

Decade-Long Stable Monostotic Fibrous Dysplasia of The Midface: A Case Report on Conservative Surgical Recontouring

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ABSTRACT

Introduction: Fibrous dysplasia is a benign fibro-osseous disorder that typically presents in childhood or adolescence with gradual, painless bony expansion. While craniofacial involvement is well documented, cases presenting with isolated, long-standing unilateral midface enlargement without progressive growth or systemic association are less commonly reported. This case highlights the diagnostic and surgical considerations in managing a decade-long stable monostotic lesion confined to the midface, and adds to the limited literature on conservative recontouring as a definitive esthetic solution in such stable presentations, with particular emphasis on the role of piezoelectric osteotomy in reducing postoperative morbidity relative to conventional rotary techniques.

Case Presentation: An 18-year-old female presented with left-sided facial asymmetry of 10 years' duration, involving the midface region extending anteroposteriorly from the left alar base to the left lateral canthus, and superoinferiorly from the infraorbital rim to the left oral commissure. The swelling was asymptomatic and had shown no notable increase in size. A prior biopsy was reported as consistent with fibrous dysplasia.

Intervention, and Outcome: Comprehensive clinical, radiographic, and hematological evaluation confirmed monostotic fibrous dysplasia limited to the left midface, with no evidence of systemic or polyostotic involvement. The patient underwent surgical recontouring of the affected region under general anesthesia, performed primarily with a piezoelectric device rather than conventional rotary instrumentation, to achieve precise, selective bone reduction while preserving adjacent soft tissue and neurovascular structures. Postoperative recovery was uneventful, with satisfactory esthetic correction of facial symmetry, minimal intraoperative bleeding, and notably reduced postoperative morbidity compared with outcomes typically reported for conventional burr-based recontouring.

Conclusion: Stable, long-standing monostotic fibrous dysplasia of the midface can be effectively managed with conservative surgical recontouring alone, achieving favourable esthetic outcomes without need for more extensive resection, provided systemic involvement is thoroughly ruled out. The use of piezoelectric instrumentation for this recontouring offered a precision-driven alternative to conventional rotary osteotomy, associated with lower postoperative morbidity, supporting its preferential use in similar conservative craniofacial recontouring procedures.

KEYWORDS: Fibrous Dysplasia · Facial Asymmetry · Maxilla · Reconstructive Surgical Procedures · Esthetics.

Introduction

Fibrous dysplasia is a rare, non-inherited skeletal disorder arising from a post-zygotic activating mutation in the GNAS gene, resulting in replacement of normal bone by immature, poorly mineralized fibro-osseous tissue [1,2]. It may present as monostotic, polyostotic, or craniofacial disease, with the latter defined by involvement confined to contiguous bones of the craniofacial skeleton [1]. Craniofacial involvement is reported in up to 90% of polyostotic cases and 10–27% of monostotic cases, most commonly affecting the maxilla, and typically manifests as painless, slowly progressive facial asymmetry that stabilizes after skeletal maturity [1,3].

It is the most common fibro-osseous lesion but no universally accepted management protocol has been established for craniofacial fibrous dysplasia. Treatment decisions are individualized, with observation to radical resection or conservative recontouring depending on the extent of the lesion, symptomatology and activity of the disease [1,5]. Surgical recontouring is particularly favored in stable, non-progressive lesions causing predominantly cosmetic deformity, as it avoids the morbidity associated with radical excision and reconstruction while achieving satisfactory aesthetic correction [1,5]. However, the literature on isolated midfacial monostotic-pattern involvement with long-standing stability and successful single-stage recontouring remains limited, with most published series emphasizing polyostotic, syndromic, or orbit-involving disease requiring multimodal or staged intervention [3,5].

This case is notable for the decade-long stability of a well-demarcated left midfacial deformity in a young adult without systemic, ocular, or functional compromise, and for the favorable aesthetic outcome achieved through

a single conservative recontouring procedure using piezoelectric osteotomy. Reporting such cases contributes to the limited evidence base guiding conservative surgical management of stable craniofacial fibrous dysplasia.

CASE PRESENTATION

Patient Information

An 18-year-old female patient presented with a chief complaint of gradually noticed swelling and facial asymmetry involving the left mid-face, first observed by the family approximately 10 years earlier (at approximately 8 years of age). The deformity had remained clinically stable in size and contour over this period, according to patient and parental report, with no recent acceleration in growth.

Primary concerns and symptoms: The patient's sole concern was cosmetic disfigurement of the left mid-face. There was no associated pain, paresthesia, diplopia, visual disturbance, nasal obstruction, hearing impairment, headache, or malocclusion. No history of trauma preceded the onset of swelling.

Medical, family, and psychosocial history: The patient had no relevant contributory medical history, no history of fractures, precocious puberty, endocrinopathy, or café-au-lait skin pigmentation, making an association with McCune-Albright syndrome or Jaffe-Lichtenstein syndrome clinically unlikely [2,3]. Family history was non-contributory, and no similar swellings or skeletal abnormalities were reported in first-degree relatives, consistent with the sporadic, non-inherited, post-zygotic mutational origin of fibrous dysplasia [2]. Psychosocially, the long-standing facial asymmetry had contributed to the patient's decision to seek definitive aesthetic correction at this stage of early adulthood, a period during which fibrous dysplasia lesions are known to become quiescent following skeletal maturity [1,3].

Past interventions and outcomes: Prior to presentation at our institution, the patient had undergone an incisional biopsy of the left mid-facial lesion elsewhere, histopathologically reported as fibrous dysplasia. No previous surgical, medical or orthodontic intervention had been undertaken for this condition.

Clinical Findings

Extraoral examination revealed a firm, non-tender, bony-hard swelling causing facial asymmetry of the left mid-face. The lesion extended anteroposteriorly from the left ala of the nose to the left lateral canthus of the eye, and superoinferiorly from the left infraorbital rim to the left commissure of the mouth.[Figure 1 and 2] The overlying skin was normal in color and texture, without sinus tracts, ulceration, or local rise in temperature. There was no proptosis, globe displacement, dystopia, or restriction of extraocular movements, and pupillary and visual function were clinically unremarkable. No cranial nerve deficits were elicited.



Figure 1: Frontal view of the patient (A) preoperatively and (B) postoperatively, showing improvement in facial symmetry.

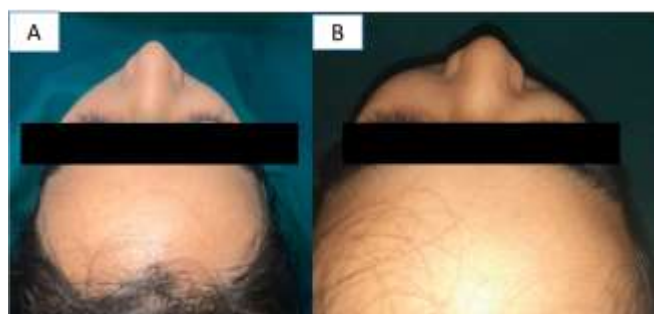


Figure 2: Bird's-eye view of the patient (A) preoperatively and (B) postoperatively

Intraoral examination revealed buccal cortical expansion, with no evidence of tooth displacement, malocclusion, or mucosal ulceration. No cervical lymphadenopathy was noted. No café-au-lait macules or cutaneous pigmentation were observed on general examination, and no other skeletal swellings were palpable, supporting a monostotic/craniofacial pattern of involvement rather than polyostotic disease [1,3].

Timeline

The patient first noticed painless swelling of the left mid-face at approximately 8 years of age, which remained stable in size over the following decade without associated functional symptoms. Prior to presentation, an incisional biopsy performed elsewhere confirmed fibrous dysplasia. At 18 years of age, she presented to our department, where clinical examination and photographic documentation were carried out, followed by radiographic imaging and baseline blood investigations. After multidisciplinary review, surgical recontouring of the left mid-face was performed under general anesthesia using a piezoelectric device. The postoperative course was uneventful, and follow-up visits showed satisfactory wound healing with a stable, symmetric aesthetic outcome.

Diagnostic Assessment

Diagnostic methods:

- Clinical examination: Extraoral and intraoral assessment of swelling extent, consistency, and functional status as above.
- Laboratory testing: Serum calcium, phosphate, and alkaline phosphatase (ALP) were assessed, as ALP is reported to be elevated in a substantial proportion of fibrous dysplasia patients and correlates with disease activity, particularly in craniofacial involvement [3]. ALP as per blood investigation was 108 U/L [normal value: 45 to 87 U/L].
- Imaging: Computed tomography (CT) of the face [Figure 3] was performed to delineate lesion extent, characterize the classic “ground-glass” radiopacity, and assess involvement of adjacent structures including the orbit, infraorbital canal, and maxillary sinus [1,3]. Bony expansion with ground-glass matrix involving the anterolateral and posterolateral walls of left maxillary sinus, left superior alveolar arch. There is reduced maxillary sinus pneumatization on the left side. There is nerve canal traversing in between the expanded medullary cavity- likely intraorbital nerve canal. Right maxillary sinus is normal in outline and attenuation.

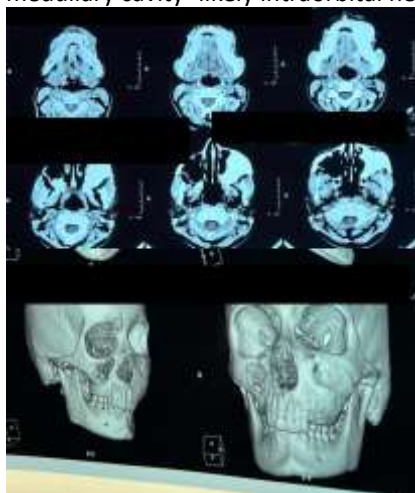


Figure 3: CT of the facial bones showing bony expansion with a ground-glass matrix involving the anterolateral and posterolateral walls of the left maxillary sinus and the left superior alveolar arch

- Bone scintigraphy was not performed to exclude additional occult skeletal lesions and confirm a monostotic/craniofacial distribution [1].

Diagnostic challenges: As fibrous dysplasia shares overlapping radiographic and histopathological features with other fibro-osseous lesions (ossifying fibroma, osseous dysplasia), the correlation of clinical stability, characteristic imaging, and prior histopathology was essential to confirm the diagnosis and exclude alternative or malignant fibro-osseous pathology [3]. The absence of GNAS mutation testing in this case relied on the classical clinoradiological–histopathological triad for diagnostic confirmation, as recommended when genetic testing is not readily accessible [3].

Diagnosis: Craniofacial fibrous dysplasia (monostotic pattern), isolated to the left mid-face, non-syndromic (no evidence of McCune-Albright or Jaffe-Lichtenstein syndrome).

Differential diagnoses considered: Ossifying fibroma, osseous dysplasia, Paget's disease of bone, and low-grade osteosarcoma were considered and excluded based on the stable decade-long clinical course, characteristic imaging, and confirmatory histopathology [3].

Prognostic characteristics: The absence of disease progression over a documented 10-year period, patient age at skeletal maturity, and lack of syndromic association were favorable prognostic indicators supporting a conservative recontouring approach over radical resection, consistent with reported outcomes in similarly stable, non-progressive lesions [1,5].

Therapeutic Intervention

Given the long-standing stability of the lesion, absence of functional compromise, and the patient's primary concern being aesthetic, conservative surgical recontouring of the left mid-face was elected over radical resection and reconstruction, in line with literature supporting debulking as a viable, lower-morbidity option for stable, non-aggressive craniofacial fibrous dysplasia lesions in patients presenting after the period of active skeletal growth [1,5]. The procedure was performed under general anesthesia via an intraoral maxillary vestibular incision. Recontouring was achieved primarily using a piezoelectric device, supplemented selectively by a high-speed burr, to reduce the expanded bone to match the contour of the contralateral unaffected side, with intraoperative assessment of symmetry [Figure 4].



Figure 4:(A) Preoperative intraoral view. Intraoperative views showing (B) exposure of the pathologic site, (C) marking of the osteotomy with a piezoelectric device, (D) the surgical site after recontouring, and (E) primary wound closure.

The piezoelectric system was favored as the primary cutting modality because of its recognized advantages over conventional rotary instrumentation in this anatomically delicate region: selective, microvibration-based cutting of mineralized bone with relative sparing of adjacent soft tissue, lower intraosseous temperature generation, and improved intraoperative visibility of the operative field through the cavitation effect of its irrigation system [10]. These properties allowed controlled, incremental bone reduction close to the infraorbital rim and orbital floor with a reduced risk of thermal bone necrosis, inadvertent soft-tissue injury, and excessive intraoperative bleeding compared with conventional burr-based osteotomy [10]. No intraoperative or postoperative modification to the planned surgical approach was required and no adjunctive pharmacologic therapy was administered, as the patient was asymptomatic for pain and had stable, non-progressive disease.

Follow-up and Outcomes

Postoperatively, the patient achieved satisfactory facial symmetry and aesthetic outcome, as assessed both clinically by the surgical team and subjectively by the patient [5]. The patient tolerated the surgical procedure well, with an uneventful postoperative recovery notable for minimal swelling and a comfortable early postoperative course, consistent with the blander postoperative period reported for piezoelectric osteotomy relative to conventional rotary techniques [10]. Wound healing was assessed clinically at routine follow-up visits; the patient reported no functional deficits (e.g., paresthesia, malocclusion, ocular symptoms) postoperatively. No intraoperative or postoperative complications (infection, hematoma, nerve injury, or asymmetry recurrence) were reported during the follow-up period. Long-term follow-up was recommended given the reported

potential, albeit low, for recurrence or continued growth, particularly given that some regrowth after debulking procedures has been documented in the literature even in adult patients [1,5].

Discussion

This case demonstrates a decade-long, stable, monostotic fibrous dysplasia confined to the left midface, successfully managed with conservative surgical recontouring alone, using a piezoelectric device as the primary bone-cutting instrument. The principal strength of this case lies in the long duration of follow-up documenting a genuinely quiescent lesion, allowing a clear distinction from the more aggressive fibro-osseous entities that are frequently confused with fibrous dysplasia, such as juvenile trabecular ossifying fibroma, which characteristically shows rapid growth and a high recurrence potential [6]. Thorough hematological and radiographic screening excluding polyostotic disease and McCune–Albright syndrome strengthens the diagnostic confidence. A limitation is that molecular confirmation of a GNAS mutation was not performed; although histopathology and imaging were considered sufficient for diagnosis, genetic analysis can add specificity where clinical and radiological findings are equivocal [9]. Notably, the postzygotic GNAS mutation underlying fibrous dysplasia produces a mosaic population of abnormal osteoblasts, and disease severity reflects the ratio of mutated to normal cells at the affected site rather than the embryological timing of mutation, which may partly explain why some monostotic lesions remain confined and stable [2]. Additionally, as a single case, the long-term recurrence risk after recontouring — a recognized concern in fibrous dysplasia surgery — cannot be generalized from this outcome alone; larger case series similarly report variable recurrence and emphasize long-term follow-up even after seemingly definitive surgery [3].

Craniofacial fibrous dysplasia typically manifests as painless, slowly progressive bony expansion during childhood or adolescence, with a tendency to become quiescent after skeletal maturity, unlike gnathic fibrous dysplasia, which frequently remains active into adulthood [8,9]. Computed tomography remains the imaging modality of choice, characteristically demonstrating a “ground-glass” pattern with poorly defined, blending borders and an intact, though thinned, cortex — findings that were consistent with our patient’s presentation [7,9]. The maxilla is the most frequently affected craniofacial bone, and lesions may cross sutures into contiguous bones without technically qualifying as polyostotic disease [7,8]. A large systematic review of craniofacial fibrous dysplasia management similarly concluded that surveillance is a viable option for asymptomatic, non-progressive lesions, and proposed a management algorithm reserving radical or debulking surgery for symptomatic or progressive disease [1]. Surgical literature increasingly favors conservative contouring procedures over radical resection once the lesion has stabilized post-puberty, as this reduces operative morbidity while achieving acceptable aesthetic outcomes, albeit with a somewhat higher reported recurrence rate compared with more extensive resection and reconstruction [9]. Where recontouring alone proves insufficient, three-dimensional modeling techniques using mirror-image biomodels of the unaffected side have been described to improve precision in facial recontouring for stable fibrous dysplasia [5]. This is distinct from more aggressive fibro-osseous lesions such as JTOF, which typically necessitate wider excision given the disease’s propensity for rapid growth and recurrence [6].

The choice of a piezoelectric device over conventional rotary burs for the recontouring in this case merits emphasis, as it reflects a broader shift toward minimally invasive, tissue-preserving instrumentation in maxillofacial bone surgery. Compared with conventional drills and saws, piezosurgery performs a selective, ultrasound-based cut that is limited to mineralized tissue, sparing adjacent soft tissue, vessels, and nerves, and generates substantially less intraosseous heat, thereby reducing the risk of thermal bone necrosis and border injury during osteotomy. The cavitation effect produced by the device’s irrigation system also improves intraoperative visibility by limiting bleeding into the operative field, which is particularly advantageous when recontouring bone close to the infraorbital rim and orbital floor, as in this case [10]. These characteristics have been associated in the broader literature with reduced intraoperative bleeding, greater precision and predictability of the osteotomy, and a comparatively bland postoperative course with less edema relative to conventional rotary instrumentation, without increasing overall operative time [10]. Taken together with reports of increased bone morphogenetic protein expression and reduced inflammatory cytokine activity at piezoelectrically cut bone margins, these properties provide a plausible mechanistic basis for the uncomplicated, low-morbidity recovery observed in the present case, and support piezosurgery as a preferable alternative to conventional burr- or saw-based recontouring in similar stable craniofacial fibrous dysplasia presentations, particularly where the lesion approximates critical neurovascular or orbital structures.

Given the decade-long clinical stability, absence of systemic features, absence of pain — notable since nociceptive mechanisms in fibrous dysplasia are increasingly linked to active lesion remodeling and inflammation rather than quiescent disease [4] — and classic radiographic ground-glass appearance confirmed

histologically, a diagnosis of quiescent monostotic fibrous dysplasia was established with reasonable certainty. This clinical stability justified a conservative surgical approach targeting aesthetic correction rather than radical excision, and the use of piezoelectric osteotomy for this correction is consistent with current best-practice recommendations for stabilized, non-aggressive craniofacial fibrous dysplasia [8,9,10].

Conclusion

This case underscores that not all craniofacial fibro-osseous swellings require aggressive resection; when fibrous dysplasia has clearly reached a stable, non-progressive phase as evidenced by a prolonged asymptomatic clinical course, absence of systemic or polyostotic features, and characteristic radiographic findings — conservative surgical recontouring can achieve excellent aesthetic and functional outcomes with lower morbidity than radical surgery. The findings of this case further suggest that piezoelectric instrumentation, by virtue of its selective bone-cutting profile, soft-tissue and nerve preservation, and favorable postoperative recovery relative to conventional rotary methods, represents the preferred tool for such conservative recontouring, especially in regions bordering critical anatomical structures [10]. Careful longitudinal clinical assessment, combined with appropriate imaging and biochemical screening to exclude McCune–Albright syndrome, is essential before selecting a minimally invasive surgical strategy, and clinicians should maintain a high index of suspicion for more aggressive fibro-osseous mimics when growth patterns deviate from this quiescent course.

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