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Case Report

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Fibrous Dysplasia of the Frontal Sinus: A Case Report

Wadii Thabet MD[1,2*], Mehdi Hasnaoui MD[1,2], Nouha Ben Taher MD[1,2], Hela Abdallah MD[1,2], Marwane Mfathia MD[1,2], Khelifa Mighri MD[1,2]

[1]Department of Otorhinolaryngology, Tahar Sfar hospital, Mahdia, Tunisia
[2]Faculty of Medicine of Monastir, University of Monastir, Monastir, Tunisia

[*]**Corresponding author:** Wadii Thabet, Address: ENT department, Tahar Sfar Hospital, Jbal dar weja, 5111 Mahdia, Tunisia. Tel: +216 21 612 012, E-mail: thabetwadii@gmail.com; ORCID: 0000-0002-1686-989X

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Abstract

Fibrous dysplasia is a rare, non-hereditary skeletal disorder. The frontal sinus location is uncommon. The management of fibrous dysplasia of the frontal sinus is clinically challenging due to the complex anatomy of this area and the proximity of vital neurovascular and ocular structures. Here, we report a case of fibrous dysplasia of the frontal sinus in a 32-year-old man who presented with a left periorbital swelling lasting for 3 days. The computed tomography scan showed fibrous dysplasia of the left frontal sinus; with the characteristic 'ground-glass appearance' and erosion of the superior orbital wall; complicated with preseptal and orbital cellulitises. The patient was treated with intravenous antibiotics. Given the absence of cranial nerve dysfunction or facial deformity, surgical treatment was not required. The purpose of this article was to report our case and discuss its clinical and radiological features, as well as the management of this rare disease.

Keywords: *Fibrous dysplasia; paranasal sinus; frontal sinus; management.*

Introduction

Fibrous dysplasia (FD) is a rare, non-hereditary skeletal developmental disorder characterized by abnormal replacement of normal bone with excessive proliferation of cellular fibrous connective tissue mixed with irregular bony trabeculae [(1)]. FD can present as a single lesion, known as monostotic FD, or as multiple lesions involving several bones, referred to as polyostotic FD [(2)]. It mostly affects the maxilla and the mandible. The fontal sinus location is uncommon [(2)].

The purpose of this article was to report a case of fibrous dysplasia of the frontal sinus and to discuss its clinical and radiological features as well as the management of this rare disease.

Case Report

A 32-year-old male, without medical history, presented with a 3-day history of left periorbital swelling. The patient did not report any history of headaches, childhood fractures, recurrent rhinosinusitis, or trauma. Physical examination revealed an inflammatory edema of the left eyelids, conjunctival hyperemia, and frontal sinus tenderness on the left side **(Fig. 1)**. Visual acuity was normal and there were no signs of vision restriction, ophthalmoplegia, proptosis, bony prominence, or nasal obstruction/discharge. In addition, there were no café-au-lait spots.

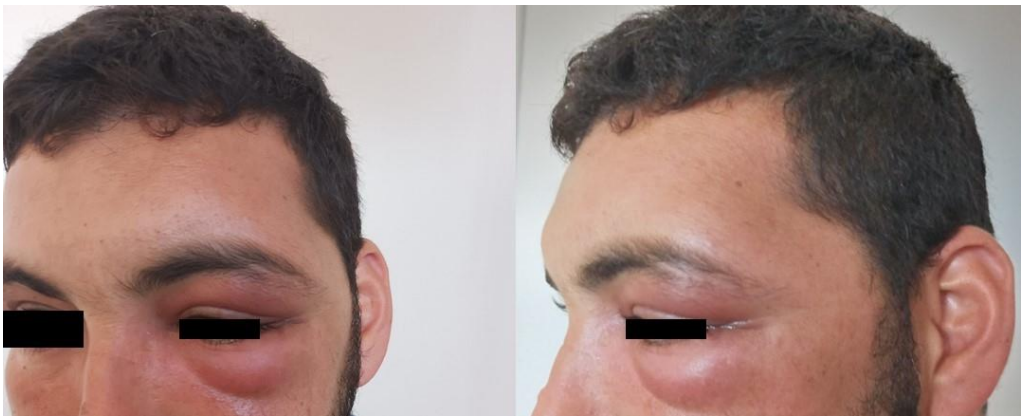

Figure 1: Inflammatory edema of the left eyelids.

The computed tomography (CT) scan showed fibrous dysplasia of the left frontal sinus: dense and sclerotic lesions with the characteristic "ground glass" appearance. Erosion of the superior orbital wall was observed **(Fig. 2)**. These findings were associated with preseptal and orbital cellulitises. The diagnosis of fibrous dysplasia of the left frontal sinus complicated with acute rhinosinusitis was retained.

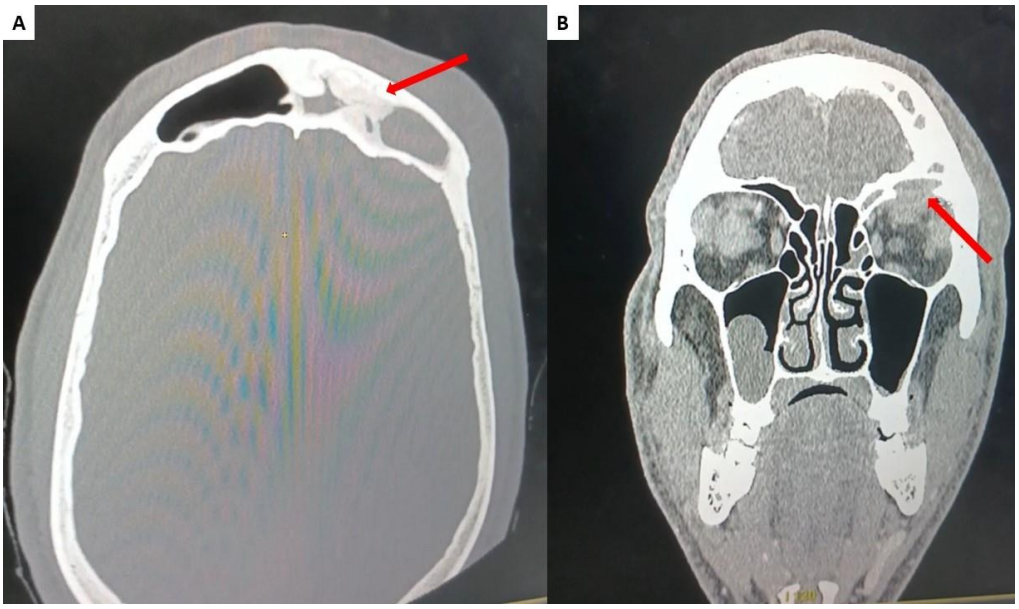

Figure 2: (A) Axial CT scan showing a ground-glass bony lesion of the left frontal sinus. (B) Coronal CT scan showing an erosion of the superior orbital wall associated with orbital cellulitis.

The patient was treated with intravenous antibiotics (Cefotaxime, Vancomycin, and Metronidazole) for 10 days, resulting in significant improvement. Given the absence of cranial nerve dysfunction or facial deformity, no surgical intervention was required.
After 6 months of follow-up, the patient remained asymptomatic.

Discussion

FD is a rare, non-inherited genetic disorder, often underdiagnosed due to its asymptomatic forms. It accounts for approximately 2.5% of all bone lesions and about 7% of benign bone lesions [(1)]. This condition occurs when normal bone connective tissue is replaced by immature, non-lamellar fibrous tissue [(2)]. The pathogenesis is primarily attributed to post-zygotic activating mutations in the GNAS gene, located on chromosome 20q13, which encodes the alpha subunit of the stimulatory G protein [(2)]. These mutations inhibit the differentiation and proliferation of bone-forming stromal cells during the early stages of embryonic development [(2)]. FD affects both sexes, with a slight predominance in females [(2)]. Males are more commonly affected during the second decade of life, when the highest prevalence is observed, accounting for approximately 63% of cases [(3)]. The enlargement of FD lesions typically begins in early childhood and tends to stabilize by late adolescence. However, most diagnoses occur during the second and third decades of life, particularly in monostotic fibrous dysplasia.

The phenotype of FD is highly variable and can be classified as monostotic (involving a single skeletal site), polyostotic (involving multiple sites), or associated with other systemic disturbances. The craniofacial bones are involved in 10%–25% of cases in monostotic forms and 50% in polyostotic forms [(2)]. Monostotic FD represents the most common form, accounting for approximately 80% of cases. The maxilla and the mandible are the most commonly affected, each involving approximately 12% [(2)]. Involvement of paranasal sinuses and temporal bones is less frequent. The ethmoid sinus is the most affected one [(3)]. Monostotic FD is often asymptomatic, and the diagnosis is typically incidental. On the contrary, polyostotic FD usually presents with earlier manifestations and may be associated with extraskeletal features, such as café-au-lait spots and hyperfunctioning endocrinopathies, as seen in McCune–Albright syndrome. Polyostotic FD can also be associated with conditions such as hyperparathyroidism, tuberous sclerosis, and soft-tissue myxomas, particularly in Mazabraud's syndrome [(3)].

The most common manifestations of fibrous dysplasia of the frontal sinus include bone deformities and swelling. In some cases, patients report nasal obstruction or congestion, recurrent rhinosinusitis, headaches, and visual disturbances such as proptosis, epiphora, diplopia, and even vision loss due to optic nerve compression [(3)]. Although malignant transformation is rare (less than 1%), FD can exhibit locally aggressive behavior, potentially extending intracranially and leading to neurological deficits and encephalitis [(3)].

Computed tomography (CT) is the gold standard for diagnosing FD [(4)]. The most common radiographic feature of FD is the 'ground-glass' appearance, characterized by a mixture of radiolucent and radiopaque fibrous tissue, observed in 56% of adult cases [(4)]. The second most common pattern is the sclerotic variant, which has a homogeneous appearance, and the least common form is the cystic variant, accounting for 21% of reported cases, where the lesion contains fibrous tissue [(4)]. On MRI, FD typically exhibits a low to intermediate signal intensity on T1-weighted images. On T2-weighted sequences, the signal intensity can be variable, with areas of high intensity reflecting the heterogeneous nature of FD lesions [(4)]. MRI is a valuable imaging modality, especially for assessing the compression of neurological structures and detecting other soft tissue lesions [(4)].

The goal of FD management is to achieve both functional and aesthetic outcomes. Treatment options include clinical observation, medical therapy, and surgical intervention.

Clinical observation is typically recommended for asymptomatic patients, with annual facial CT scans performed for the first two years to monitor any enlargement of lesions [(3)]. After this period, the imaging frequency is adjusted according to clinical findings. In our case, given the absence of cranial nerve dysfunction or facial deformity, no surgical intervention was required and we opted for clinical observation.

Medical treatment is based on bisphosphonates. Their effect in suppressing the progression and activity of FD lesions, as well as in reducing pain remains controversial [3,5,6].

Surgical therapy is considered the only curative treatment for FD, with its main goal being restoration / preservation of function and avoidance of cosmetic deformity [3,5]. Surgical approaches can be either radical (followed by reconstruction) or conservative. However, there is no consensus on the most effective method. The choice of approach should be tailored to the individual patient, taking into account factors such as age, the presence of functional or aesthetic concerns, the potential benefits and risks and proximity to critical structures [7]. The endoscopic endonasal approach should usually be preferred to external approach [7]. Surgery is indicated in symptomatic patients (especially pain), cosmetic deformity, and compression of vital organs [3,8]. Surgery is considered, before adulthood, in cases of extensive bone involvement or functional impairment [8].

Long-term follow-up is essential to monitor for potential complications, malignant transformation or recurrence (25%) [3].

Conclusion

Although FD is a benign condition, it is crucial to promptly recognize the disease and ensure appropriate follow-up and management. Due to the locally expansive nature of FD, complications can arise that often require surgical intervention. The disease can present with various radiological appearances. Early detection and careful follow-up of patients are essential in preventing potential complications associated with this significant condition.

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None.

Author contributions

Conceptualization: Wadii Thabet. Data collection: Nouha Ben Taher. Writing: Wadii Thabet and Nouha Ben Taher. All authors reviewed the manuscript.

Data Availability

All data generated or analyzed during this research are included in this published article. Data supporting the findings of this study are available from the corresponding author on reasonable request.

Declaration of Conflicting Interests

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Ethical considerations

Not applicable

Consent to participate

Not applicable

Consent for publication

Written informed consent was obtained from the patient to publish the case report.

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