

Case Report

Juvenile Trabecular Ossifying Fibroma (TrJOF) Originating from Lateral Fronto-Orbital Bone with Secondary Aneurysmal Bone Cyst (ABC) Formation: Report of an Unusual Case and Literature Review

Isra Abdulla¹, Ali Basalamah¹, Omar alwhaibi², Ammar Alrikabi³, Ahmad Al Boukai⁴, Sherif Elwatidy¹

¹Department of Surgery, Division of Neurosurgery, King Saud University, Medical City

²Department of Surgery, Division of Neurosurgery, King Saud University

³Department of pathology, King Saud University

⁴Department of Radiology, King Saud University

***Corresponding Author:** Isra Abdulla, Neurosurgery Intern, Department of Surgery, Division of Neurosurgery, King Saud University, King Saud University Medical City, Riyadh, Saudi Arabia

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Abstract:

Juvenile trabeculated Ossifying Fibroma (JTrOF) is commonly arising from the maxilla and the mandibular bones, while Psammomatoid Juvenile Ossifying Fibroma (PsJOF) commonly arises in the sinonasal and calvarial bones. In our paper, we described a 12-year-old pediatric female who

presented with a right fronto-orbital disfiguring swelling. She underwent total resection, peripheral osteotomy, and reconstruction. Pathology reported a TrJOF associated with Aneurysmal Bone Cyst (ABC) formation. TrJOF rarely originates from superolateral wall orbital bone, and it is exceedingly rare when it is associated with ABC formation. Pathologically, it is

difficult to distinguish TrJOF from other FOLs due to similarities and overlap. The ideal treatment is total surgical excision and reconstruction. The Surgical resection is challenging as regards functional and cosmetic outcomes.

Keywords: Juvenile ossifying fibroma; Aneurysmal bone cyst; Trabecular ossifying fibroma; Orbital tumors; Craniofacial surgery

Abbreviations: FOLs- Fibro-osseous lesions; TrJOF: Trabecular juvenile ossifying fibroma; PsJOF- Psammomatoid juvenile ossifying fibroma; JOF- Juvenile ossifying fibroma; OF- Ossifying fibroma; COF- cemento-ossifying fibroma; FD- Fibrous dysplasia; OFD- Osteofibrous dysplasia; ABC- aneurysmal bone cyst.

1. Introduction

Juvenile trabeculated Ossifying Fibroma (TrJOF) is one of the benign fibro-osseous lesions (FOLs) that may occur anywhere in the skeleton predominately affecting jaw bones (the maxilla and the mandible “gnathic”). It is extraordinarily rare that (TrJOF) involves the paranasal sinuses, orbital bones, and other extra-gnathic bones [1-3]. An aneurysmal bone cyst (ABC) is an expansile blood-filled multi-cystic lesion that commonly affects children. It's arising de novo in most cases or secondary to another osseous lesion [4, 5] Herein, we report an exceedingly rare case of TrJOF originating from the fronto-orbital bone (calvarial) with a secondary ABC formation.

2. Case Report

A 12-year-old girl presented with a 1-year history of right fronto-orbital swelling involving the superior and lateral borders of the orbit. The lesion was slowly increasing in size over 2 years then stabilized. The patient did not have any previous history of trauma and did not report any visual problems. Physical examination revealed a right orbital disfiguring swelling (Figure 1-A), hard in consistency and tender when exerting pressure over it, otherwise, the examination was unremarkable with no neurological deficits. Computed tomography (CT) scan of the Brain and Orbit showed an expansile multiseptated bony lesion in the right frontal bone extending into the right orbital superolateral wall. The lesion measures approximately 4 x 3.6 x 4.2 cm (AP, transverse and craniocaudal dimensions). It exerts a mass effect on the right orbital structures with subsequent proptosis. Moreover, a marked thinning of the cortex of the involved bone with areas of cortical breach intracranially and intraorbitally (Figure B, C, D) Magnetic resonance imaging (MRI), the tumor appeared multicystic, well-circumscribed with small areas of fluid-fluid levels (Figure 1-E, F). focal osteoblastic and osteoclastic activity with osteoid formation seen at the periphery as well. Postcontrast images showed diffuse intense heterogeneous enhancement with areas of high perfusion on rCBV and few small foci of diffusion restriction. The differential diagnosis based on radiological findings were fibro-osseous tumors including (osteo-fibrous dysplasia, fibrous dysplasia, ossifying fibromas, and osteoblastoma), intraosseous hemangioma, and aneurysmal bone cyst.

2.1 Surgical Technique

After taking consent from the family, the patient was taken for total resection and reconstruction. Steps of surgery are described below.

1. General anesthesia, supine position, head fixed in a 3- point fixator and rotated to the left about 40 degrees with slight extension.
2. Curvilinear skin incision extending from the just above the zygomatic arch half a cm anterior to the tragus, extending upward behind the hair line to cross the midline, Figure 2-A.
3. Skin, temporalis fascia and pericranium were elevated as one flap, temporalis muscle detached subperiosteally. We exposed the frontal, temporal bone, and orbit, we dissected the periorbital fascia completely from the orbital bones.
4. We did a strip craniotomy around the tumor (leaving a thin rim of healthy bone around the tumor), Figure 2 B.
5. The dura was carefully and completely dissected from the involved bone.
6. With the oscillating saw and craniotome we removed the whole tumor en-bloc Figure 2- C, the resected part included the superior and lateral orbital walls with the superior and lateral orbital rim

7. We used split calvarial bone grafts fixed together with long titanium microplate and screws to reconstruct the superior and lateral rim of the orbit, it took a lot of efforts and trials to achieve symmetry of both orbits.
8. We used titanium mesh to cover defects in the roof and lateral wall of the orbit as well as the skull defect, Figure 2-D. The Titanium mesh was finally covered with bone cement (Hydroxy Apatite) and contoured to merge with the normal bone, Figure 2-E.
9. The wound was closed in layers as routine with suction drain, Figure 2-F.

The final histopathology report was consistent with trabeculated ossifying fibroma with a secondary multiloculated aneurysmal bone cyst. In addition, there was a focal osteoblastic and osteoclastic activity with osteoid formation at the periphery of the lesion Figure-3.

Postoperative course was uneventful with no postoperative complications. Postoperative CT scan showed no residual tumor and satisfactory reconstruction, Figure-4. One year after surgery, the patient was doing well, there was no delayed complications and she was happy with the cosmetic outcome, Figure 5.



Figure. 1-A: photograph for the patient before surgery showing the tumor.

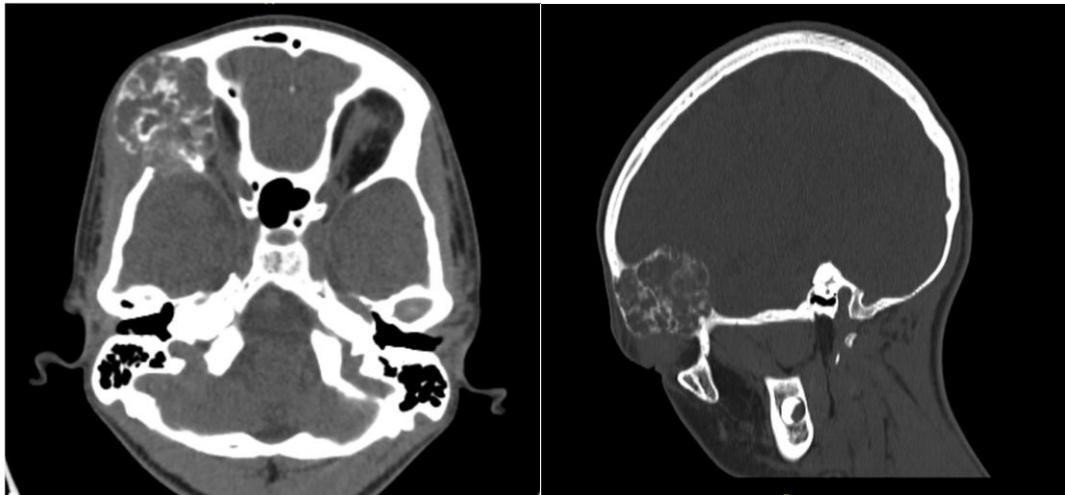


Figure-B

Figure- C



Figure-D

Figure.1- B, C, D: CT scan of the brain, axial, sagittal, and coronal cuts respectively (bone window at the level of the orbit), showing the expansile bony lesion at the superior and lateral aspect of the orbit with cortical thinning and faint ground glass density and trabeculated calcification.

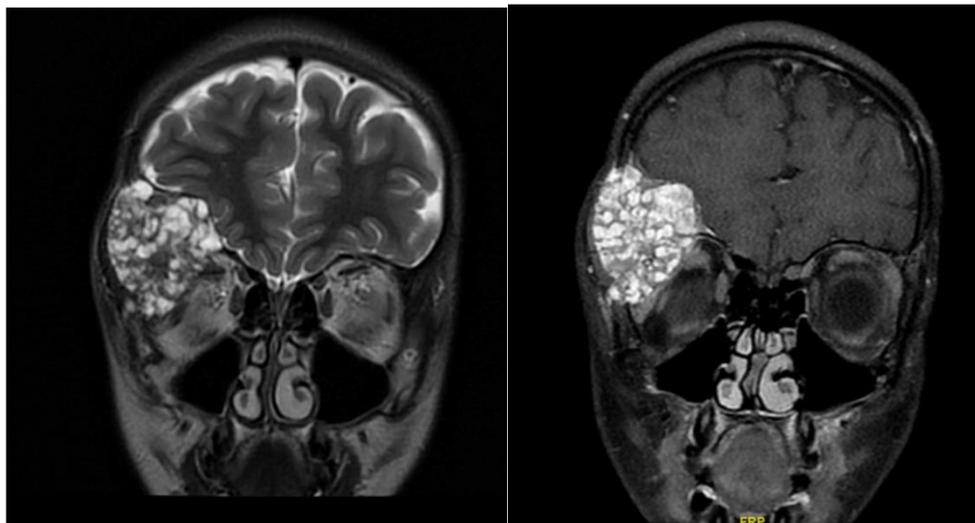


Figure-E

Figure-F

Figure 1-E, F: MRI of the brain T2 coronal and T1 coronal after gadolinium injection, showing the scattered high T2 Cysts-like lesion with central scattered areas of low T2 and the lesion shows septal and peripheral enhancement post-contrast. there is no sign of local invasion.



Figure 2-A: Intraoperative picture with the head in 3-point fixator and incision line.

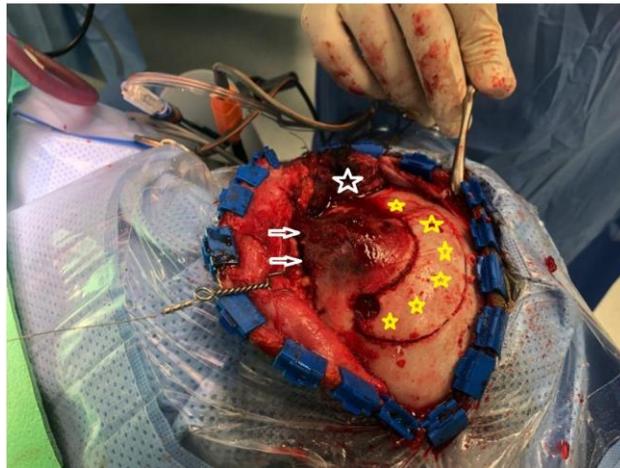


Figure 2-B: Skin flap is reflected, temporalis muscle detached subperiosteally and retracted inferiorly (white star), orbital fascia dissected superior and lateral orbital rim are exposed (white arrows). Strip craniotomy around (yellow stars) the tumor is performed.



Figure 2-C: Tumor resected totally.



Figure 2-D: Intra-operative photograph showing:

- Reconstruction of superior and lateral orbital rim using split calvarial graft (blue line).
- Reconstruction of skull defect using titanium mesh (green star).
- Strip craniotomy (white star).

-Globe (yellow arrow).



Figure 2-E: Skull & orbit after completion of reconstruction, N.B. the blue lines over the superior and lateral orbital rims.



Figure 2-F: Immediate postoperative photograph demonstrating acceptable symmetry of both orbits after.

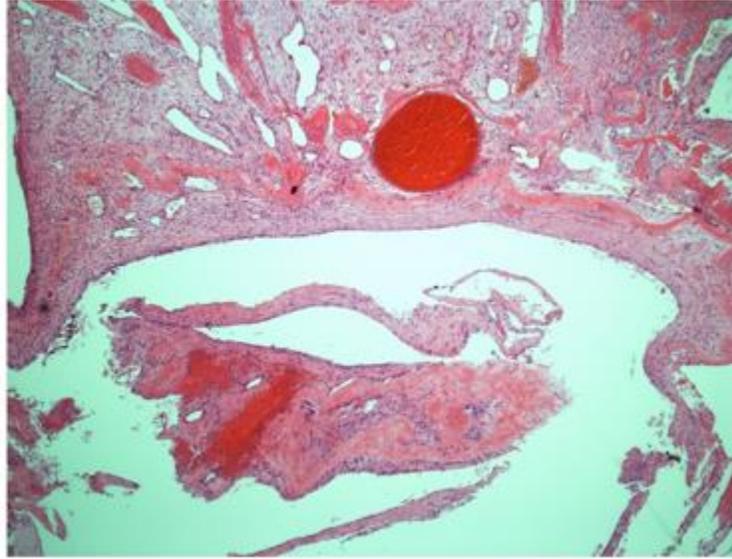


Figure 3-A: Pathology slide, low power view of the lesion (H&E stain x40) showing a large fibrous walled cystic space with surrounding congested blood vessels and bone trabeculae.

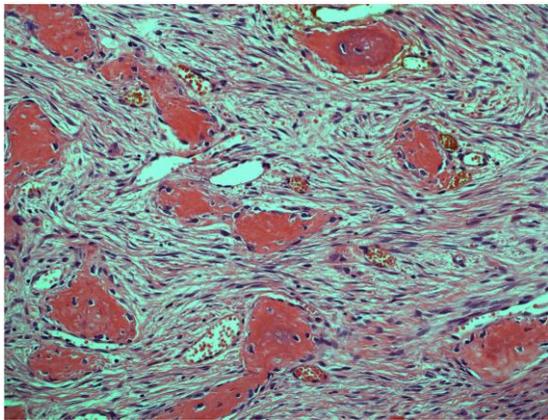


Figure-B

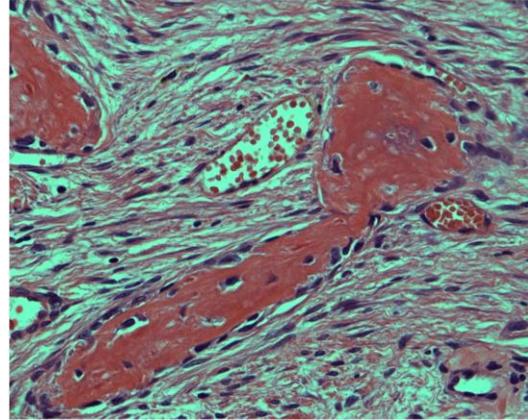


Figure-C

Figure 3-B, C: Bone trabeculae with the osteoblastic periphery and surrounding fibroblastic spindle cell proliferation on both (H&E stain x200) and (H&E stain x400) respectively.

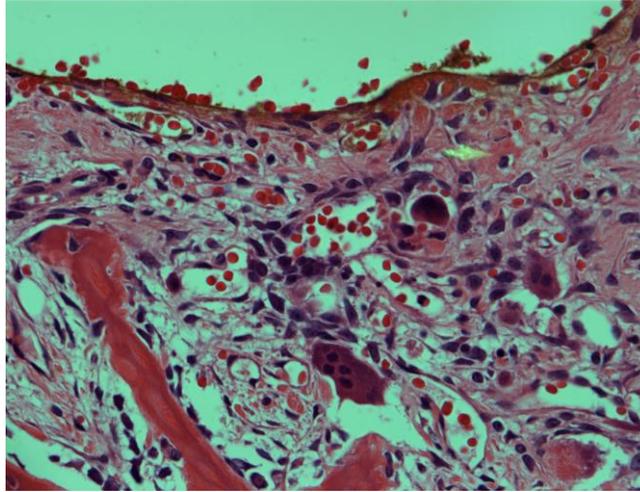


Figure 3-D: High power microscopic view shows the wall of the aneurysmal bone cyst as well as the presence of osteoid material and several giant cells.

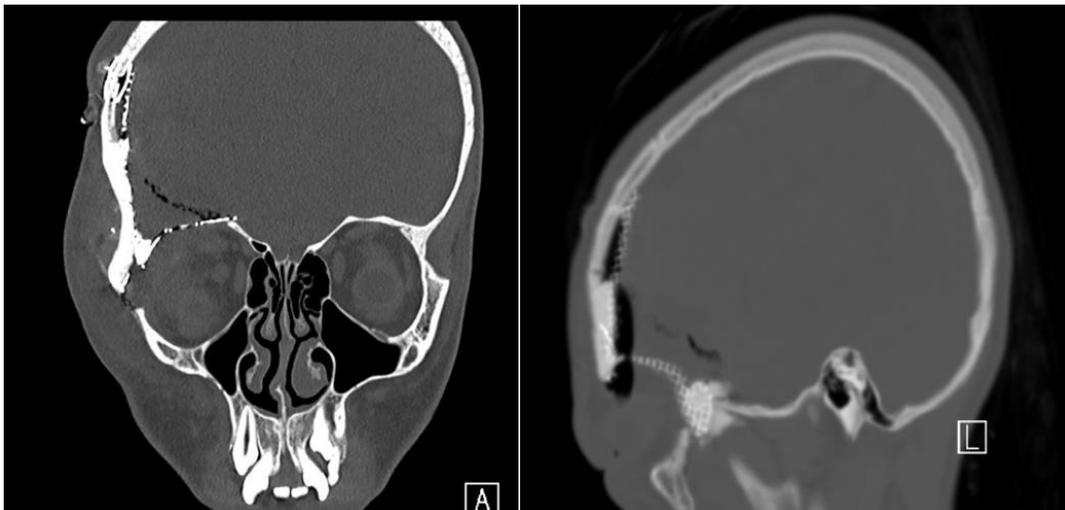


Figure-A

Figure-B

Figure 4-A, B: Postoperative CT scan (bone window) , coronal and sagittal reconstruction images showing complete excision of the tumor.

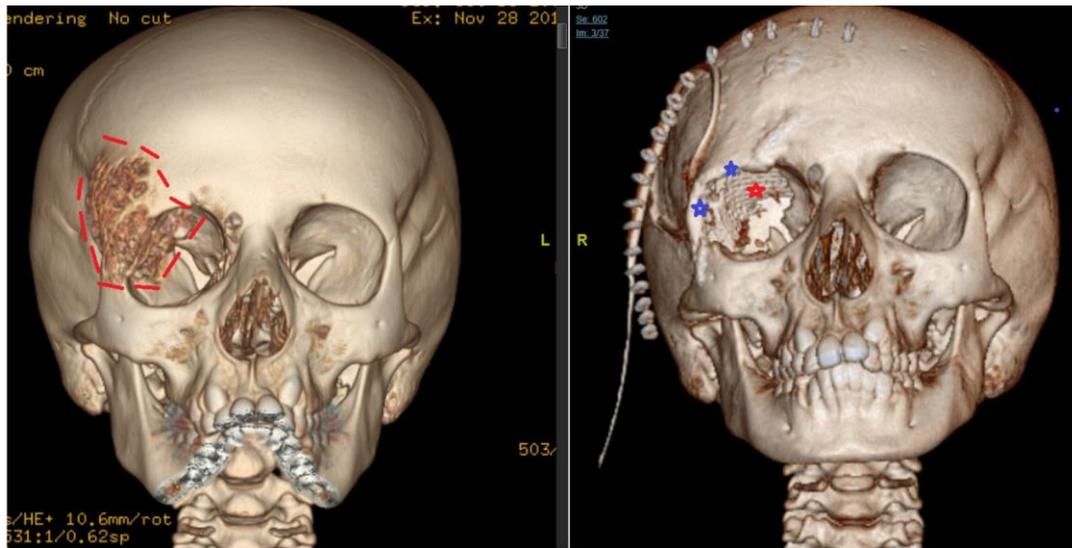


Figure 4-C: CT scan, 3-D reconstruction before and after surgery demonstrating.

- 1- Area resected (outline in red dashed line)
- 2- Reconstruction of superior and lateral orbital rim (blue stars)
- 3- Reconstruction of the orbital roof with titanium mesh (red star)

3. Discussion

Fibro-osseous lesions (FOLs) are a group of benign conditions arising when the bone is replaced by connective tissue. In 2008, Eversole et al classified FOLs of the craniofacial complex into five categories comprising; bone dysplasias, cemento-osseous dysplasias, inflammatory/reactive processes, metabolic disease, and neoplastic lesions. The juvenile ossifying fibroma (OF) was considered a subtype of neoplastic lesions [6]. According to the 4th edition of WHO classification of the head and neck tumors which was released in 2017, OF was classified under the fibro-osseous and osteochondromatous lesions subcategory of the Odontogenic and maxillofacial bone tumors, furthermore, OF can be subclassified into Juvenile Ossifying Fibroma and cemento-ossifying fibroma [7].

OF is thought to arise from mesenchymal blast cells. It is seen in pediatrics and adults with a female predilection [8]. Juvenile Ossifying Fibroma (JOF) is a term used to describe the OF variant occurring in young patients with a mean age of 11 years old. It typically involves craniofacial bones, and it comprises two distinct histopathological types including psammomatoid juvenile ossifying fibroma (PsJOF) and TrJOF [1, 9-11]. In literature, TrJOF was first described in 1965 by Reed et al under the general name of JOF while PsJOF was 1st described by Benjamin in 1938 [9]. It is usually presenting at an earlier age on average being 8-12 years while PsJOF presenting on average between 16-33 years[1, 2, 9]. The cemento-ossifying fibroma (COF) was 1st described by Menzel

in 1872. COF is peaking at the 3rd to 4th decades of life with female predominance [10].

In the craniofacial bones, TrJOF predominately arises from the maxilla and mandibular areas (gnathic), and it's extremely rare to arise from any other extragnathic areas [9, 12]. Table 1 summarizes all reported extragnathic cases of Tr JOF.

In the presented case the TrJOF originated from the lateral fronto-orbital bone wall.

A wide range of differential diagnoses was proposed based on the patient's initial clinical and radiological presentation including OF, fibrous dysplasia (FD), ABC, osteoid osteoma, osteblastoma, and osteosarcoma. The initial frozen section that was sent intraoperatively reported as FD. The final histopathological report was JOF of the trabecular subtype (TrJOF) with an associated ABC. Ossifying fibroma, FD, and osteofibrous dysplasia (OFD) are closely related, all are benign proliferative FOLs in which there is a replacement of normal bone with fibrous tissue [13]. They overlap clinically, histologically, and radiographically, with some key differentiating features.

Radiologically, FD typically exhibits a characteristically diffused ground-glass appearance that merges with the normal surrounding bone. It is polycystic and usually has an irregular border and it may present as completely radiolucent or homogeneously sclerotic lesions. On contrary to the FD, TrJOF, PsJOF and COF are monostotic and are usually associated with bony expansion, well-defined

borders with central mixed lytic and osteoblastic areas, and spherical architecture, while FD maintains the same architecture of the involved bone. The ground-glass appearance is also seen occasionally with PsJOF, while TrJOF lacks this feature and manifests as scattered trabeculated calcifications within a primarily radiolucent structure [9]. Cemento-ossifying fibroma is usually not destructive and has an intact cortex on imaging while JOF typically demonstrates cortical thinning, disruption, and/or infiltration of surrounding tissue due to its more locally aggressive nature [7]. Furthermore, a hyperdense center surrounded by hypodensity (target-like appearance) may be seen in OF especially PsJOF and COF [14]. On MRI, OF is usually iso to hypo intense on T1 and has variable intensity on T2. Cysts, non-ossified regions, and mucocele are associated usually with a high T2 signal. Post-contrast administration, an enhancement is expected especially for the outer lesional layer and the septa, and due to the high osteoblastic activity, and the lesions demonstrate high Technetium-99 radioisotope uptake [7, 15]. On the other hand, FD usually has a low signal intensity on T2 weighted MRI and enhancement is seen on the expanded diploe only [10, 15].

Histopathologically, the FOLs may have similar microscopic features including the increase of fibroblastic-stroma, variable degrees of osteoid or cementum-like tissue, and variable degrees of calcified structures [10]. On the otherhand, FD is commonly described with the appearance of chine letters due to the thin curved bone trabeculae interspersed with fibroblast-like spindle cells, but it characteristically lacks the osteoclastic multinucleated giant cells seen in TrJOF along with anastomosing

trabeculae of woven mature lamellar bone that is lined with osteoclasts and osteoblasts. Furthermore, FD has no osteoblasts rimming, less stromal cellularity, collagen, less vascularity, and maintains a similar pattern throughout the lesion. Both subtypes of JOF are unencapsulated infiltrating normal surrounding bone, with PsJOF having a typical curved or spherical ossicle (psammomatoid calcifications), while COFs are encapsulated, rimmed with osteoblasts, and have a variable ratio of bone/cementum-like tissue [7-9, 16]. Presence of areas of ABC in association with these FOLs may indicate rapid growth [17].

From a genetic point of view, FD has been proven to have a characteristic α subunit mutation of single-transducing G protein, which is lacking in OFs. PsJOF may have chromosomal breakpoints at Xq26 and 2q33 leading to (X;2) translocation. Few studies reported HRPT2 gene and germ-line mutation in COF. Although genetic studies of TrJOF are generally not well understood, MDM2 gene amplification was found in both TrJOF and PsJOF [16, 18].

The clinical course is another key aspect to differentiate JOFs. The majority of benign FOLs demonstrate slow progressive non-infiltrative mass effect over years. JOFs have an aggressive nature causing facial deformity occasionally. The initial asymptomatic growth may rarely exhibit symptoms. Other than local pain, paresthesia and deformity, symptoms are primarily related to the primary site of origin and invaded structures including but not limited to visual loss, proptosis, nasal blockage, anosmia, and hearing disturbance [1, 3, 19] The aggressive nature may be attributed to the young age of onset and the

association of ABC. Continuous growth is expected if JOF is not treated [17]. Contrary to JOF, COF demonstrates a slow rate of growth and no reports of recurrence of malignant transformation [7, 10]. Several Investigators reported that OF originating from paranasal sinuses areas tend to be more aggressive when they are compared to those originating from the mandibular area while others have concluded a lack of relationship between location and aggressiveness [20].

FD, on the other hand, usually spontaneously regresses at puberty and can be observed if no local complications are associated. Table 3, summarizes radiological, histopathological, and genetic differences between fibrous FD and subtypes of OF.

A proper initial diagnosis is suggested to guide the treatment plan⁷. Complete surgical excision of OF is recommended by the majority of authors [21]. According to a recent systematic review reported by Chrcanovic et al in 2019, enucleation and curettage alone are associated with a high recurrence rate, and this risk is significantly reduced when adjunctive management is added such as ostectomy. This study found that the treatment of choice for both JOF subtypes should be enucleation with peripheral osteotomy as it is associated with complete resection that renders recurrence [17]. Conservative approaches for lesions behaving benignly and not producing deformities such as curettage and osteotomy may be attempted as 1st line management for these lesions [7, 22] The risk of recurrence for OF is considerably high ranging between 30-56% and this risk is significantly

present in cases of incomplete resection [3, 20]. JOF may require removal multiple times to reach a cure. No malignant progression was reported to date [10, 23] On the other hand, COF tends not to recur after removal [7]. Interferon-alpha systemic therapy use was reported to decrease the rate of recurrence while radiotherapy is contraindicated due to the risk of malignant transformation [9]. In our case, upfront total resection, peripheral osteotomy, and reconstruction were attempted due to a relatively fast-growing rate (less than a year), painful locally deforming nature, and controversial radiological diagnosis.

ABC is another benign FOLs that is osteolytic, expansile in nature with connective tissue septations in between blood-filled spaces [4]. ABC may arise primarily or secondary to another bone lesion and it has been reported secondary to FD and JOF. The pathogenesis is not well understood. it was postulated

that the formation of an ABC starts as myxoid change (myxofibrous cellular stroma overproduction) within a hemorrhagic stroma with osteoclastic giant cells and reactive bone that is gradually expanding and forming multiple cysts with thin fibrous septae. Moreover, due to local pressure, intracellular edema, weak supporting tissue, and maldeveloped vascularity, cysts may form, and blood may be pooled into these cystic cavities following small vessels rupture [12, 24-26]. when OF is associated with a fluid-fluid level, ABC is strongly suggested. However, this sign may be associated with other lesions such as simple bone cyst, cavernous haemangioma, osteosarcomas, and others [7].

ABC associated with JOF was reported predominantly with psammomatoid variant of JOF, while the association with TrJOF were reported in a total of 8 cases so far that are summarized in Table 2 [10, 12, 25-29].

No. of cases reported	Reference / date	Trabecular Juvenile Ossifying Fibroma (TrJOF)	Location of origin
1	Thomas et al. [14]	TrJOF	Orbital roof
1	Nakagawa et al. [23]	TrJOF	Ethmoid sinus, orbit, and anterior cranial fossa
1	Fakadej et al [21]	TrJOF	Ethmoid sinus
1	Marvel JB et al. [30]	TrJOF	Ethmoid and sphenoid sinuses
1	Tunc M et al. [20]	TrJOF	Lateral orbital wall.
1	Caylakli F et al. [31]	TrJOF	Middle turbinate
2	Antonio et al. [11]	TrJOF	Orbital roof.
1	Yazici B et al. [10]	TrJOF	Lateral Orbit.
1	Bohn OL et al. [32]	TrJOF	Nasal cavity and anterior cranial fossa

1	B T Yang et al. [12]	TrJOF	Paranasal sinuses
1	Marian Christoph Neidert et al. [33]	TrJOF	Sella
1	Marglani OA et al. [34]	TrJOF	Nasal cavity and Paranasal sinuses
1	Owosho AA et al. [24]	TrJOF	Ethmoid sinus
1	Ciniglio Appiani M et al. [35]	TrJOF	Nasal cavity and Paranasal sinuses
1	Bipasha Mukherjee et al. [9]	TrJOF	Superolateral orbital wall.
1	Ta NH et al. [19]	TrJOF	Ethmoid sinus
Total number: 17			

Table 1: Confirmed Cases of Trabecular Juvenile Ossifying Fibroma (TrJOF) reported in the literature arising from the Skull excluding maxilla and mandible bones.

No	Reference / date	Diagnosis	Location of origin
1	Noffke CE et al. [27]	TrJOF and ABC	Mandible
1	Yazici B et al. [10]	TrJOF and ABC	Lateral Orbit wall.
1	Sankaranarayanan S et al. [28]	TrJOF and ABC	Maxilla.
1	Silva CA et al. [25]	TrJOF and ABC	Maxilla.
1	B T Yang et al. [12]	TrJOF and ABC	Paranasal sinuses
2	Urs AB et al. [29]	TrJOF and ABC	Maxillae and Mandible
1	Reddy AVS et al. [26]	TrJOF and ABC	Mandible.
Total number: 8			

Table 2: Confirmed Cases of Trabecular Juvenile Ossifying Fibroma (TrJOF) reported in the literature associated with aneurysmal bone cyst (ABC).

	FD	TrJOF	PsJOF	COF
	“polyostotic”	“monostotic”	“monostotic”	“monostotic”
Histopathology	Chinese letters appearance. Lack of osteoblastic rimming. Similar architecture to involved bone.	Un-encapsulated infiltrating surrounding bone. Osteoclastic multinucleated giant cells, anastomosed with trabeculated lamellar bone. Osteoblastic rimming.	Un-encapsulated infiltrating surrounding bone. Psammomatoid calcifications. Osteoblastic rimming.	Encapsulated. Variable ratio of bone/cementum tissue. Osteoblastic rimming.
Radiology	Diffused ground-glass appearance. Radiolucent or homogeneously sclerotic.	Scattered trabeculated calcifications within radiolucent structure. Locally aggressive with well-defined borders.	Target-like appearance, occasionally ground-glass. Locally aggressive with well-defined borders.	Target-like appearance. Non-destructive with intact cortex.
(MRI)	Low intensity (T1). Enhancement on expanding dipole (post-contrast)	Iso to hypo intense (T1). Variable intensity (T2). Enhancement of outer layers and septae (post-contrast)	Similar to TrJOF. High technetium-99 uptake. MDM2 gene amplification.	
Genetics	α Subunit Mutation of single-transducing G protein	MDM2 gene amplification	Chromosomal breakpoints at Xq26 and 2q33 leading to (X;2) translocation.	HRPT2 gene and germ line mutations
Course	Spontaneous regression at puberty	Locally aggressive. No spontaneous regression. May recur if incompletely resected.	Similar to TrJOF.	Slow growth rate. No recurrence or malignant transformation reported.

Table 3: Comparison between subtypes of ossifying fibroma (OF) and fibrous dysplasia (FD).

4. Conclusion

TrJOF commonly arises from maxilla and mandible, while PsJOF commonly arises in the sinonasal and calvarial bones. Pathologically, it is difficult to distinguish TrJOF from other FOLs due to similarities and overlap. The ideal treatment is total surgical excision and reconstruction. The Surgical resection is challenging as regards functional and cosmetic outcome.

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