



Fibrous dysplasia with secondary aneurysmal bone cyst—a rare case report and literature review

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Abstract

Introduction Fibrous dysplasia (FD) and aneurysmal bone cyst (ABC) are uncommon non-neoplastic intraosseous lesions, and the occurrence of concomitant FD and ABC is extremely rare.

Case report An 11-year-old boy presented with gradual progressive enlargement of his right zygomatic bone over 4 years prior to presentation. Computed tomography revealed a lesion with a central bony area showing a ground-glass appearance surrounded by a well-defined expansile lesion with internal septations. An incisional biopsy was performed and suggested a fibro-osseous lesion. Resection was performed, followed by immediate reconstruction using autogenous bone graft from the iliac crest. Histopathological examination revealed irregularly shaped trabeculae comprising immature woven bone in a fibroblastic cell-rich stroma. Blood-filled sinusoidal spaces lined by fibrous septa containing scattered multinucleated giant cells were observed peripherally. These findings were compatible with concomitant FD and ABC. The patient was disease-free at the time of his 10-month follow-up.

Conclusions Based on our literature search, this report is the first to describe concomitant monostotic FD and ABC in the zygomatic bone. Accurate diagnosis requires careful investigation and examination of clinical, radiographical, and histopathological features. The treatment of choice should provide an esthetic and functional improvement in the patient.

Keywords Fibrous dysplasia · Fibro-osseous lesions · Aneurysmal bone cyst · Craniofacial surgery

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Introduction

Fibrous dysplasia (FD) is a skeletal anomaly in which normal bone is replaced by poorly organized and inadequately mineralized immature bone and fibrous tissue. FD may affect any bone in the body; however, the ribs, the long bones, the pelvis, as well as the skull and the jaw are most commonly involved [1].

The disease may affect a single bone (monostotic) (representing most cases of FD) or multiple bones (polyostotic) [2]. The polyostotic type may be related to the McCune-Albright syndrome in which skeletal abnormalities are associated with characteristic “café-au-lait spots” and endocrine abnormalities, as well as the Jaffe-Lichtenstein syndrome in which patients show the same characteristics, except the associated endocrine abnormalities [3, 4].

In the craniomaxillofacial skeleton, FD more commonly affects the maxilla than the mandible and usually affects adjacent bones including the zygomatic, sphenoid, and the frontal bones. The main clinical characteristic of FD is painless swelling in the affected bones [5]. Bony overgrowth can cause facial asymmetry, disfigurement, malocclusion, and exophthalmos depending on the anatomical sites and structures involved [6]. Radiographically, the characteristics of FD vary from radiolucent (initial stages) to radiopaque (mature) lesions secondary to a combination of fibrous and osseous elements. The affected bone shows expansion and a ground-glass appearance is commonly reported in advanced stages [7].

An aneurysmal bone cyst (ABC) is an intraosseous osteolytic lesion that primarily involves the metaphysis of long bones and vertebrae [8]. An ABC rarely occurs in maxillofacial bones, and if it does, it usually presents as a primary type in this region. Secondary types of ABC are associated with well-recognized pre-existing bone lesions including osteoblastomas, ossifying fibroma (OF), and FD [9]. Radiographically, an ABC is characterized by uni- or multilocular radiolucent lesions with typically “ballooned” cortical expansion [10]. Histopathologically, an ABC is characterized by blood-filled spaces separated by fibrous septa containing osteoclast-like giant cells [9].

Concomitant FD and ABC lesions are rare in the head and neck region [11]. We performed a literature search in the PubMed/MEDLINE/Google Scholar databases, as well as the gray literature and identified 36 case reports describing the occurrence of concomitant FD and ABC in the skull bones (Table 1). However, no case report has described concomitant FD and ABC exclusively affecting the zygomatic bone. To our knowledge, this is the first report to describe a case of concomitant ABC and monostotic FD affecting the zygomatic bone.

Case report

An 11-year-old boy presented with gradual progressive enlargement of his right zygomatic bone, which was noticed

after a fall from a bicycle over 4 years prior to presentation. He reported that the lesion had doubled in size over 6 months prior to presentation, although this enlargement was not associated with pain, paresthesia, or functional disturbance. The patient denied systemic, metabolic, or endocrine disease. After clinical evaluation, preoperative and imaging examinations were performed.

Computed tomography (CT) revealed a bony lesion measuring approximately $3.0 \times 5.2 \times 4.9$ cm in the right zygomatic bone. The lesion showed a central area of bone density (400 HU) with a ground-glass appearance surrounded by a well-defined expansile lytic lesion with internal septations and low-attenuation content (2–25 HU), suggesting thinning of cortical bone (including the orbital region) (Fig. 1).

An incisional biopsy was suggestive of a fibro-osseous lesion. Surgical removal was planned using a three-dimensional (3D) printed biomodel developed by Compass Diagnóstico, Planejamento e Prototipagem Odontológica LTDA (Fig. 2a). Significant bone involvement necessitated zygomatic bone resection performed through the Weber-Ferguson incision (Fig. 2b). Simultaneous reconstruction was performed using a previously harvested autogenous bone graft from the iliac crest, which was fixed using low profile 1.5-mm titanium miniplates (Fig. 2c). The soft tissue incisions were sutured using 5-0 monofilament nylon. Despite the proximity to the orbit, the patient’s vision remained unaffected, and he was discharged from the hospital a few days later, without complications.

Histopathological evaluation of the resected specimen showed a central area of well-defined whitish colored mixed tissue with large cystic spaces observed peripherally. The remaining cortical bone showed marked thinning (Fig. 3a). Further evaluation revealed irregularly shaped trabeculae comprising immature woven bone in a fibroblastic cell-rich stroma. Blood-filled sinusoidal spaces lined by fibrous septa containing scattered multinucleated giant cells were observed peripherally. These findings were compatible with concomitant FD (Fig. 3b) and ABC (Fig. 3c).

Subsequent postoperative follow-up revealed an excellent cosmetic result. The patient was without recurrence at the time of his 10-month follow-up. A CT scan showed zygomatic bone reconstruction without evidence of relapse (Fig. 4).

Discussion

Both FD and ABC are uncommon non-neoplastic intraosseous lesions, which characteristically occur in young patients [9]. The concomitant occurrence of these lesions can be explained by the probable pathogenesis of ABC. ABC formation could be attributed to a local circulatory disturbance causing a markedly elevated venous pressure and the development of enlarged vascular spaces within the affected bone

Table 1 Results of a literature search showing cases with concomitant fibrous dysplasia and aneurysmal bone cyst involving the skull bones

Authors	Cases	Site	Age	Gender
Arden et al., 1997 [12]	1	Mandible, ethmoid and sphenoid sinuses, clivus, petrous apex, pterygoid bone, sphenoid wing, and maxilla	19	F
Birk et al., 2017 [13]	1	Parietal	23	M
Branch et al., 1986 [14]	1	Frontal and parietal	9	F
	1	Parietal	19	M
Buraczewski et al., 1971 [15]	1	Mandible	26	F
Dani et al., 2016 [16]	1	Frontal and ethmoid	23	M
El-Deeb et al., 1980 [17]	1	Mandible	19	M
Elleuch et al., 2016 [18]	1	Parietal	16	F
Haddad et al., 1998 [19]	1	Temporal, middle cranial fossa, sphenoid	6	M
Iseri et al., 2005 [20]	1	Clivus, temporal and occipital	35	F
Itshayek et al., 2002 [21]	1	Clivus, petrous apex and occipital	19	M
Lee et al., 2010 [11]	1	Fronto-parietal region	18	F
Lee et al., 2014 [22]	1	Zygomatic, orbit and maxilla	–	M
Lin et al., 2004 [23]	1	Frontal	18	M
Lucarelli et al., 1995 [24]	1	Orbit	19	M
Malik et al., 2006 [25]	1	Anterior cranial fossa, nasal cavity and orbit	13	M
Manjila et al., 2013 [26]	1	Nasal cavity, ethmoidal sinuses, orbital wall, and anterior cranial fossa	10	M
Mattei et al., 2005 [27]	1	Occipital and parietal	19	F
Moorthy et al., 2015 [28]	1	Ethmoid, frontal, maxillary, and sphenoid	19	F
Ngo et al., 2015 [29]	1	Sphenoid and cavernous sinus	15	F
Oliver et al., 1973 [30]	1	Mandible	20	F
Pasquini et al., 2002 [31]	1	Maxillary sinus, nasal fossa, and orbit floor	5	–
Rappaport et al., 1989 [32]	1	Occipital	25	M
Salmasi et al., 2011 [33]	1	Mandible, sphenoid, sella turcica, clivus, nasal cavity, paranasal sinus	16	M
Skladzierin et al., 2008 [34]	1	Nasal cavity, paranasal sinus, orbits, and anterior cranial fossa	16	M
Suzuki et al., 2001 [35]	1	Maxillary sinus	23	M
Terkawi et al., 2011 [36]	1	Sphenoid and ethmoidal sinuses, nasal cavity, and maxillary sinus	7	F
Urgun et al., 2016 [37]	1	Occipital, parietal, and orbital roof	14	F
Wojno et al., 1994 [38]	1	Temporal and middle cranial fossa	14	F
	1	Maxillary and ethmoidal sinus	22	F
	1	Mandible	19	M
	1	Sphenoid	22	M
Yokoyama et al., 1988 [39]	1	Frontal	40	M
	1	Parietal and occipital	29	M
	1	Paranasal and maxillary sinus	27	M
Yuen et al., 2002 [41]	1	Temporal, orbit, frontal, and sphenoid	22	M
Total	36		Mean 18.8	F 40% M 60%

M male, F female

[42]. This hemodynamic disturbance could be secondary to local trauma or the presence of a pre-existing lesion in the affected bone [43, 44].

Clinically, FD usually presents as slow-growing painless lesions, whereas an ABC is known to be a rapidly growing painful lesion [45]. Our patient reported trauma that occurred 4 years prior to presentation and had noticed accelerated growth of the lesion only in recent months. Therefore, we

concluded that the bicycle fall-related trauma was a mere coincidence and that the development of the ABC was secondary to a pre-existing lesion. Pre-existing lesions usually associated with an ABC are chondroblastoma, giant cell tumor, chondromyxoid fibroma, non-OF, and FD [46].

A few studies have reported concomitant FD and ABC affecting the skull bones. Our literature search performed in the PubMed/MEDLINE/Google Scholar databases and the

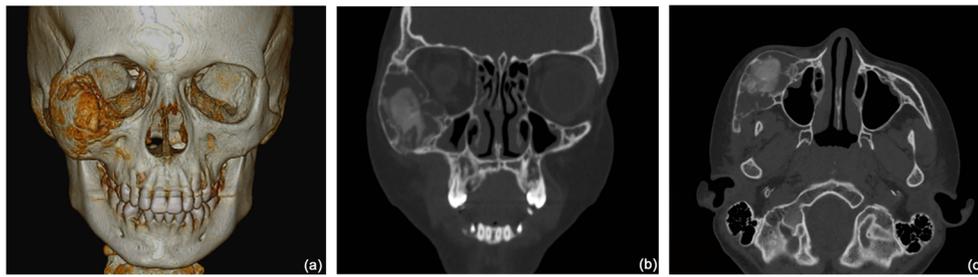


Fig. 1 **a** Three-dimensional computed tomography (3D-CT) reconstruction images show an expansile lesion affecting the right zygomatic bone. **b** and **c** Coronal and axial views show an expansile lesion with a central

ground-glass bony area surrounded by a well-defined expansile lytic lesion with internal septations

gray literature revealed 36 cases (Table 1), and most cases described involvement of the skull base [29, 33]. Only one case reported involvement of the zygomatic bone with the disease extending to the maxilla and the orbit. However, this case was of a patient with McCune-Albright syndrome showing the classical triad of polyostotic FD, “café-au-lait” skin macules, and endocrinopathies [47]. Thus, we conclude that this is the first report to describe a case of concomitant ABC and monostotic FD in the zygomatic bone.

Our literature search demonstrated that most patients showing concomitant FD and ABC are diagnosed in the second decade of life (mean age 18.8 years), compatible with the age observed in patients with isolated lesions of each disease [2, 42]. Moreover, men (60%) were more commonly affected than women (40%). Interestingly, isolated FD usually affects more women (ratio 2:1) [48] similar to isolated ABC (ratio 1.4:1) [49]. Our patient presented with characteristics that concurred with previous reports in the literature describing concomitant FD and ABC (Table 1).

CT is an important diagnostic modality to study these cases. CT helps to determine the extent of the lesion and the radiodensity of the FD in craniofacial bones thereby facilitating the development of an adequate diagnostic hypothesis [5]. The findings of FD can vary depending on the amount of bone present in the lesion, and FD may present as: cystic (radiolucent or lytic), sclerotic, and mixed (radiolucent/radiopaque) types [50]. Owing to the varying degrees of internal calcified or ossified matrix, FD may often be indistinguishable from other lesions including an OF [51]. Our patient showed a characteristic FD lesion with the most common pattern of mixed tissue with a central “ground-glass” appearance [7].

Peripherally, the lesion showed a multiloculated appearance with septations and soap-bubble features suggestive of an ABC [10, 42]. The multiloculated appearance was attributed to the blood-filled cavernomatous spaces surrounded by connective tissue and bone [29]. CT and magnetic resonance imaging show the presence of fluid-fluid levels, which helps in the diagnosis [42]. In our patient, these fluid-fluid levels

Fig. 2 **a** Three-dimensional (3D) biomodel used to perform preoperative planning. **b** Images show that the lesion is accessed through a Weber-Ferguson incision. **c** Reconstruction of bone is observed using autogenous bone graft from the iliac crest fixed with titanium miniplates

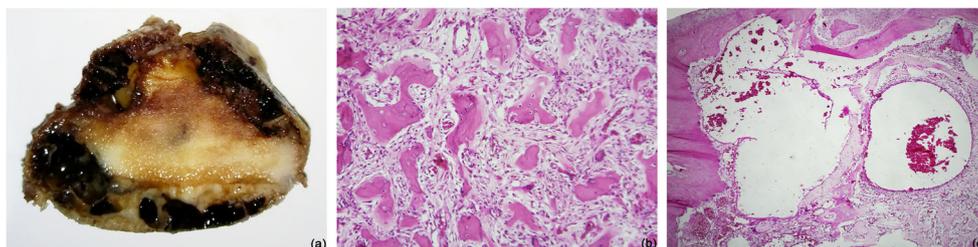
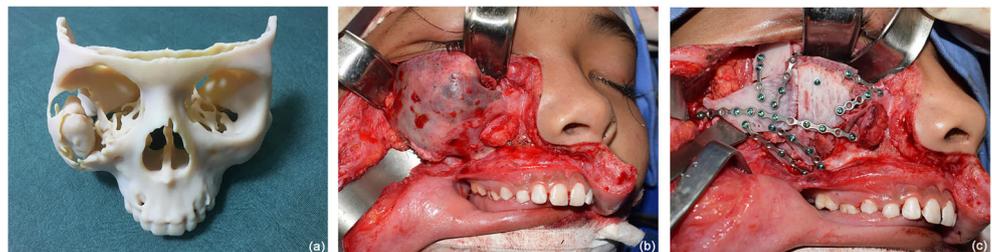
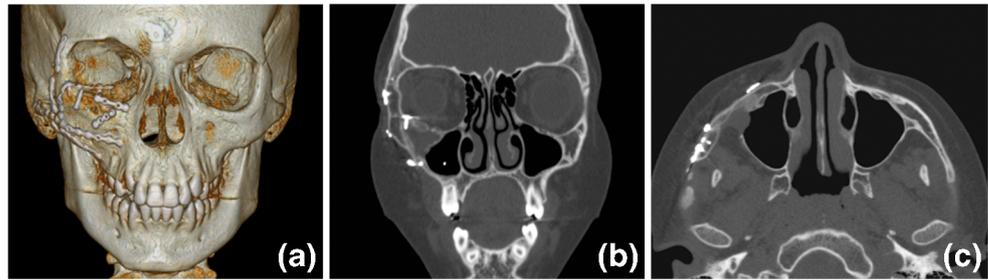


Fig. 3 **a** Histopathological examination of the resected surgical specimen shows a well-defined whitish colored fibrous area with peripheral blood-filled cystic spaces. **b** Irregularly shaped trabeculae comprising immature

woven bone in a fibroblastic cell-rich stroma characterizing fibrous dysplasia are observed. **c** Blood-filled sinusoidal spaces lined by fibrous septa, characteristic of an aneurysmal bone cyst are observed

Fig. 4 **a** Three-dimensional computed tomography (3D-CT) reconstruction images show post-operative results. **b** and **c** Coronal and axial views show restoration of facial symmetry, and no relapse is observed after 10-month follow-up



could not be viewed on CT images; however, the blood-filled spaces were clearly identified in the surgical specimen after removal of the central dysplastic tissue. The lesion in our patient showed a central ground-glass appearance with peripheral multiloculated areas of low attenuation, which concurs with the findings of previous studies that have reported concomitant FD and ABC involving the skull bones [33, 37, 38].

Several types of fibro-osseous lesions may be indistinguishable from FD histopathologically; therefore, microscopic examination alone cannot conclusively diagnose this condition and additional clinical, radiographic, and intraoperative examinations are often necessary [2]. In this patient, the osseous component showed a curvilinear trabecular bony arrangement characteristic of FD with occasional small spicules of woven bone, resembling a presentation of OF [2]. However, the predominant pattern was that of irregularly shaped trabeculae comprising immature woven bone without osteoblastic rimming in a fibroblastic cell-rich stroma—characteristics compatible with FD [52].

The management of craniomaxillofacial FD includes a “wait and see” strategy, or conservative/radical surgery [53]. In patients with concomitant lesions (for example, an ABC), the management is determined by the associated lesion [47]. Therefore, we performed complete resection owing to the risk of involvement of adjacent structures and the high rates of recurrence associated with ABC curettage [8].

There is a lack of consensus regarding the optimal treatment strategy; however, factors that must be considered in formulating a treatment plan include the site of involvement, rate of growth, esthetic complications, functional disruption, patient preference, general health of the patient, and the surgeon’s experience [54]. Regarding the site of involvement, a study by Chen et al. (2006) has recommended that lesions affecting the fronto-orbital, zygomatic, and the upper maxillary regions ought to be treated with total excision of the dysplastic bone owing to the risk of visual changes caused by displacement of the eyeball or optical nerve compression [54].

Currently, resection involves fewer risks because surgical planning now involves sophisticated modern techniques including assessment using a 3D biomodel as was used in our patient [55]. The use of this technique

enables more reliable interpretation of imaging data and improved preoperative planning, reduces the operative time, improves a patient’s understanding of the surgical intervention, and aids in teaching and team communication. These factors can significantly reduce the risks and complexity of the resection [56].

Conclusions

We report an interesting case of concomitant FD and ABC affecting the zygomatic bone. The occurrence of these concomitant lesions produces a clinical, radiographical, and histopathological picture that may be indistinguishable from several other similar conditions; therefore, accurate diagnosis warrants careful investigation and examination. Optimal management involves the consideration of multiple factors in the differential diagnosis to select the appropriate treatment. The most modern techniques should be used to reduce the surgical risk and ensure functional and esthetic improvement to provide a better quality of life to the patient.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest of any nature (financial or non-financial).

Ethical approval All procedures performed in this study involving human participants were in accordance with the ethical standards of the Institutional and/or National Research Committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from the patient included in the study.

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