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Fibrous Dysplasia of the Mandible Presenting with Facial Asymmetry: A Case Report

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Abstract

Fibrous dysplasia is a benign fibro-osseous disorder caused by a genetic mutation, replacing normal bone with fibrous tissue. A 24-year-old female presented with a 10-year history of progressive right mandibular swelling post-trauma, causing facial asymmetry. Examination revealed a hard, non-tender swelling. Radiographs (OPG, CBCT) showed a radiopaque lesion with cortical expansion and mandibular canal obliteration. Histopathology confirmed Fibrous dysplasia, displaying irregular trabeculae with fibrocellular stroma. Surgical recontouring was performed and asymmetry of the jaw was corrected. This case provides a clinically-focused comprehensive description of Fibrous dysplasia, its natural progression, the components of the diagnostic evaluation and its surgical management.

Keywords: Cone beam computed tomography, Fibrous Dysplasia, Mandible, Cortisectomy

Introduction

Fibrous dysplasia is a benign intramedullary fibro-osseous disorder caused by a genetic mutation that hinders normal bone maturation, replacing it with fibrous tissue.[1] It comprises 2–5% of all bone tumors and about 7% of benign bone tumors.[2] The maxilla is more frequently affected than the mandible, with lesions often unilateral in nature.[3] Fibrous dysplasia occurs in three forms: monostotic, polyostotic, and McCune-Albright syndrome. Monostotic is the most common and usually affects craniofacial bones, with a female predominance.[4] Lesions are slow-growing and self-limiting but may rapidly expand during puberty. Diagnosis is made through clinical, radiological, and histopathological examination. Treatment may be

conservative or surgical, depending on lesion size, location, patient age, and extent of cosmetic or functional issues.[1] Mandibular involvement may cause nerve compression, occlusal disturbances, and facial asymmetry.[5] This report presents a case of a 24-year-old female with a progressive swelling on the right side of mandible, supported by clinical, radiographic, and histopathological findings.

Case Report

A 24 year old female reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of facial asymmetry since 10 years. Patient gave a history of fall from tractor at the age of 14 years, 3 months after which she developed a swelling over right lower third of face. The swelling gradually increased in size over a period of 3 months with no further progression till the time of evaluation. On clinical examination, a distinct facial asymmetry was noted with a diffuse swelling on the right side of the mandible measuring 5x3 cm^2 spanning medially from midline to angle of mandible and superiorly from corner of the mouth – tragus till the inferior border of mandible with normal overlying skin. On palpation, the swelling was hard in consistency. (Figure 1) Intra oral examination revealed expansion of the buccal cortical plate extending from mandibular right canine to mandibular right third molar which was hard in consistency. An orthopantomogram was advised which showed loss of trabecular bone pattern with radiopaque lesion on the right side of the mandible extending from 43to 48 region accompanied by obliteration of the mandibular canal. (Figure 2A) The cone beam computed tomography images revealed a radiopaque lesion with expansion of both buccal and lingual cortical plates. (Figure 2B) Based on clinical and radiographic findings, a differential diagnosis of fibrous dysplasia, ossifying fibroma, osteoid osteoma was made. An incisional biopsy was performed, and it was histopathologically reported as fibrous dysplasia. Based on histopathology, radiographic and clinical investigations, surgical recontouring was planned under general anaesthesia.

Figure 1 : Pre- operative clinical picture of the patient

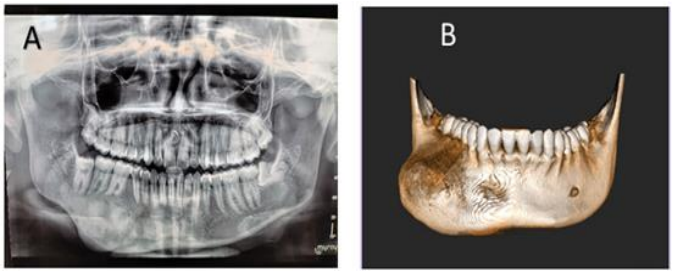

Figure 2: Pre- operative : Orthopantomogram (A) and Cone beam computed tomography (CBCT) (B)

Surgical Procedure

The procedure was performed under general anesthesia with nasal intubation. A vestibular incision was made from 35 to 47 teeth region, and mucoperiosteal reflection was done to expose the underlying bone. The inferior border of the mandible was marked and by using rotary instruments, chisel and mallet inferior and lateral cortisectomy was performed to reduce the prominence and the bone was contoured gradually, ensuring smooth and symmetrical mandibular outline. The excised sample was sent for histopathological evaluation. Closure was achieved through 3-0 vicryl suture. (Figure 3 A, B, C) Patient has been kept under periodic follow up. (Figure 4 A,B)

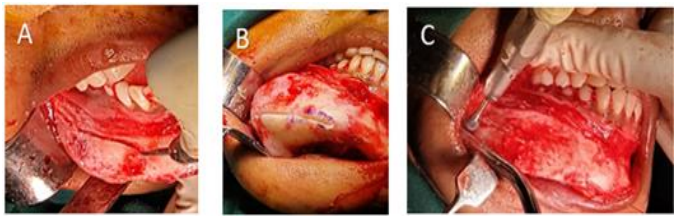

Figure 3: Intra- operative pictures (A) Lateral cortisectomy (hyperplastic bone from the lateral cortex was excised) (B) Inferior cortisectomy (hyperplastic bone from the inferior border was excised) (C) Recontouring

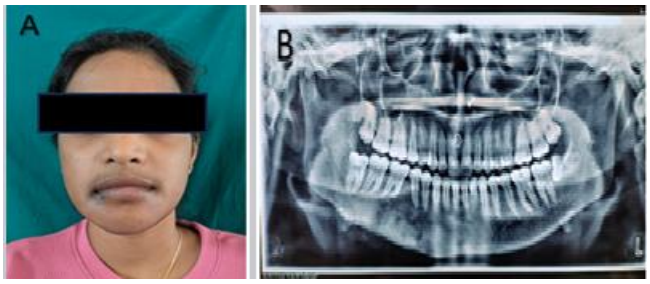

Figure 4: Post- operative (A) Clinical picture, (B) Orthopantomogram of the patient

Histopathology

Gross examination of the specimen included 4 bony tissues, largest measuring 3 x 1.5 x 1 cm and smallest measuring 2.5 x 1.5 x 0.5 cm. H&E-stained decalcified section showed numerous irregular bony trabeculae. Most of the trabeculae exhibits C shaped/Chinese letter pattern with fibro cellular connective tissue stroma. Osteoblastic rimming of bony trabeculae and resting lines were noted. Focal area with howship lacunae with osteoclast was evident, suggestive of Fibrous dysplasia. (Figure 5)

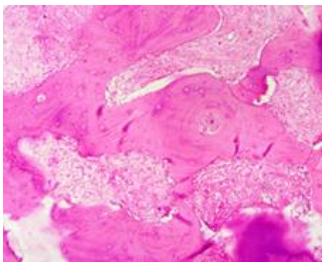

Figure 5: Photomicrograph showing Chinese letter bony trabeculae in fibro-cellular stroma (H&E 40x)

Discussion

Fibrous dysplasia was first reliably reported a century ago by von Recklinghausen. It is a relatively uncommon condition with an incidence rate of 1:4000-1:10,000. In the craniofacial region, it is more common in the maxilla compared to the mandible whereas in this case mandible was the affected site[6] Fibrous dysplasia can occur at any age however, it is usually observed in children and young adults with 75% of patients presenting before the age of 30.[2]

In this case, the patient reported with a chief complaint of facial asymmetry following trauma. However, the exact mechanism linking head trauma to the pathogenesis of fibrous dysplasia remains unclear, and the association between the two could simply be coincidental. Nevertheless, one study has suggested a connection between Fibrous Dysplasia development and elevated expression of the *c-fos* gene. Since *c-fos* upregulation has also been previously linked to trauma, it is possible that injury could trigger Fibrous Dysplasia through a *c-fos*-mediated pathway.[7]

These lesions are most commonly seen in the premolar to molar region with anterior segment being the least affected. Similar findings were observed in our patient. The most common presenting symptom in fibrous dysplasia is a gradual, painless enlargement of the involved bone, clinically seen as facial asymmetry. For such bony lesions, the preferred diagnostic approach is 3D imaging modality. The most common radiographic finding in Fibrous Dysplasia is the ground-glass appearance with an abnormal bone pattern.[4] Treatment modalities include conservative surgery, radical excision, and medical treatment with bisphosphonates.[8] The treatment approach is determined based on a thorough clinical and radiographic examination to achieve considerable functional and aesthetic outcomes.

Conservative surgery involves a remodelling procedure aimed at achieving reasonably acceptable aesthetics. The recurrence of fibrous dysplasia is reported in relation to puberty or skeletal maturation; however, its evolution is constant into adulthood .The surgical procedure is usually postponed till puberty as the disease will undergo remission.[9]

Various studies have shown that therapy with intravenous bisphosphonates (Zoledronic acid or Pamidronate) and Denosumab (RANKL monoclonal antibody) have been effective in relieving pain and normalizing bone turnover however, their long- term use should be limited as it can lead to necrosis of bone. For quiescent fibrous dysplasia without evident facial deformity or significant risk of pathological fracture, conservative management through regular follow ups represents an appropriate therapeutic approach.[5,10,11] In the present case, facial asymmetry was a concern for the patient hence, surgical management was planned in this case. Histopathological confirmation is important in establishing a definitive diagnosis but in some cases the histopathological examination has shown to be inconclusive due to similarity between histopathological characteristics with other fibro-osseous lesions such as ossifying fibroma. Hence, it is important to consider the stage of maturation of the lesion and the variations in its histological patterns for the establishment of a definitive diagnosis.[5] The reported prevalence of malignant transformation in fibrous dysplasia (FD) ranges from 0.4% to 4%, emphasizing the importance of periodic follow up to detect potential progression or malignant changes.[3]

Conclusion

Fibrous dysplasia affecting only the maxilla or mandible as a single, localized lesion is uncommon. Distinguishing fibrous dysplasia from malignant bone lesions can be challenging, requiring a comprehensive diagnostic approach that includes detailed patient history, clinical examination, and radiographic evaluation. Each case is different with peculiar symptoms and unique clinical findings. Therefore, the management should be tailored to the location, extent of involvement, and individual patient factors.

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