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Case Report

Ossifying Fibroma with Coexistent Squamous Cell Carcinoma in Maxilla

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ABSTRACT

Introduction

Ossifying fibroma is a rare benign bone lesion characterized by gradual development and asymptomatic presentation, typically affecting craniofacial bones in individuals, particularly females, during their second to fourth decades of life. The concurrent occurrence of oral squamous cell carcinoma (OSCC) with ossifying fibroma is exceedingly rare, with only one previous case reported.

Case Report

We describe a 71-year-old male presenting with a gradually enlarging swelling in the oral cavity, initially treated five years prior for a mandibular fracture. Clinical examination revealed a large swelling in the right maxillary area, and imaging showed a radiolucent lesion with expansile characteristics. Histopathological analysis of the excised tissue initially suggested a benign fibro-osseous lesion. However, upon recurrence, a further biopsy revealed a coexistence of ossifying fibroma and well-differentiated squamous cell carcinoma in the previously affected palatal region.

Discussion

The simultaneous presence of ossifying fibroma and OSCC is extremely rare. This case underscores the importance of thorough diagnostic evaluation and highlights potential associations between trauma and tumor development. Further research is needed to elucidate the relationship between these lesions and improve diagnostic and therapeutic approaches for such rare occurrences.

Keywords

Fibro-osseous Lesion; Maxillary Sinus; Ossifying Fibroma; Oral Squamous Cell Carcinoma

Ossifying fibroma is an uncommon benign bone lesion characterized by its gradual development, asymptomatic presentation, and predilection for affecting craniofacial bones. It is particularly seen in females, during the second and fourth decades of life.[1,2] The simultaneous presence of Oral squamous cell carcinoma (OSCC) with ossifying fibroma is exceedingly rare, and to the best of our knowledge with only one documented case reported by Karube et al.[3] This case report presents a similar co-occurrence of lesions in a 71-year-old male patient.

Case Presentation

Seven months back, a 71-year-old gentleman presented to the hospital with complaints of swelling inside his oral cavity that had developed gradually over the past year. He gave a history of a traumatic fracture of the mandible 5 years back which was treated by internal fixation. Clinical examination revealed a 5 x 3 x 2 cm swelling in the right maxillary area (Fig. 1), extending from the lateral aspect of the nose to the infraorbital region with granular swelling on the palate with erythematous surfaces. The expansion of the swelling had led to enlargement of the buccal cortical plate, obliteration of the vestibule, and distortion of the nasolabial fold.

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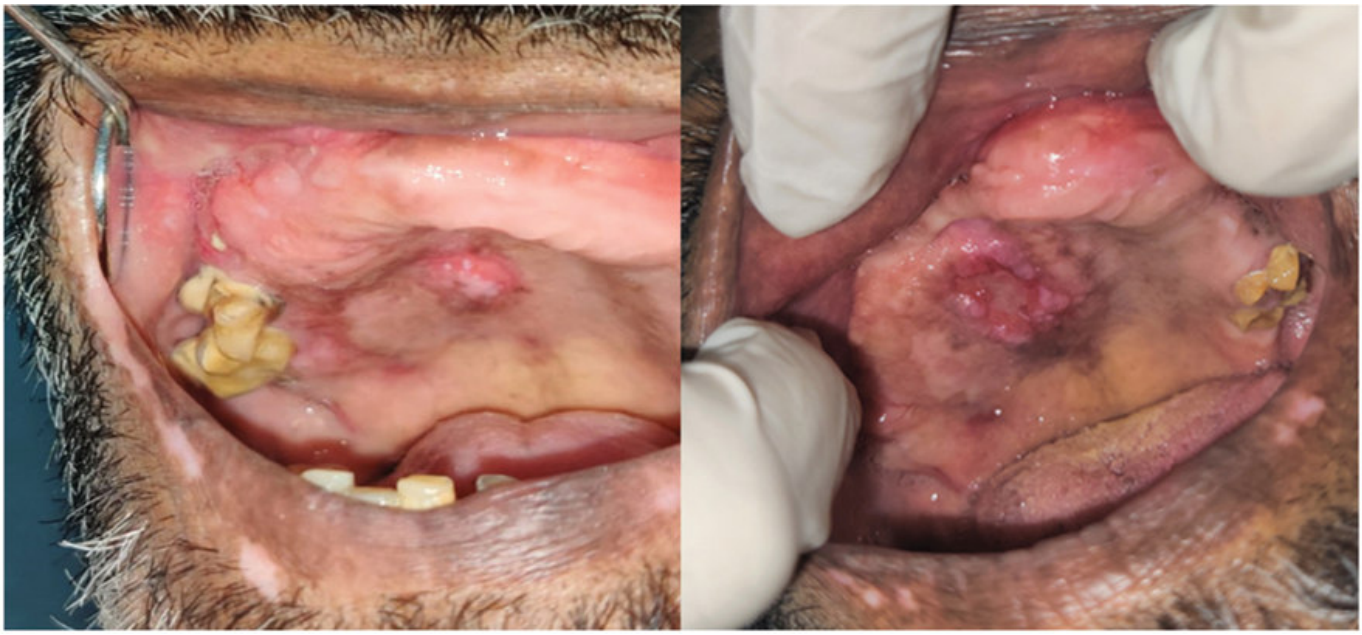

Fig. 1. Clinical picture of the initial presentation of the lesion in the oral cavity with a palatal ulcer.

OPG revealed a radiolucent lesion in the right upper jaw (Fig. 2), and he underwent excision under GA at that time. Histopathological examination of the excised tissue revealed a cellular spindle cell neoplasm with low mitotic activity supporting the diagnosis of a benign fibro-osseous lesion.

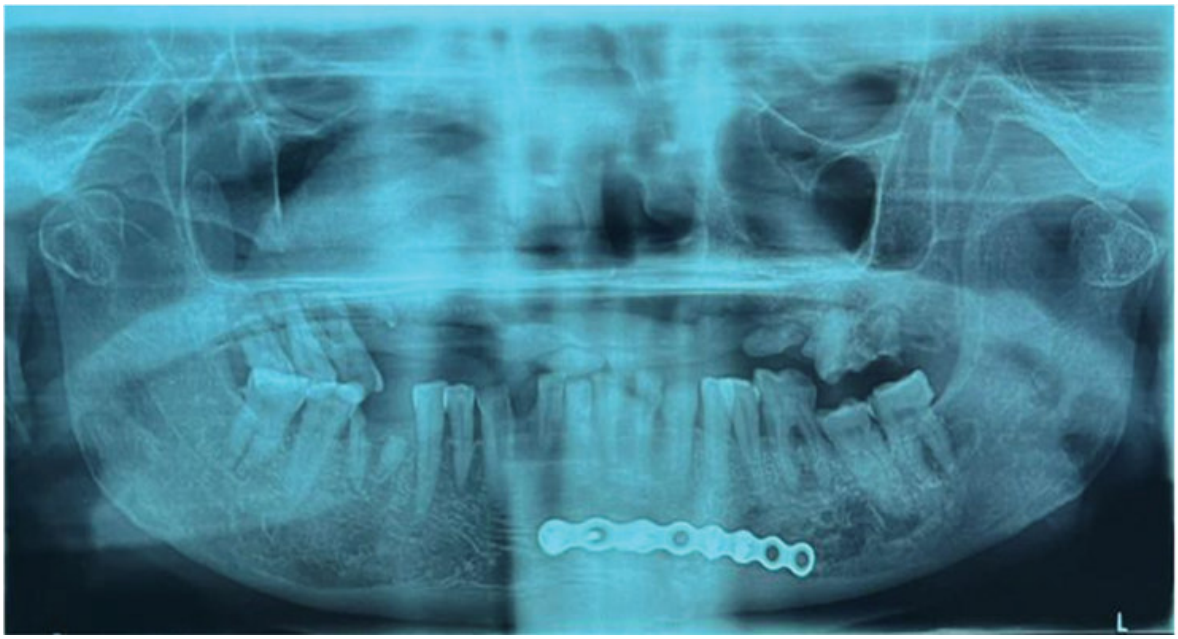

Fig. 2. OPG showing radiolucent lesion in right maxilla.

Now he has presented again with, swelling on the right side of the face. Intraorally, a 4 x 4 cm swelling was identified on the hard palate anteriorly in the prior excision scar region, exhibiting tenderness, a hard consistency, and granular and erythematous surface characteristics. Contrast-enhanced computed tomography (CECT) revealed bony expansion with lytic areas and ground glass attenuation in the right maxilla, extending into adjacent regions. The lesion displayed an expansile pattern involving the anterolateral and posteromedial walls of the right maxillary sinus (Fig. 3). A soft tissue component extended into various surrounding structures, notably the premaxillary soft tissue, gingival mucosa, gingivobuccal sulcus, and gingivolabial sulcus.

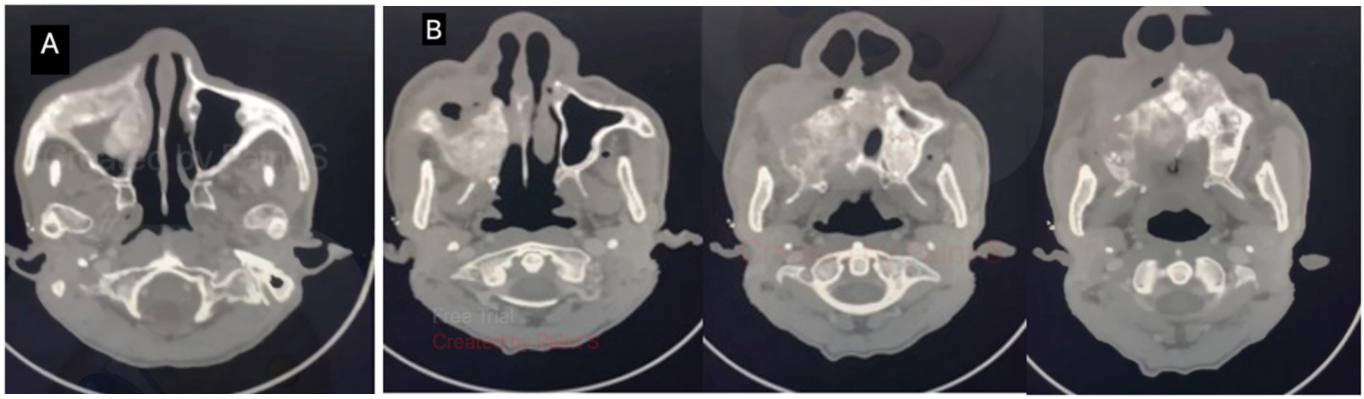

Fig. 3a. CT Axial sections of the face showing an expansive ground glass attenuating lesion involving the anterolateral, posteromedial wall of the right maxillary sinus, canine fossa, and canine eminence of the maxilla.

Fig. 3b. Ill-defined lytic lesions involving posterolateral maxilla with extensive soft tissue component -likely coexisting squamous cell carcinoma.

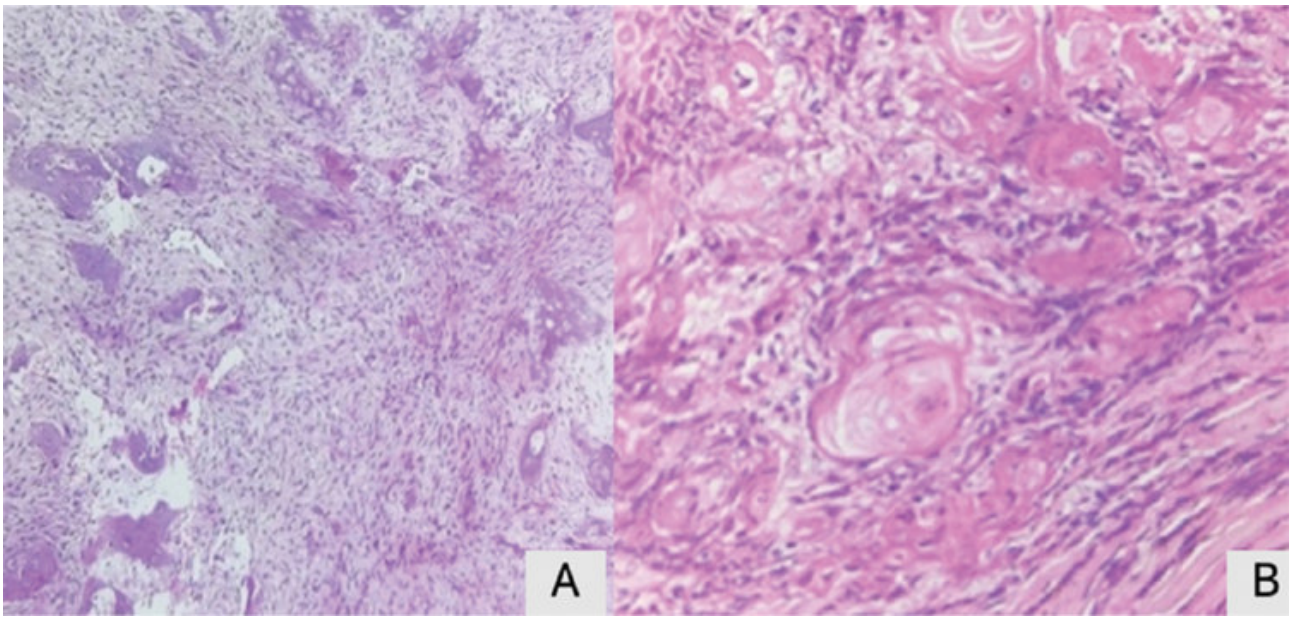

Fig. 4a. H and E stain (10X) Photomicrograph shows tumor epithelial islands invading into connective tissue.

Fig. 4b. H and E staining (40 X) Photomicrograph shows bony trabeculae dispersed in cellular stroma.

An incision biopsy revealed the following: The dysplastic, stratified squamous epithelium was observed overlying a fibrous connective tissue stroma. Epithelial cells showed proliferation into the superficial lamina propria as nests and islands, accompanied by focal chronic inflammatory cell infiltration (Fig. 4A). An area of hypercellular connective tissue with irregular bony trabeculae containing osteocytes and occasional giant cells was identified (Fig. 4B). Moderate vascularity and the presence of mucus lobules and salivary gland acini were observed. Based on the histopathological analysis, the mucosal lesion was diagnosed as a benign ossifying lesion with superficial epithelial features consistent with well-differentiated squamous cell carcinoma. The coexistence of a squamous cell carcinoma over a palatal ossifying fibroma is a very rare occurrence and hence presented here.

Discussion

Lesions like ossifying fibroma occur when normal bone tissue is replaced by fibroblasts and collagen fibers, with altering amounts of mineralized material.[4] The origin of ossifying fibroma can be attributed to various factors such as uncontrolled multiplication of periodontal ligament (PDL)cells, metaplastic process arising in connective tissue fibers or steered by a fault in the tissue induction process, trauma, periodontitis, or previous extractions.[4] The frequent location of these lesions is the mandibular premolar-molar area and only 30% of cases are in the maxilla.[1,4] The maxillary ossifying fibroma is prone to recurrence and is more aggressive. [5] Ossifying fibroma are seen in imaging as round or oval well-defined, expansile mass with a corticated border and a variable degree of central density. [6] Previous research has shown that the radiographic borders of ossifying fibroma appear relatively smooth, well-defined, and mostly corticated. The lesion exhibits a regular contour and tends to show concentric growth within the medullary section of the bone, expanding outward in all directions.[7]

In the maxilla, ossifying fibroma most often appears in the canine fossa and zygomatic arch area. The clinical presentation of these lesions can range from being relatively inert to displaying aggressive behavior.[8] The lesion is usually asymptomatic at discovery.[7] However, the growth of cemento-ossifying fibroma can produce a noticeable swelling and slight deformity, with the displacement of teeth being an early clinical indicator.[9] Central cemento-ossifying fibromas usually present as solitary and well-defined lesions. Initially, they present as radiolucent lesions without any apparent internal radiopacities. As the tumor progresses, noticeable calcification occurs, causing the radiolucent area to be flecked with opacities, ultimately transforming the lesion into an extremely radiopaque mass.

The coexistence of ossifying fibroma and OSCC, in this case, is exceptionally rare; this may be the second case in the literature.[3] Both cases have co-existential squamous cell carcinoma and maxillary ossifying fibroma. As ossifying fibroma is a mesenchymal tumor, there has been speculation that the presence of squamous cell carcinoma, in this case, could be linked to trauma or other stimuli. However, the exact etiology of the coexistence of ossifying fibroma and squamous cell carcinoma remains uncertain and must be examined by accumulating additional cases to establish a definitive understanding. This synchronous occurrence adds complexity to both diagnostic and therapeutic approaches.

Thorough histopathological examination and immunohistochemical analysis remain crucial in elucidating the unique characteristics of such rare entities and guiding appropriate treatment strategies.

Conclusion

This case highlights the uncommon convergence of ossifying fibroma and squamous cell carcinoma within the maxilla, a scarcely documented phenomenon in the medical literature.

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