CASE REPORT
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AN UNUSUAL HYBRID TUMOUR OF JAW; OSSIFYING FIBROMA WITH ODONTOME.

Oral & Maxillofacial Pathology

Dr. Chennuru Geetha
Dr. M. Rajini Kanth
Dr. Pavani Vidhyadhari
Dr. K. S. Vidya
Dr. K. Sowmya
Dr. T. Vaishnavi

ABSTRACT

Background: The coexistence of ossifying fibroma (OF) and odontome is an exceptionally rare clinicopathological entity, with very few cases reported in the literature. Accurate diagnosis requires correlation of clinical, radiographic, and histopathological findings.**Case Presentation:** A 40-year-old female presented for replacement of a missing tooth without any associated symptoms. Routine panoramic radiography incidentally revealed a mixed radiolucent–radiopaque lesion in the right retromolar region. Surgical excision was performed, and histopathological examination demonstrated a fibrocellular stroma containing irregular bony trabeculae along with dentinal tubules and dental papilla-like tissue, confirming the diagnosis of ossifying fibroma associated with odontome. **Conclusion:** This case highlights a rare hybrid lesion detected incidentally in an asymptomatic patient without cortical expansion or an impacted tooth. Careful clinicoradiographic correlation and histopathological evaluation are essential for accurate diagnosis and appropriate management. Reporting similar cases will enhance understanding of the biological behaviour and pathogenesis of these uncommon lesions.

KEYWORDS

Ossifying Fibroma; Odontome; Hybrid Odontogenic Tumour; Fibro-Osseous Lesion

INTRODUCTION

Fibro-osseous lesions represent a fascinating group of conditions where normal bone is gradually replaced by fibrous tissue and collagen, interspersed with varying amounts of mineralized material . Within this spectrum, cemento-ossifying fibroma (COF) stands out as a true osteogenic neoplasm, as described by Kramer et al. These benign tumors most often arise in the head and neck region particularly the jaws with a strong predilection for the mandibular premolar–molar area with female predominance. Clinically, COF tends to grow slowly but can cause noticeable swelling, tooth displacement, or facial asymmetry, while radiographically it evolves from radiolucent to mixed radiopaque with sharply defined borders[1].

In contrast, odontome is the most common odontogenic tumor, and it is better understood as a hamartomatous malformation rather than a true neoplasm. It is composed of enamel, dentin, cementum, and pulp arranged either in a tooth-like pattern (compound odontome) or as a disorganized calcified mass (complex odontome). Odontomes are often silent, discovered incidentally on radiographs, but they can interfere with normal tooth eruption. Surgical removal is straightforward and curative, with recurrence being exceedingly rare[2].

Here we present a rare case report of COF and Odontome.

Case Report

A 40-year-old female presented to the Department with the chief complaint of replacement of a missing tooth. Her medical and dental histories were non-contributory. Intraoral examination was unremarkable, with no evidence of cortical expansion, mucosal changes, tenderness, or discharge. As part of the pre-prosthetic assessment, a panoramic radiograph was obtained, which incidentally revealed a well-defined mixed radiolucent–radiopaque lesion with ill defined borders noticed in the right side retromolar region. An excisional biopsy was performed, and the specimen was submitted to the Department of Oral and Maxillofacial Pathology for histopathological examination. On gross examination multiple tissue bits were received in which few were soft – firm in consistency and one of the bit was hard which was kept for decalcification, Hematoxylin and eosin (H&E)-stained soft tissue sections revealed connective tissue composed of numerous irregular bony trabeculae having osteocytes within lacunae and without osteoblastic rimming. The intervening stroma was densely collagenized and highly cellular, with plump fibroblasts, mild inflammatory cell infiltrate, numerous endothelial-lined blood capillaries, areas of extravasated erythrocytes, and focal hemorrhage. The decalcified sections demonstrated irregular eosinophilic dentin with well-defined dentinal tubules associated with loosely arranged fibrovascular connective tissue resembling dental papilla. These histopathological features were consistent with a ossifying fibroma with odontome.

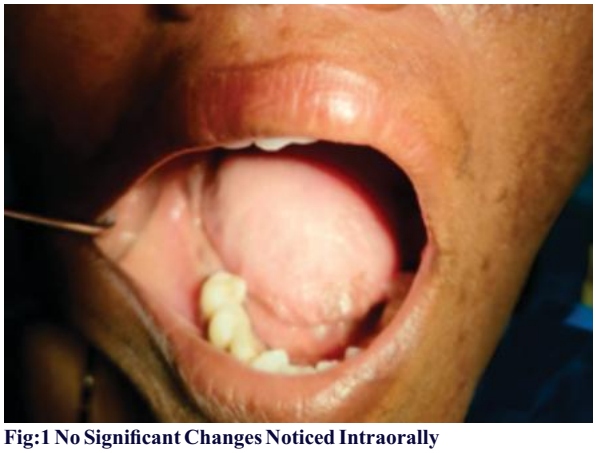

Fig:1 No Significant Changes Noticed Intraorally

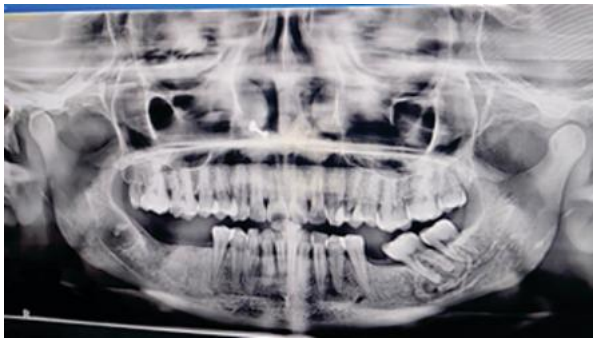

Fig:2 OPG Revealed Mixed Radio Opaque And Radiolucent Area.

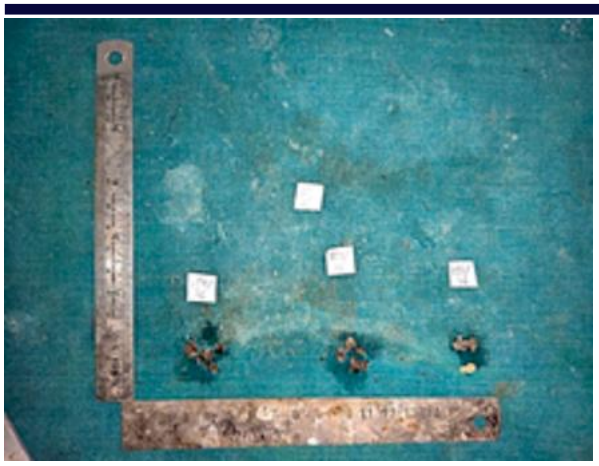

Fig:3 Gross Specimen Showing Multiple Soft Tissue and Hard Tissue.

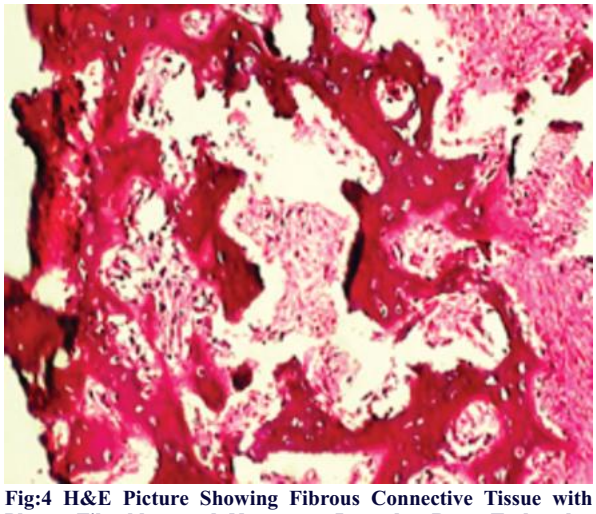

Fig:4 H&E Picture Showing Fibrous Connective Tissue with Plump Fibroblast and Numerous Irregular Bony Trabeculae Having Osteocytes within Lacunae and Without Osteoblastic Rimming.(20x)

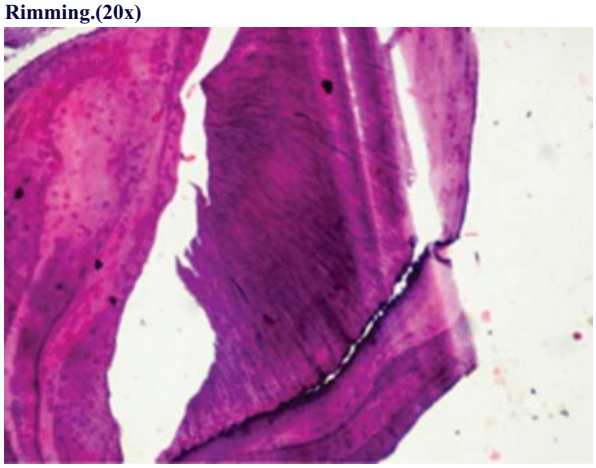

Fig:5 Decalcified Section Showed Irregular Eosinophilic Dentin with Well-defined Dentinal Tubules Associated with Loosely Arranged Fibrovascular Connective Tissue Resembling Dental Papilla(20x)

DISCUSSION

Odontogenesis is a complex and highly regulated process involving coordinated interactions between the odontogenic epithelium and ectomesenchyme. Disturbances during this process may give rise to odontogenic tumours (OTs), a diverse group of lesions derived from the epithelial, ectomesenchymal, and/or mesenchymal components of the tooth-forming apparatus. These lesions exhibit a broad spectrum of biological behaviour, ranging from hamartomatous proliferations and benign neoplasms to locally aggressive and, rarely, malignant tumours. Owing to their varied histopathological characteristics and clinical presentations, odontogenic tumours often pose significant diagnostic challenges[1].

Occasionally, odontogenic tumours present as a combination of two or more distinct odontogenic entities within the same lesion. Such lesions are described as hybrid odontogenic tumours, in which the histopathological features of two or more recognised odontogenic tumours coexist and intermingle within a common neoplasm. This differs from collision tumours, where two independent tumours arise separately and merely come into contact without histological integration. Accurate identification of hybrid odontogenic tumours is essential because they may mimic mixed odontogenic tumours or represent metaplastic changes, potentially leading to diagnostic errors and inappropriate management[3].

Literature reports odontome associated with the other odontogenic lesion like Adenomatoid Odontogenic Tumour (AOT)[4], Calcifying Epithelial Odontogenic Tumour (CEOT)[5] in the present case The coexistence of an odontome with ossifying fibroma imparts a unique clinicopathological dimension to this rare case, broadening the spectrum of hybrid odontogenic–fibro-osseous lesions.

Odontomes are developmental hamartomas composed of enamel, dentin, cementum, and pulp tissue and are generally divided into compound and complex variants depend up on the arrangement of hard tissue.[1]

The classification of fibro-osseous lesions has evolved considerably over the years. In the 1971 World Health Organization (WHO) classification, four cementum-containing lesions were recognized: fibrous dysplasia, ossifying fibroma, cementifying fibroma, and cemento-ossifying fibroma. In the subsequent WHO classification, benign fibro-osseous lesions of the jaws were categorized into osteogenic neoplasms and non-neoplastic bone lesions, with cementifying ossifying fibroma included among the osteogenic neoplasms. However, based on the overlapping clinical, radiographic, and histopathological features of cementifying and ossifying fibromas, the 2005 WHO classification unified these entities under the single term ossifying fibroma (OF)[67].

Ossifying fibroma is a benign fibro-osseous neoplasm of the craniofacial skeleton characterized by the replacement of normal bone with a cellular fibrous stroma containing varying amounts of mineralized tissue. Histologically, the lesion consists of fibroblastic connective tissue interspersed with immature woven bone, mature lamellar bone, osteoid, and rounded cementum-like calcifications, which may occur individually or in combination. These features reflect different stages of tissue maturation within the tumour[6].

Clinically, ossifying fibroma is a slow-growing, well-circumscribed neoplasm with the potential for progressive expansion if left untreated. It occurs most frequently in the premolar and molar regions of the mandible, predominantly affecting individuals in the third and fourth decades of life, with a marked female predilection. Most small lesions are asymptomatic and are detected incidentally during routine radiographic examinations. As the lesion enlarges, patients typically present with a painless bony swelling, resulting in cortical expansion and facial asymmetry, while pain and sensory disturbances are uncommon[1,6].

They are usually diagnosed during the first two decades of life and are frequently discovered incidentally because of delayed eruption or impaction of teeth. In contrast, ossifying fibromas commonly affect females during the third and fourth decades and predominantly involve the mandibular premolar-molar region. The present patient, a 40-year-old female with a posterior mandibular lesion, therefore corresponds well with the demographic profile of ossifying fibroma but differs from the usual age at presentation of odontome, further emphasizing the unusual coexistence of these lesions.

An interesting feature of the present case is that the lesion was detected incidentally during routine radiographic examination performed before prosthetic rehabilitation. Unlike most previously reported cases, which presented with facial asymmetry, cortical expansion, missing or impacted teeth, or progressive swelling, in the present case patient was completely asymptomatic and showed no clinically evident extraoral or intraoral abnormalities. This highlights the importance of routine panoramic radiography in identifying occult jaw

lesions before dental treatment.

Microscopically, the diagnosis was established by identifying two distinct yet intimately associated components. The fibro-osseous component exhibited densely collagenized fibrocellular connective tissue with plumb fibroblast and irregular trabeculae of bone without conspicuous osteoblastic rimming, whereas the odontogenic component consisted of dentinal tubules associated with loose fibrovascular tissue resembling dental papilla. These findings fulfilled the diagnostic criteria for ossifying fibroma associated with odontome. Similar microscopic combinations have been reported by Bakhtiari et al., Santosh et al., and Lina et al., although their cases demonstrated more extensive mineralization and associated impacted teeth[2,10,14].

To date, only few cases have been described. In the literature Ohtake et al. first reported multiple odontomes associated with ossifying fibroma in a 10-year-old boy[8]. Matsuo et al. later documented the subsequent development of ossifying fibroma following surgical removal of multiple odontomes, suggesting that residual odontogenic tissue or pluripotent mesenchymal cells might contribute to the later development of fibro-osseous neoplasia[9]. Bakhtiari et al. reported a compound odontome associated with ossifying fibroma in the anterior mandible of a 37-year-old female[14], while Santosh et al. described a hybrid tumour composed of complex odontome and central ossifying fibroma requiring en bloc resection[2], the present case was odontome with ossifying fibroma where conservative excision of the lesion was performed Other reports by Hosseini et al., Borghesi et al., Srivastava et al., and Lina et al. further demonstrate the rarity and clinicopathological diversity of this association.[Table-1][2]

Table - 1 Published Case Reports of Odontome Associated with Central Ossifying Fibroma (COF)

Author	Year	Age/ Sex	Location	Radiographic Features	Histopathologic Diagnosis
Ohtake et al.8	1993	10/M	Mandibular right posterior	Mixed radiolucent–radiopaque lesion	Multiple odontomes with central ossifying fibroma (COF)
Matsuo et al.9	2012	3/M	Mandibular left posterior	Well-defined unilocular radiolucency with buccal cortical expansion	Multiple odontomes coexisting with central ossifying fibroma (COF)
Lina et al10.	2016	19/M	Mandibular left posterior	Dense hyperopaque mass associated with ramus enlargement	Compound odontome with central ossifying fibroma (COF)
Hosseini et al11.	2016	46/F	Bilateral mandible	Multiple radiopaque and mixed-density lesions with anterior radiolucency	Central ossifying fibroma (COF) associated with odontome and cemento-osseous dysplasia
Bakhtiari et al14.	2016	37/F	Anterior mandible	Mixed radiolucent–radiopaque lesion with bicortical expansion and impacted canine	Compound odontome with central ossifying fibroma (COF)
Borghesi et al12.	2017	50/F	Mandibular right posterior	Pedunculated exophytic radiopaque mass with peripheral mixed-density rim	Peripheral osteoma, compound odontome, central ossifying fibroma (COF), and focal cemento-osseous dysplasia
Srivastava et al13.	2020	35/F	Bilateral mandibular posterior region	Radiopaque lesion on the left side and radiolucent lesion on the right side	Compound odontome with central ossifying fibroma (COF)
Santosh et al2.	2021	15/M	Mandible	Large mixed radiolucent–radiopaque expansile lesion with cortical expansion	Hybrid tumor comprising central ossifying fibroma and complex odontome

The pathogenesis of these hybrid lesions remains speculative. Several hypotheses have been proposed, including simultaneous development of two independent lesions, differentiation of pluripotent odontogenic ectomesenchymal cells into multiple tissue types, and induction of fibro-osseous proliferation by remnants of odontogenic tissue following odontome formation. Matsuo et al. suggested that immature elements of an odontome might subsequently develop into ossifying fibroma, whereas Ohtake et al. proposed that odontogenic mesenchyme surrounding developing odontomes could possess the potential for fibro-osseous differentiation. Alternatively, these lesions may simply represent coincidental coexistence within a common developmental field. At present, no molecular or genetic evidence conclusively supports any of these mechanisms, underscoring the need for future genomic and molecular studies[8].

Management depends primarily on the biological behaviour of the most aggressive component. While odontomes are effectively treated by simple enucleation, ossifying fibromas possess greater growth potential and require complete surgical excision with adequate curettage to minimize recurrence. Larger lesions may require segmental resection and reconstruction, whereas smaller, well-circumscribed lesions can be managed conservatively with complete removal and long-term follow-up, which was the treatment plan in the present case as the lesion was accidentally found and patient was devoid of any signs and symptoms related to lesion. In the present case, excisional biopsy was both diagnostic and therapeutic, and careful postoperative surveillance is recommended because long-term biological behaviour of hybrid lesions remains uncertain[11].

Another distinguishing feature of our case is the absence of an impacted tooth. Most previously reported odontome-associated ossifying fibromas have been closely related to impacted or unerupted teeth, supporting the developmental origin of odontomes. In contrast, the present case patient presented only with a missing tooth and no impacted tooth was identified radiographically. This observation indicates that the coexistence of odontome and ossifying fibroma should not be excluded solely because an impacted tooth is absent and expands the clinicopathological spectrum of these uncommon lesions.

Current evidence indicates that complete surgical excision remains the treatment of choice. Because odontomes behave as hamartomas with virtually no recurrence, the long-term prognosis is largely determined by the biological behaviour of the ossifying fibroma. Most reported hybrid lesions have been successfully managed by conservative enucleation and curettage, whereas larger lesions required segmental resection. Raza et al., in the largest recent series of hybrid odontogenic lesions, also reported excellent outcomes following complete surgical removal with no recurrences during follow-up[15]. Our lesion was localized and successfully treated by excisional biopsy, and periodic radiographic follow-up has been advised because long-term behaviour of odontome-associated ossifying fibroma remains insufficiently documented.

The present report contributes an additional example of this exceedingly uncommon hybrid lesion and expands its known clinical spectrum. Unlike previously documented cases, the lesion was identified incidentally in an asymptomatic middle-aged woman without cortical expansion or impacted teeth. This emphasizes that hybrid odontogenic lesions should be considered in the differential diagnosis of well-defined mixed radiolucent-radiopaque lesions of the jaws and that definitive diagnosis requires meticulous clinico-radiographic and histopathological correlation. Further accumulation of similar cases together with molecular investigations may clarify whether these lesions represent true hybrid neoplasms arising from a common progenitor cell or merely the coincidental coexistence of two independent pathological processes.

CONCLUSION

This case adds to the limited body of evidence on the rare coexistence of ossifying fibroma and odontome and broadens the recognized clinicopathological spectrum of hybrid odontogenic lesions. Reporting similar cases with long-term follow-up and molecular characterization will be instrumental in elucidating the pathogenesis of these uncommon lesions and determining whether they represent true hybrid neoplasms or the coincidental coexistence of two independent pathological entities.

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