Case Report

Large odontogenic fibroma of the mandible mimicking malignancy in an adolescent male: Diagnosis unveiled on histopathology

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Abstract

Background: Odontogenic fibroma is a very rare benign tumour of the jaw with an overall incidence of less than 1% of all odontogenic tumours. It can either be unilocular or multilocular cystic tumour with a significant overlap between clinical and radiological findings amongst different odontogenic tumours of oral cavity.
Case report: We present a case of an odontogenic fibroma of mandible in a 14-year old boy, which was clinically suspected to be osteosarcoma with radiological differentials of odontogenic myxoma and ossifying fibroma. Histopathological examination showed a tumour composed of variably cellular fibroblastic stroma with bits of lamellar bone showing osteoblastic rimming. Interspersed strands of bland odontogenic epithelium and multinucleated giant cells were noted. There was no mitosis or atypia. Final diagnosis of odontogenic fibroma was rendered on histopathologic examination of the biopsy specimen.
Conclusion: This case report highlights the role of histopathological examination in diagnosing odontogenic fibroma, which can easily be missed due to clinical and radiological similarities with other benign and malignant entities.

Keywords: Odontogenic; fibroma; oral cavity; mandible; tumour

Introduction

Odontogenic fibroma is a rare, benign mesodermal tumour of odontogenic origin, first described by Schultz in 1957. [1] It accounts for less than 1% of all odontogenic neoplasms. [2] It usually presents as a mandibular swelling in young patients. The diagnosis is challenging on clinical and radiological examination. We report a case of a 14-year-old boy presenting with a large jaw swelling, clinically mimicking malignancy. Subsequent radiological examination revealed features of benign odontogenic neoplasm without a specific diagnosis. Histopathological examination of the biopsy specimen rendered the final diagnosis of odontogenic fibroma.

Case Report

A 14-year-old male presented to the Department of Dentistry, with the chief complaints of painless swelling over the right side of cheek from the past 6 months. The swelling was gradually increasing in size. There was no history of trauma, nasal complaints or visual disturbances. On external examination, facial asymmetry was present (Figure 1). The swelling was non

tender, non-fluctuant, hard in consistency without any evidence of pus discharge or bleeding. Mouth opening was reduced. An expansile lesion was noted in the right mandibular ramus on intraoral examination. In view of the large expansile mass with a short history, a clinical diagnosis of osteosarcoma was suspected. Radiological investigations and histopathologic examination were sought.

Radiograph (X-ray) of the skull showed a large well-defined radiolucent lesion surrounded by sclerotic border on right side of mandible (Figure 2). Contrast enhanced computed tomography (CECT) revealed a well-defined heterogeneously enhancing lesion measuring 53.1mm (CC) × 33.3mm (TR) × 54.1mm (AP) with soft tissue component involving the ramus, angle and adjacent posterior body of the right mandible. The lesion showed multilocular soap bubble appearance and thus caused marked thinning of the overlying cortex with focal areas of cortical disruption in the mandibular ramus (Figure 3). Based on these findings, the radiological differentials considered were odontogenic myxoma, ameloblastoma and ossifying fibroma.

Due to the misalliance of clinical and radiological diagnoses, incisional biopsy was taken and histopathological examination was sought. Microscopic examination showed a tumour composed of variably cellular fibroblastic stroma with bits of lamellar bone showing osteoblastic rimming. (Figure **??**) Interspersed within this stroma were seen strands of bland odontogenic epithelium and multinucleated giant cells. However, no mitosis or atypia was noted (Figure **??**). No myxoid stroma was noted. A final diagnosis of odontogenic fibroma was established. The tumour was removed by enucleation.

Figure 1: Large swelling in mandible causing facial asymmetry.

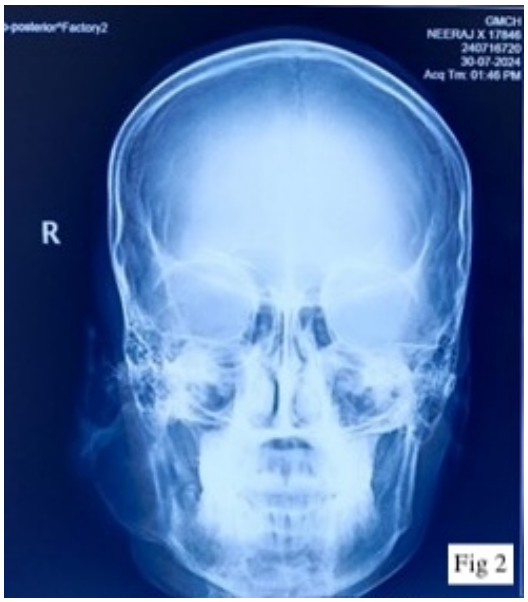

Figure 2: X-ray of the skull showing a large well-defined radiolucent lesion surrounded by sclerotic border on right side of mandible.

Discussion

Odontogenic fibroma is a relatively rare and benign entity. It is defined as "a benign odontogenic neoplasm having fibroblastic origin and is characterized by relatively mature collagenous fibrous tissue along with varying amounts of odontogenic

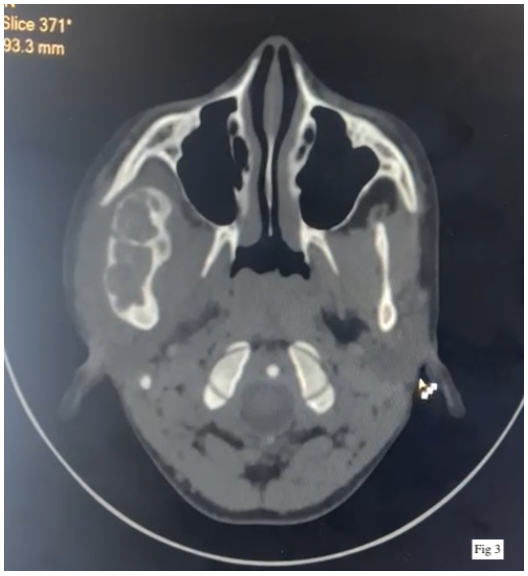

Figure 3: CECT showing a well-defined heterogeneously enhancing lesion measuring 53.1 × 33.3 × 54.1 mm involving the ramus, angle and adjacent posterior body of the right mandible. Multilocular soap bubble appearance with marked thinning of the overlying cortex noted.

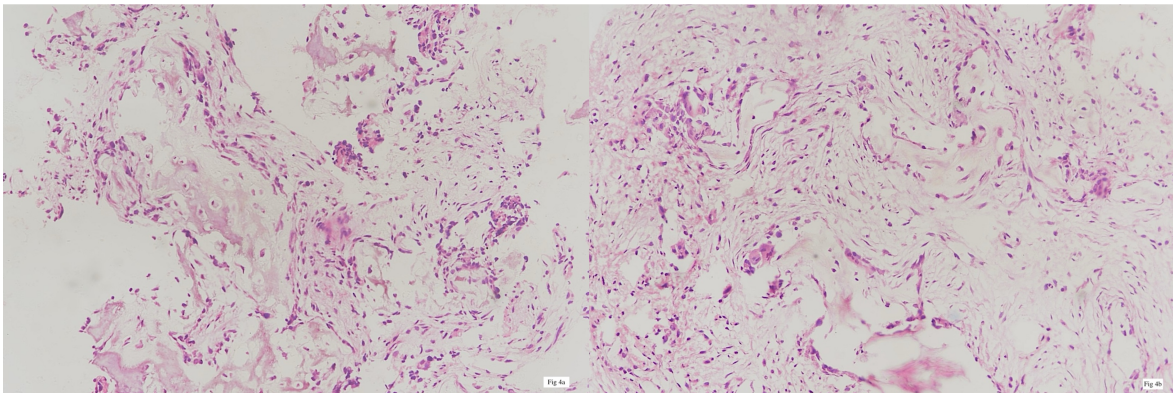

Figure 4: a. Microscopic examination showing a tumour composed of variably cellular fibroblastic stroma with bits of lamellar bone showing osteoblastic rimming. (H&E, ×100). b. Tumor showing interspersed strands of bland odontogenic epithelium and multinucleated giant cells. (H&E, ×100))

epithelium." [3] The incidence of odontogenic fibromas is 0.1% of the benign odontogenic tumours. [4] This entity was described in 1971 by the WHO for the first time and underwent many modifications over the period of time. [5] In the year 1975, Wesley et al suggested a set of diagnostic criteria for odontogenic fibroma based on clinical, radiological and histopathological parameters. [6] The terms central and peripheral odontogenic fibroma refer to intraosseous and extraosseous tumours respectively.

Most of the patients with odontogenic fibroma are between 2nd to 4th decade with a slight female preponderance. Clinically, this lesion presents as a painless and gradually progressive cystic enlargement of jaw causing displacement of the adjacent teeth in the affected area and leading to facial deformity. It may be associated with roots of unerupted teeth and may affect mandible or the posterior region of maxilla. [2, 3, 7] A very large mass may raise the suspicion of malignancy, as in the present case who presented with a progressively increasing swelling, with significant facial deformity.

Radiologically, odontogenic fibroma has been described both as a well-defined radiolucent area, that either simulates a unilocular ameloblastoma or odontogenic cyst or a multilocular radiolucent lesion having well-defined borders. [7] The great variability in radiologic appearance of odontogenic fibroma emphasizes that despite its rarity, it should be considered in the differential diagnosis of all the radiolucent lesions of the jaws. Although the radiological examination in the present case could rule out the clinical likelihood of malignancy, it could not pin point to a specific diagnosis. Owing to the multilocular appearance and well- defined borders, radiological differential diagnoses also included other more common benign odontogenic neoplasms such as ameloblastoma and ossifying fibroma.

On histopathologic examination, Gardner classified central odontogenic fibroma (COF) into three different groups: 1) Hyperplastic dental follicle 2) Simple COF 3) The WHO-type COF with features of dysplastic dentin or cementum-like

Table 1: Differential diagnosis of odontogenic fibroma

Entity	Odontogenic fibroma	Odontogenic myxoma	Ossifying fibroma	Ameloblastoma	Desmoplastic fibroma
Presentation	Maxilla and mandible	Mandible at posterior region and maxilla at premolar region	Mandible and maxilla with a predilection for mandibular premolar and molar	Often accompanied by unerupted teeth, frequently mandibular in the region of third molar	Locally aggressive benign lytic lesion in mandible
Radiology	Well-defined radiolucent lesions. May be unilocular or multilocular (Multilocular appearance in the present case)	Multilocular as "honeycomb", "soap bubble" or "tennis racket" aspect with well-defined borders	Mixed radiolucent opaque imaging	Unilocular, well-demarcated radiolucencies	Well-defined, almost multilocular radiolucent lesions
Histopathology	Collagenous connective tissue with variable amounts of odontogenic epithelium	Stellate and spindle shaped cells in a rich myxoid or mucoid stroma with few collagen fibrils	Cellular collagenized stroma with calcifications, psammoma bodies, or cementum	Single cyst lined by epithelium with palisaded columnar basal layer with reverse polarity and stellate reticulum-like cells.	Whorled aggregates of densely collagenous tissue containing uniform spindled cells.

tissue and varying amounts of odontogenic epithelium. [8] With the advent of the latest edition of WHO of head and neck tumours in the year 2022, the classification of odontogenic tumours was revisited and the use of terminology odontogenic fibroma was suggested. Newer subtypes were added which included amyloid subtype, granular subtype, ossifying subtype, hybrid odontogenic fibroma with central giant cell granuloma type. [2]

Odontogenic fibromas show moderately cellular bland fibrous tissue with moderate to dense collagen content, accompanied by varying amounts of dispersed inactive-appearing odontogenic epithelial nests and cords. There may be minor hard tissue formation in the form of mineralised dentinoid or round dense basophilic cementum-like mineralisation, both associated with the odontogenic epithelium. It can have varied histological appearance, and may either be epithelium poor type or epithelium rich type. [2, 3] Odontogenic fibroma with deposition of amyloid like protein is associated with deposition of acellular amyloid material around the epithelial strands of the tumour. Granular cell variant shows granular appearance of the epithelial cells and ossifying variant is characterised by presence of bony trabeculae admixed with epithelial strands.

Furthermore, due to its non-exclusive histological findings, the differential diagnosis of odontogenic fibroma may include a wide spectrum of tumours including odontogenic myxomas, ossifying fibroma and ameloblastomas, and fibrous tumours like fibromyxoma and desmoplastic fibroma. [3] Table 1 shows the differential diagnosis of odontogenic fibroma.

Histopathological examination of the present case showed a predominantly fibrous tumor with lamellar bone and very few strands of epithelial fragments. There was no atypia or mitosis. Thus, malignancy was definitely ruled out. Typical palisaded appearance of cells with reverse polarity, characteristic of ameloblastoma was not seen. Due to the prominent fibrous component of the tumor, ossifying fibroma and desmoplastic fibroma were the main histopathologic differentials. However, the presence of fragments of odontogenic epithelium helped in clinching the diagnosis of odontogenic fibroma. Since many bony trabeculae were found intermixed with fibrous tissue and epithelial strands, our case fits in ossifying type of odontogenic fibroma, according to the WHO classification.

Treatment of odontogenic fibroma consists of conservative surgery including curettage with complete removal of the lesion. The prognosis of central odontogenic fibroma is good with low recurrence rate if the lesion has been completely enucleated. [7]

Conclusion

Jaw lesions might often be misdiagnosed clinically and radiologically due to their rarity, and the very subtle differences between the various odontogenic neoplasms. In cases with discordance between clinical and radiological findings, as

encountered in the present case, prudent histopathological examination becomes absolutely essential in arriving at the confirmatory diagnosis.

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