration by a squamous cell carcinoma. The follow-up includes regular checkup in consultations associated with nasal endoscopy and an imaging, generally a CT scan. The CT scan allows certain tumor residuals to be visualized. This may lead to surgical revision when the recurrence is significant, symptomatic or progressive. The persistence of small areas of bone tumor does not imply systematic surgical revision. In our first and third cases, the residual tumor did not evolve over the years. However, caution should be the rule, particularly in young patients.

Conclusion

Ossifying fibromas are rare, benign slow growing tumours originating in the paranasal sinus cavities and skull base. It is recommended to classify them as trabecular or psammatoid tumour that can affect young patients or adults. The tumour behave more aggressively in teenagers. That is why the terminology of juvenile aggressive tumour must be maintained.

Imaging is necessary to visualize the size and origin of the tumour and to demonstrate any loco regional complications. In case of extensive, progressive or symptomatic mass, a surgery is recommended. A subtotal resection can be an option when an obvious plan of dissection with the skull base is not found.

The surgery has to be performed by an experienced surgeon endoscopically assisted with drills and a navigation system.

A long follow-up is mandatory to rule out any recurrence.

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