CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 53 (2018) 513–516

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Aggressive ossifying fibroma of right ethmoidal sinus: A case report

Yazeed Alghonaim, Salwa ALRashed ALHumaid*, Abdullah Arafat

King Abdullah International Medical Research Center/King Saud bin Abdulaziz University for Health Sciences, ENT Division, Department of Surgery, King Abdulaziz Medical City, NGHA, Saudi Arabia

ARTICLE INFO

Article history:
Received 27 August 2017
Received in revised form
20 December 2017
Accepted 21 December 2017
Available online 26 December 2017

Keywords:
Ossifying fibroma
Ethmoid sinus
Psammomatoid
Paranasal CT
Functional endoscopic sinus surgery
Case report

ABSTRACT

BACKGROUND: Ossifying fibroma is a rare benign fibro-osseous lesion seen in the bones of the head and neck area. It is mostly found in the mandible followed by the maxilla and rarely in the paranasal sinuses along with the orbit and skull bones.
CASE PRESENTATION: A 30-year-old male patient presented with headache and incidental finding of a right ethmoidal sinus ossifying fibroma by paranasal-sinuses CT scan and MRI of the face. A flexible fiber-optic nasal endoscopy examination revealed a right side fullness. A non-contrasted CT scan of the paranasal sinus showed hyperdense lesion at the right ethmoid air cells. Functional endoscopic sinus surgery was performed and multiple nasal-mass biopsies were taken which showed fibro-osseous lesion most consistent with aggressive ossifying fibroma. Later, a near total resection of skull base tumor by endoscopic surgery was done and patient was discharged next day in a good condition.
CONCLUSION: Ossifying fibroma is a rare lesion found in the head and neck area and it is unusual to be found in the paranasal sinuses. Thus crucial attention to the clinical, radiographical and histopathological examination should be taken for more accurate diagnosis and thus appropriate management.

1. Introduction

Ossifying fibroma is a fibro-osseous tumor, mostly found in the craniofacial bones [1]. It is a rare, benign and aggressive lesion. The mandible is considered as the most common location of this neoplasm. However, it could also be seen in the maxilla and paranasal sinuses in some occasions [1–4].

The first case of ossifying fibroma in literature was described in 1872 by Menzel. It was later coined with this terminology by Montgomry in 1927 [5,6]. We report the occurrence of this rare condition of an aggressive ossifying fibroma of the right ethmoidal sinus in King Abdulaziz Medical City (KAMC), Riyadh, Saudi Arabia.

This project has been reported according to the SCARE criteria [7].

2. Case presentation

A 30-year-old male patient, medically free with past surgical history of right mandible bone graft to the right maxilla, with no known existing mandibular pathology at that time, presented with headache for one-week duration, he denied any other sino-nasal symptoms. Due to the asymmetry between his eyes, a para-nasal sinuses CT scan was performed. The CT showed a right soft tissue mass consistent with the para-nasal MRI images done in a private hospital. The patient is otherwise healthy, not complaining of any visual disturbances, nasal obstruction symptoms, epistaxis nor hyposmia. He is not on any medications nor known for any allergies with unremarkable family history.

Upon examination, fiber-optic nasal endoscopy was done and right side fullness was found along with a polyp in the left side with left sided anterior septal deviation. Other head and neck examination was unremarkable while slight enophthalmus and outward eye deviation was noted during the exam.

Laboratory results were all within the normal ranges. As for the radiological findings by non-contrasted CT scan of the paranasal sinuses, three head and neck radiologist were unable to diagnose the nature of the lesion at first. The images showed a hyperdense mass at the right ethmoid air cells extending to the right frontal sinus and left ethmoid sinus and inseparable from the right middle turbinate. Dehiscence of the right lamina papyracea and right cribriform plate was also found (Fig. 1). These radiological findings were consistent with those found by sinus navigation CT and para-nasal MRI study (Fig. 2).

Initially, endoscopic sinus surgery was performed by an otorhinolaryngology and head and neck surgeon and multiple nasal-mass biopsies were taken. Resection of the tumor from the right side and right middle turbinate showed fibro-osseous lesion. It was characterized by spindle cell proliferation, packed with globular cementum droplets and round/ovoid fragments of woven bone. These spindle cells appeared benign and were associated with

* Corresponding author.
E-mail address: salwa.r.h@gmail.com (S. ALRashed ALHumaid).

https://doi.org/10.1016/j.ijscr.2017.12.026

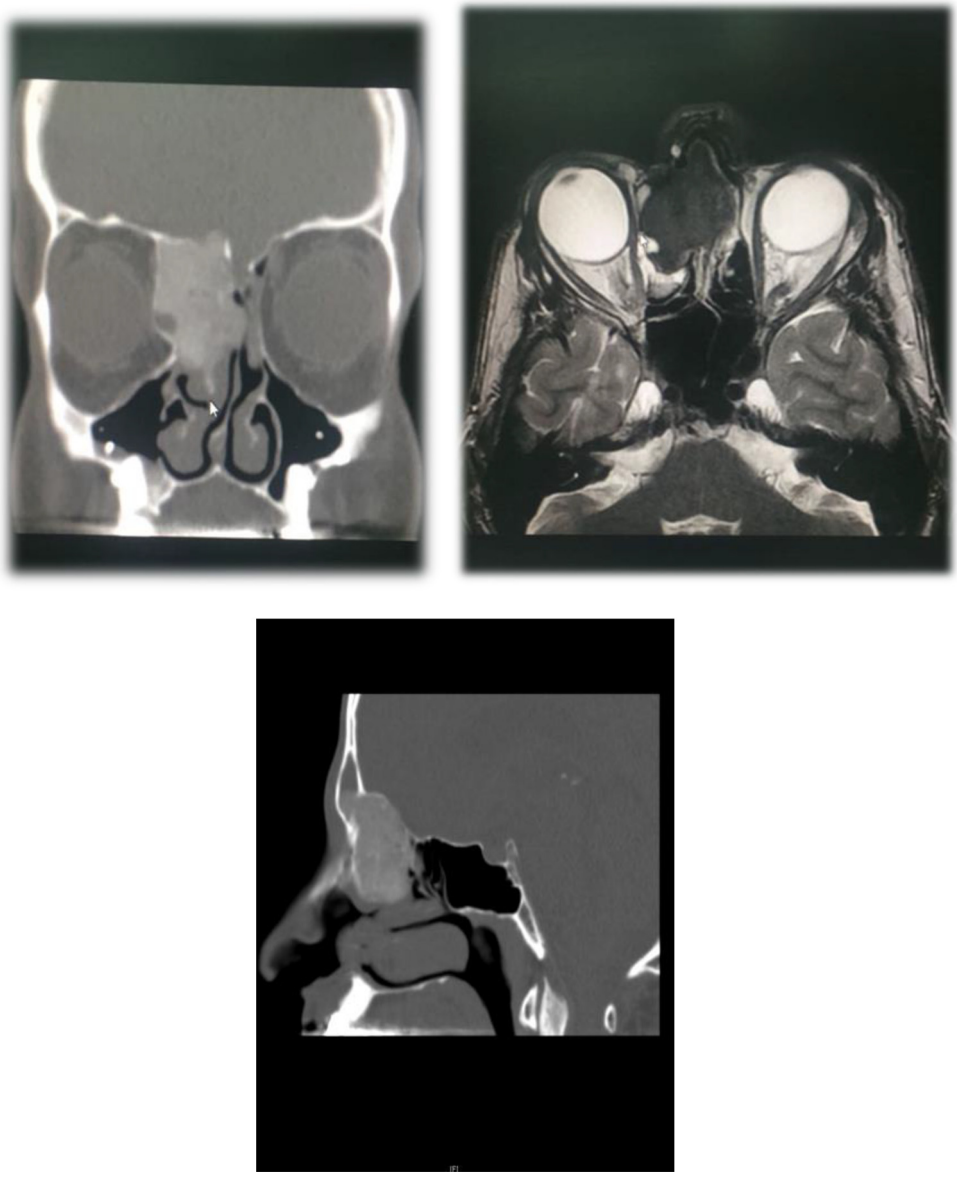

Fig. 1. Paranasal coronal, axial and sagittal CT scan shows right ethmoid sinus and nasal cavity tumor.

psammomatoid cementum droplets and some woven bone and were typical of aggressive ossifying fibroma (Fig. 3).

Later on, the patient underwent another sinus surgery, septoplasty, frontal sinusotomy and near total endoscopic resection of the tumor (Fig. 4). The patient tolerated the procedure well. Pre and post-operative frontal photos were not taken as per the patient desire. He was discharged the next day with instruction to follow up and rule out any recurrence by repeated nasal endoscopy & radiological studies.

3. Discussion

Ossifying fibroma is a benign fibro-osseous lesion seen in the bones of the head and neck area. Although it is considered as a rare, locally aggressive and slow growing tumor, it is mostly found in the mandible, accounting for more than 70% of all cases, followed by the maxilla and rarely found in the paranasal sinuses with 55 reported cases in the literature until February 2013 [1–4,8].

No specific etiology has been attributed to the development of ossifying fibroma. However, it is suggested that developmental abnormality, odontogenic or traumatic origins could be some underlying causes [1,8]. This could explain that the possible origin of the ossifying fibroma lesion in our patient was from the implanted mandible graft.

A variety of lesions, ranging from fibrous dysplasia, osteoma to ossifying fibroma could be classified under benign fibro-osseous lesions developing in the head and neck area. Moreover, a spectrum of histopathological variants of ossifying fibroma have been described; cemento-ossifying fibroma, aggressive psammomatoid ossifying fibroma and juvenile active ossifying fibroma [3].

Manes et al. reviewed 55 cases of ossifying fibroma in the paranasal sinuses and revealed that the male to female ratio with this type of tumor tends to be 1:1.04 with a mean age of 29.9 years [3].

Patients with ossifying fibroma tumors are often symptomless. They are mostly discovered incidentally when imaging is done for other indications. When they present with mass effect symptoms, facial swelling tends to be the most common sign followed by nasal obstruction symptoms, headache, proptosis and occular symptoms such as visual loss and diplopia [1,3,9].

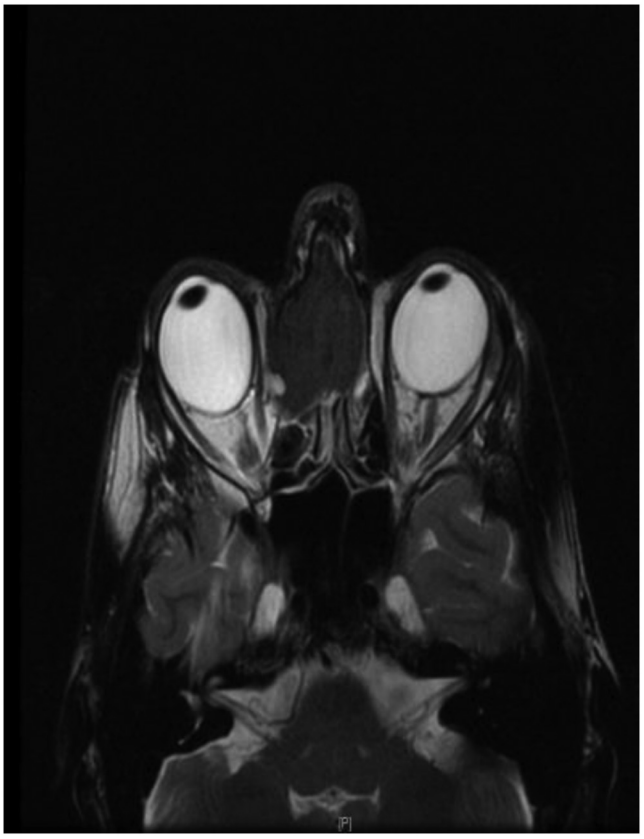

Fig. 2. The well-demarcated bony wall which is hypointense to soft tissue on axial MRI T2-weighted image. B: MRI images demonstrate right ethmoid lesion with mass effect and ethmoid air cells primary tumor.

Diagnosis of such mass is mostly done by radiographic images showing a well-circumscribed demarcated lesion. Initially, the lesion is radiolucent but progresses to radiopaque and gets surrounded by a radiolucent or sclerotic periphery. Paranasal CT scan presents the central areas with a non-homogeneous matrix including a ground-glass opacification which represents diffuse calcifications and areas of fibrous tissue or retained mucus. Further remodeling and thickening might be seen in the walls of the involved sinuses. Other diagnostic tools such as histopathological examination and MRI might be needed to confirm the diagnosis or to rule out intraorbital or intracranial extension [1,8].

The mainstay treatment of a paranasal sinus ossifying fibroma is total excision of the lesion by different modalities. Endoscopic resection of the tumor is the recommended therapeutic approach. It holds high advantages from direct visualization of the lesion to decrease in external deformity and morbidity. Other approaches such as craniofacial (craniotomy, transfacial or transoral) ones could be done. Although radiation therapy has been advocated in the treatment of bony tumor, it has not been advocated in the treatment course of ossifying fibroma [3]. Most common complication one might face in such operation is cerebrospinal fluid leakage which is mostly repaired intraoperatively. With a mean follow up time of 26 months, the recurrence rate of paranasal sinus ossifying fibroma was 7% with total resection compared to 25% with subtotal resection [3]. A 6 months follow up of our patient reveals no signs or symptoms of recurrence by physical examination and nasal endoscopy. A paranasal CT scan is planned in one year from the time of surgery. Therefore, long term follow-up with repeated nasal endoscopy and serial paranasal sinus CT is crucial to detect any tumor recurrence and prevents any possible morbidity [1,3]. No clear data in the literature reporting the malignancy transformation rate.

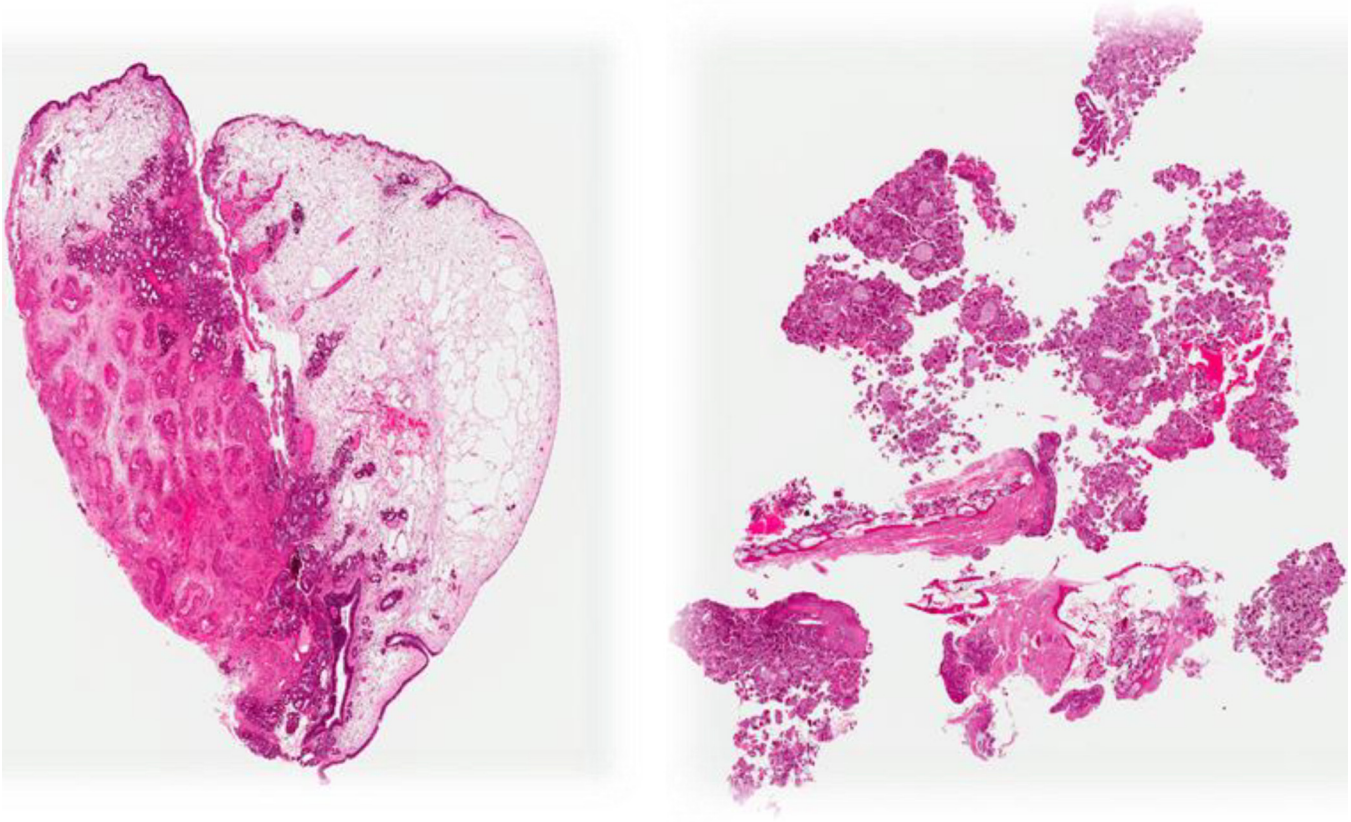

Fig. 3. Histopathologicaly, this lesion is characterized by spindle cell proliferation packed with globular cementum droplets and round/ovoid fragments of woven bone. These spindle cells appear benign and associated with psammomatoid cementum droplets and some woven bone and consistent with aggressive ossifying fibroma, psammomatous type IV.

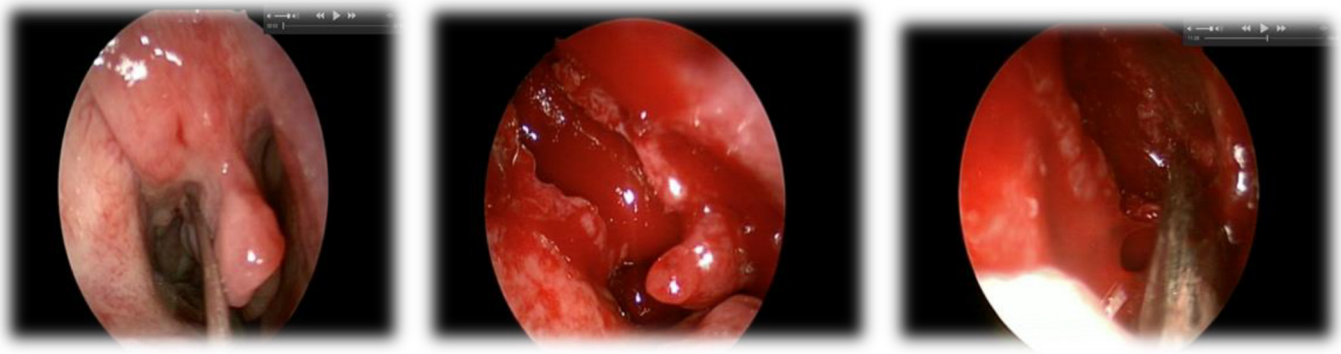

Fig. 4. Intraoperative pictures of right ethmoid tumor resection.

4. Conclusion

Ossifying fibroma is a rare benign fibro-osseous lesion seen in the bones of the head and neck area. The case presented here highlights an especially rare situation of this tumor involving the right ethmoidal sinus after implantation of this possible lesion from the mandible. Therefore, a careful and precise imaging should be done before implanting mandible graft. Also, it is very important to be aware of the clinical, imaging and histopathological features of ossifying fibroma in this uncommon site for better diagnosis and treatment of the different craniofacial tumors. Furthermore, implanted graft should be carefully inspected for such a possible rare lesion.

Competing interests

The authors declare that they have no competing interests.

Sources of funding

No source of sponsorship.

Ethical approval

Research has been approved by the Research Ethical Committee of King Abdullah International Medical Research Center.

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.

Author contribution

Salwa AlRashed ALHumaid: Data collection, data analysis & interpretation and writing the paper.

Yazeed AlGhonaim: Study concept & design, data collection, data analysis or interpretation, writing the paper.

Abdullah Arafat: Data analysis & interpretation and writing the paper.

Registration of research studies

Non Applicable.

Guarantor

Yazeed ALGhonaim.
Salwa ALRashed ALHumaid.

References

[1] Zhaleh Mohsenifar, Saeedallah Nouhi, Fatemeh Mashhadi Abbas, Sareh Farhadi, Behzad Abedin, Ossifying fibroma of the ethmoid sinus: report of a rare case and review of literature, J. Res. Med. Sci. 16 (June (6)) (2011) 841–847.
[2] Y.C. Choi, E.J. Jeon, Y.S. Park, Ossifying fibroma arising in the right ethmoid sinus and nasal cavity, Int. J. Pediatr. Otorhinolaryngol. 54 (August (2–3)) (2000) 159–162.
[3] R.P. Manes, M.W. Ryan, P.S. Batra, et al., Ossifying fibroma of the nose and paranasal sinuses, Int. Forum Allergy Rhinol. 3 (2013) 161–168.
[4] Anirban Chatterjee, Neha Ajmera, Amit Singh, Peripheral cemento-ossifying fibroma of maxilla, J. Indian Soc. Periodontol. 14 (July–September (3)) (2010) 186–189.
[5] J.C. Martins Junior, F.S. Keim, M.S. Kreibich, Peripheral ossifying fibroma of the maxilla: case report, Int. Arch. Otorhino-laryngol. 12 (2008) 295–299.
[6] I. Baumann, R. Zimmermann, F. Dammann, M.M. Maassen, Ossifying fibroma of the ethmoid involving the orbit and the skull base, Otolaryngol. Head Neck Surg. 133 (1) (2005) 158–159 [PubMed].
[7] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, the SCARE Group, The SCARE Statement: Consensus-based surgical case report guidelines, Int. J. Surg. 34 (2016) 180–186.
[8] S.Y. Lam, N.M. Ramli, D. Harikrishnan, S.F. Sia, J. Pailoor, Ossifying fibroma of the occipital bone—a case report and literature review, Eur. J. Radiol. Extra 67 (1) (2008) 19–23.
[9] Shekhar Charavarty, Debashish Datta, Esha Boro, Bijita Dutta, Ossifying fibroma in the sinonasal tract: a rare case report, J. Clin. Diagn. Res. 8 (August (8)) (2014) FD09–FD10.