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Dual Manifestation: A Rare Case Report On Combined Extra-Osseous And Intra-Osseous Lesion Of The Mandible

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ABSTRACT

The purpose of this case report is to describe a co-existing association between intra-osseous cemento-ossifying fibroma (COF), an aneurysmal bone cyst (ABC) and extra-osseous pyogenic granuloma and review the literature regarding associated benign fibro-osseous lesions (FOLs) and cysts. A 45-year-old woman presented with asymptomatic swelling in the left side of the mandible with no history of trauma. The extraosseous soft tissue lesion was 20x15x10mm in measurement. Radiographic investigation revealed a unilocular radiolucent area with radiopaque foci. A complete excisional biopsy was planned under general anaesthesia. Microscopic examination revealed a COF with secondary ABC and pyogenic granuloma. The 3-month follow-up showed no signs of recurrence.

KEYWORDS: *Pyogenic granuloma, Cement-ossifying fibroma (COF), Aneurysmal bone cyst (ABC), Intermixed lesion, Collision tumour.*

INTRODUCTION

Pyogenic granuloma (PG), sometimes known as granuloma pyogenicum, refers to a common, acquired, benign vascular tumour that arises in tissues such as the skin and mucous membranes.[1,2] The scientifically accurate term for this entity is lobular capillary hemangioma.[1,3] In the past, pyogenic granulomas were thought to be an exaggerated granulomatous reaction to an infectious or pyogenic insult, which led to the use of terms such as 'pyogenic granuloma' and 'granuloma pyogenicum'. However, the term pyogenic granuloma is a misnomer that may initially cause confusion. Multiple factors are involved in the etiopathogenesis of this entity, but the exact cause is unknown.

Cemento-ossifying fibroma (COF) is a benign fibro-osseous neoplasm that can arise from any part of the facial skeleton, although the majority occur in tooth bearing areas, particularly the mandible. More common among women between the third and fourth decades of life, these lesions present as a slow-growing, asymptomatic, intrabony mass. [4,5,6] If left untreated, COF may affect adjacent teeth, causing root resorption, displacement and eventually significant functional and aesthetic deformity and pain.[4,6] Radiographically, COF can show a number of patterns depending on the degree of mineralization, though commonly seen as round or oval, unilocular or multilocular, mixed lesion containing variable amounts of radiopaque material.[4,7]

The aneurysmal bone cyst (ABC) is a benign, osteolytic, rapidly growing, expansile, and destructive lesion of the bone[8]. Characterized by the replacement of normal bone with fibro-osseous tissue containing blood-filled sinusoidal or cavernous spaces[9,10]. As such, ABCs present radiographically as expansile unilocular of multilocular, lytic lesions, sometimes with bony septa[10]. ABCs predominantly occur in the long bones and occurrence in the

jaws is extremely rare, consisting of only 0.5%–1.5 % of all jaw cysts[4,9,10]. The average age of occurrence is about 13yrs, with the majority of patients being less than 20 years old [4,10]. Histologically, ABCs present as an expanding osteolytic lesion containing blood-filled spaces, separated by connective tissue, bone trabeculae and many osteoclast like giant cells [4,8,9]. Furthermore, ABCs are considered a pseudo cyst due to the absence of an epithelial lining [4,9]. When treated with enucleation and curettage, reports of recurrence are commonly reported between 20 and 30 % [4,9,10]. As such, recommended treatment for ABC is complete excision [4,9,10]. ABCs may arise as true mesenchymal neoplasms, termed "primary ABC", or they can develop concurrently with pre-existing bone lesions. ABCs occurring secondary to pre-existing bone lesions are termed "secondary ABCs" [4,8,10]. Secondary ABCs show similar characteristics as primary ABCs although have additional histological findings indicating the presence of an additional coexisting or intermixed lesion [4,8,10]. According to Yeom and Yoon, 70 % of ABC cases are primary, whereas 30 % are secondary [4,8].

Here we present a rare case report of a 45yr old female patient who complained of a soft swelling inside her mouth since 15 days which gradually increased in size and started coming in between her teeth while chewing. This case highlights a novel finding that may influence diagnostic accuracy and therapeutic decision-making. The following description details the clinical presentation, radiographic imaging and eventual treatment of this extraordinary co-existing extra and intraosseous lesion of left mandible.

CASE REPORT

A 45-year-old female presented to the outpatient department (OPD) of Oral and Maxillofacial Surgery with a chief complaint of a painless, soft-growing mass in the left lower jaw region of her oral cavity for approximately 15 days. The growth was initially small in size and slowly growing attaining the present size. The patient did not have any specific medical or familial history.

Extraoral examination did not reveal any swelling, expansion, cutaneous changes or cervical lymphadenopathy and there was no motor or sensory deficit.

On intraoral examination, a solitary, red, extra-osseous, soft, non-tender, pedunculated growth was noted with respect to 36-37 tooth region measuring approximately 2x1.5x1cm (Figure 1). Bleeding on provocation was positive. Moderate amount of calculus and stains was present intraorally. Clinical examination also revealed bony-hard, non-tender expansion of left lower buccal cortex along with normal dentition and occlusion.

Radiographic examination showed a well-defined, unilocular, minimally expansile, radiolucent along with few flecks of radiopacities lesion with respect to periapical region of 36,37,38 region (Figure2) measuring approximately-23.37mm (Mesio-Distal) x22.81mm (Supero-Inferior) x16.93mm (Bucco-Lingual) (Figure 3). Supero-inferiorly the lesion was extending from the alveolar crest till the inferior border of mandible Bucco-lingually it was extending from the buccal cortex till the lingual cortex. Margins were partially corticated and scalloped. Perforation of lingual cortex was present in 37 region. (Figure 4) Horizontal impaction of 38 and apical root resorption of molars was found on cone beam computed tomography (CBCT).

A complete excision of the extra-osseous soft tissue overgrowth was done, involved teeth were removed and curettage of the intra-osseous lesion was performed under general anaesthesia and the tissue was sent for histopathologic evaluation. (Figure 5-10)

SURGICAL APPROACH -

A pre-operative antibiotic coverage – Inj. Augmentin 1.2gm IV, Inj. Metronidazole 100cc IV was given. Patient was taken into the operation theatre and intubated via naso-endotracheal route. All routine aseptic procedures were done. Injection 2% lignocaine hydrochloride with adrenaline (1:80,000) 3ml + Inj. Dexamethasone 8 mg (1ml) – Twin mix was injected intraorally with respect to the lower arch in the form of nerve blocks and local infiltrations. The incision was marked and taken after which a full thickness trapezoidal mucoperiosteal flap was raised. The extra-osseous lesion was excised and 3 posterior teeth (36, 37, 38) were extracted. Intra-osseous lesion was exposed and carefully dissected along the length of inferior alveolar neurovascular bundle. Inferior alveolar nerve bundle was preserved while excising the lesion. Peripheral ostectomy was done. Thorough povidone iodine (10%) and normal saline irrigation done. An absorbable gelatin sponge was filled in the cavity and haemostasis was achieved. Closure was done using 3-0 Vicryl (absorbable sutures). Patient was extubated uneventfully. The extra-osseous and intra-osseous specimen was sent for histopathologic evaluation. (Figure 5-10)

HISTOPATHOLOGY FINDINGS – (Figure 11-12)

Extra-osseous lesion – The sections show parakeratotic stratified squamous epithelium that is ulcerated at places. Proliferation of numerous budding vascular channels and endothelial cells are observed. Intense mixed inflammatory cell reaction is noted within the connective tissue core. (Figure 11)

Intra-osseous lesion – The sections show numerous ossicles, woven bony trabeculae and cementicle like areas within the fibrocellular connective tissue core. Numerous cavernous, dilated, haemorrhagic spaces are seen in the current section. (Figure 12)

DISCUSSION

The cemento-ossifying fibromas (COF) of the jaws are well circumscribed, generally slow-growing, benign lesions which enlarge in an expansive manner. On occasion, they may reach a large size and may result in considerable deformity. The histological pattern of these lesions varies with the stages.[11] In most reported cases ossifying and cemento-ossifying fibromas occur as a solitary lesion. The term" cemento-ossifying fibroma" is used to describe fibrous lesions containing calcifications with strong similarity between bone and cementum. A COF can be classified as a peripheral cemento-ossifying fibroma (PCOF) or a central cemento-ossifying-fibroma (CCOF). The central type arises from the cells of the periodontal ligament in the apical area, causing the expansion of the lamina dura. The peripheral type occurs in the soft tissues of the teeth-bearing areas.[6,12]

Aneurysmal bone cyst (ABC) has been recognized since 1893 when it was described as an ossifying hematoma by Van Arsdale.[13] Jaffe and Lichtenstein were the first to recognize ABC as an intraosseous, osteolytic lesion, chiefly affecting the metaphyseal region of long bones and vertebrae. Bernier and Bhaskar described the first case of ABC in the jaws in 1958.[14,15] Jaffe and Lichentenstein refer to alterations in local hemodynamics causing increased venous pressures and engorgement of the vascular bed in the transformed bone, leading to resorption, connective tissue replacement and osteoid formation.[9] Hernandez et al. classified ABC as primary and secondary. Primary could be congenital or acquired and could originate from pre-existing AV malformations. The secondary type is postulated to be associated with degeneration of pre-existing lesions such as a cyst, tumour or fibrosseous lesion.[13]

The radiological features of ABC in the jaws are quite conflicting; the bone is expanded, appears cystic resembling a honeycomb or soap bubble and is eccentrically ballooned. There may be destruction or perforation of the cortex, and a periosteal reaction may be evident.[16]

In our case, a unilocular radiolucency causing expansion of the cortical plates and thinning of the lingual cortex of the mandible with root resorption of the involved teeth was present. The diagnosis based on radiographic appearance is impossible because there are other lesions having similar radiographic appearance, such as ameloblastoma, myxoma, central giant cell granuloma, odontogenic cysts or central hemangiomas of the bone.[17]

A case of Peripheral Ossifying Fibroma evolving from Pyogenic Granuloma was reported by Godinho GV et al.[18] However, to date, there is no published literature demonstrating the evolution of Central Ossifying Fibroma (COF) or Aneurysmal Bone Cyst (ABC) from Pyogenic Granuloma. This highlights the rarity and possible novelty of such a pathological progression in the present case.

A literature review of thirty-two ABC-plus-lesions revealed that males are more commonly affected, and there is a higher propensity for mandibular involvement. Sixty-eight percent of the thirty-two ABC plus-lesions were associated with Fibro-osseous lesions (FOLs), while giant cell lesions accounted for 32% of the cases.[16](TABLE 1)

Treatment of COF with secondary ABC and pyogenic granuloma is usually directed toward complete removal of the lesion. The treatment modalities are percutaneous sclerotherapy, diagnostic and therapeutic embolization, curettage, block resection and reconstruction, radiotherapy and systemic calcitonin therapy.[19] The present case was treated by curettage and regularly monitored. There was no evidence of any residual lesion after 3 months of follow-up.

AGE	1ST DECADE OF LIFE - 6	2ND DECADE OF LIFE - 15	3RD DECADE OF LIFE - 7	4TH DECADE OF LIFE - 0	5TH DECADE OF LIFE - 3	6TH DECADE OF LIFE - 1	
SEX	MALE - 19	FEMALE - 13					
SITE	MAXILLA – 9	MANDIBLE - 23					
ASSOCISTED FOL WITH ABC	GIANT CELL GRANULOMA - 4	OSSIFYING FIBROMA – 6 NON-OSSIFYING FIBROMA – 1	FIBROUS DYSPLASIA – 5	CEMENTO-OSSIFYING FIBROMA - 11	CENTRAL GIANT CELL GRANULOMA - 2	BENIGN OSTEOBLASTOMA - 1	JUVENILE OSSIFYING FIBROMA - 2

TABLE 1 – REVIEW OF ABC PLUS CASE REPORTS IN THE HEAD AND NECK REGION (1964-2022) [20]

CONCLUSION –

As the radiological features of COF with secondary ABC are varied, resembling many lesions, histopathologic analysis is a must for the diagnosis. This case of ABC associated with COF provides an insight for the diagnostic process of radiographically mixed lesions with cystic changes.

To date, there is no literature demonstrating any interrelationship or pathological overlap among Central Ossifying Fibroma (COF), Aneurysmal Bone Cyst (ABC), and Pyogenic Granuloma. This case represents a unique convergence of these lesions, underscoring an area of pathology that remains unexplored and warrants further investigation.

Authors' contributions –

C.G., M.P. - interpreted the radiologic data, surgical and medical management.
K.P., D.P., R.L., S.S - wrote the manuscript and collected the data.
All authors have read and approved the manuscript.
Availability of data and materials –
All data analyzed during this study are included in this published article.

Consent for publication -

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests –

The authors declare that they have no competing interests.

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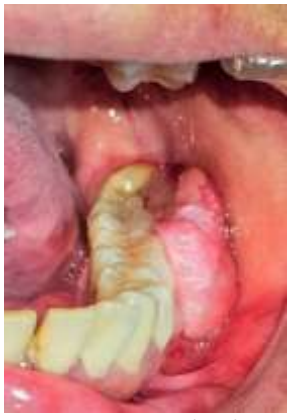

Figure 1- Frontal view showing extension of soft tissue lesion from 36-38 tooth.

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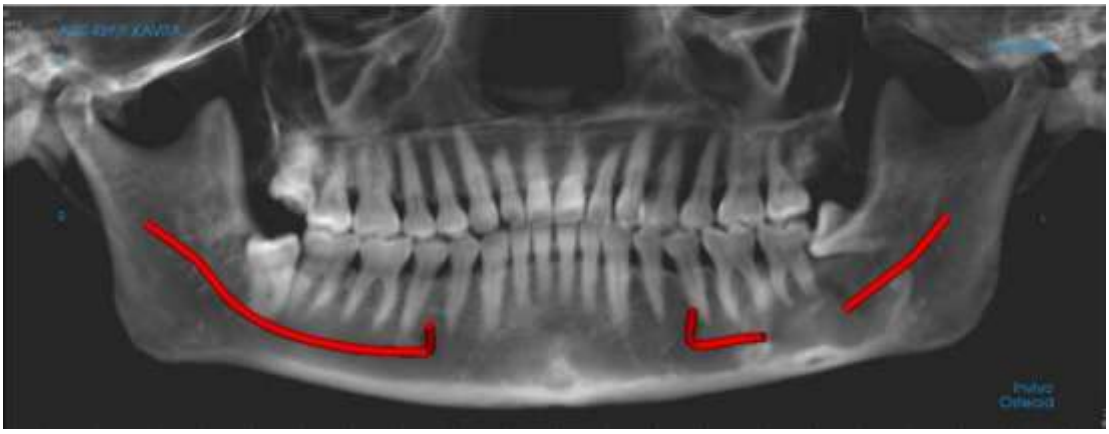

Figure 2 - Orthopantomogram view showing an intra-osseous lesion with respect to left mandible region (36,37,38) + Horizontal Impaction of 38

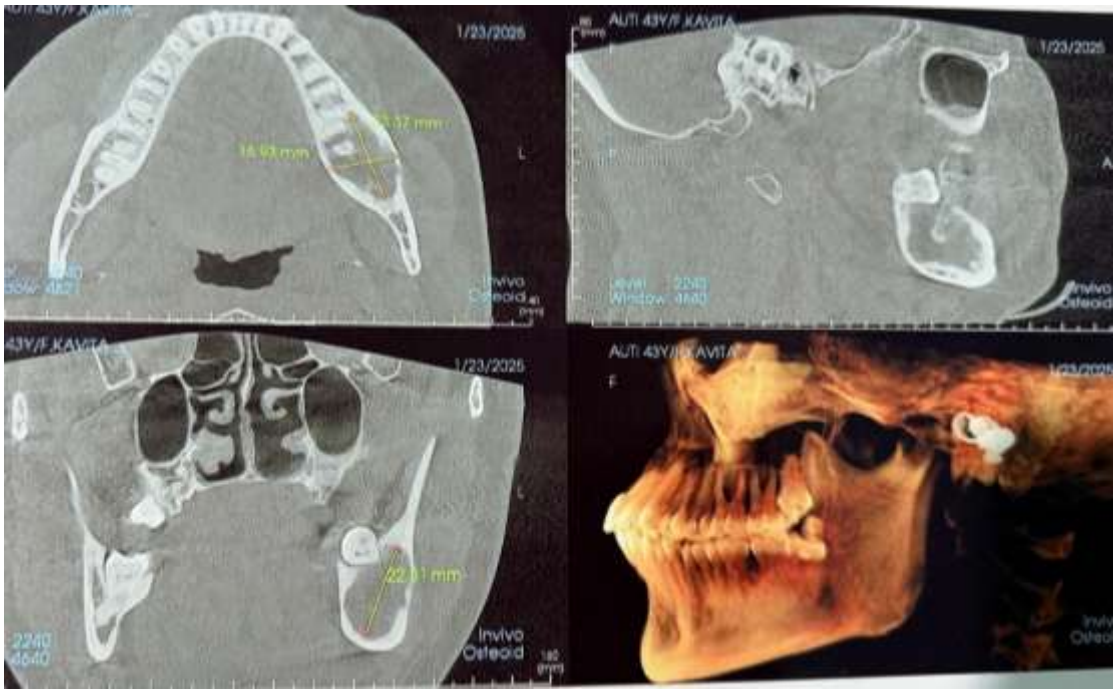

Figure 3 – CBCT cuts showing measurements of the intra-osseous lesion

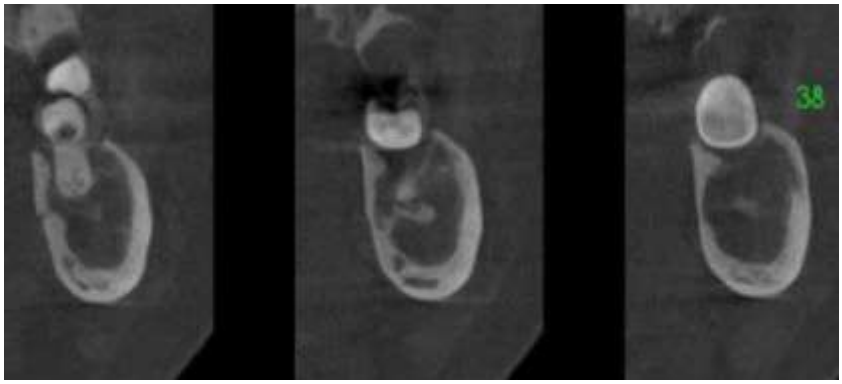

Figure 4 – CBCT cut showing a radiolucent Bucco-Lingual expansile intra-osseous lesion along with flecks of radiopacities +Perforation of lingual cortex in 37 region

Figure 5 – Intra-operative photograph

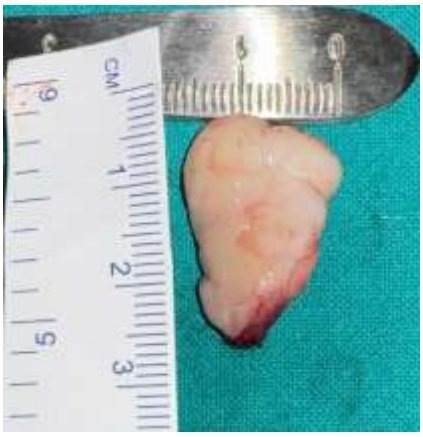

Figure 6 - Macroscopic image of excised lesion showing an irregular aspect and lobulated surface

Figure 7- Extracted teeth associated with the lesion

Figure 8 - Intraoperative photograph after the extraction of involved teeth

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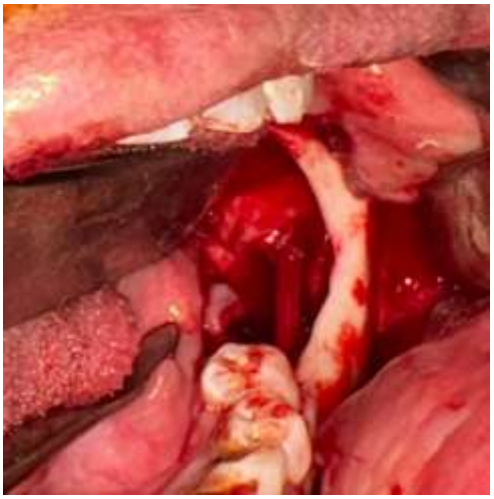

Figure 9 – Intra-operative photograph after curettage

Figure 10 – Macroscopic image of excised intra-osseous lesion

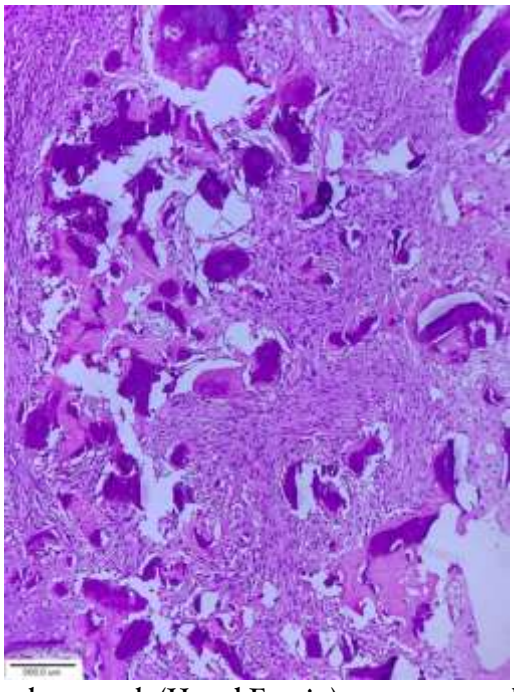

Figure 11 – Histopathologic slide photograph (H and E stain)- extra-osseous lesion (Pyogenic granuloma)

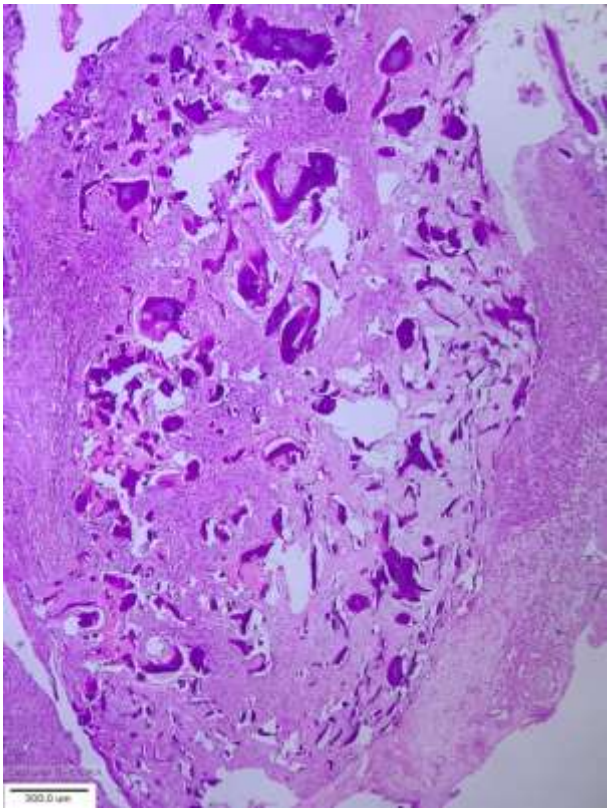

Figure 12 – Histopathologic slide photograph (H and E stain)- Intra-osseous lesion (Cemento-ossifying fibroma with secondary aneurysmal bone cyst)

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