VOLUME - 11, ISSUE - 04, APRIL - 2022 • PRINT ISSN No. 2277 - 8160 • DOI : 10.36106/gjra

Original Research Paper

Maxillofacial Surgery

CALCIFYING EPITHELIAL ODONTOGENIC TUMOR: REPORT OF A CASE AND REVIEW OF LITERATURE

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ABSTRACT The calcifying epithelial odontogenic tumour (CEOT), also known as Pindborg's tumour. It is a rare benign neoplasm of locally aggressive behaviour originating from stratum intermedium. CEOT accounts for approximately 0.4 – 3 % of all the odontogenic tumours' most frequent location is mandibular premolar and molar area; less frequently, in the maxilla, typically formed in the fourth to fifth decades. It usually presents as a slow growing, firm, painless mass and it shares many features with ameloblastoma. Due to a fairly common tendency of recurrence, estimated to be approximately 14 %, the preferred choice of treatment is radical surgical procedure and postoperative follow up . This case report presents the clinical, radiological findings, histopathology, and surgical treatment of the patient with pindborg tumour were presented. The report is supplemented by a review of literature.

KEYWORDS : Orbit, Fracture, Zygomaticomaxillary complex, Titanium mesh, Reconstruction

INTRODUCTION

CEOT is a rare benign odontogenic neoplasm of the jaws. Pindborg described this lesion as a separate clinicopathological entity. Clinically CEOT manifests as an intraosseous lesion in 95% of cases and extraosseous for less than 5%(Bouckaert, M. M. R,2000). The majority are associated with impacted or unerupted teeth. The age of the patients affected ranges from 8-92 years (Philipsen, H. P., 2000). The most common radiographic finding is a well-defined unilocular radiolucency, which resembles a dentigerous cyst. The neoplasm appears as a multilocular lesion mimicking ameloblastoma (Buyukkurt 2014). The surgical procedure involves conservative enucleation, marginal or partial or rarely composite resections in cases showing malignant transformation and invasion (Sedghizadeh, P.P 2007).

Case Report

A 25-year-old female patient with a complaint of facial asymmetry was referred to the Department of Oral and Maxillofacial Surgery with one and half-year-old history of non-fluctuant painless mandibular left side swelling. No regional nerve paresthesia or tenderness over the region was present. The extraoral inspection revealed facial asymmetry on the left lower third region, an oval swelling measuring approximately 3*2cms over the left body of the mandible. The skin over the swelling was slightly stretched with no secondary change. Submental and submandibular lymph nodes were not palpable. Intraoral examination revealed a diffused swelling present in the lower left body region extending anteroposteriorly up to the buccal gingival sulcus of tooth numbers #34 to 36. Supero- inferiorly the mucosa overlying the lesion was intact and teeth in the vicinity showed grade II mobility with tooth numbers 35 & 36. No discoloration, no tenderness and responded positively to vitality tests. Interincisal mouth opening was 40mm.

Her medical and dental history was non-contributory. General examination revealed a moderately built and nourished individual with a normal gait. Vital signs within normal range. No localised rise in temperature were noticed. Radiographical examination.

On a panoramic radiograph, a 2*2cms multilocular radiolucency with a sclerotic border involving the left body with a honeycomb appearance at the site of tooth numbers 34,35 and 36. Cone-beam computed tomography showed multilocular radiolucent lesions about 40*20 mm in diameter involving both buccal and lingual cortical plates. After clinical and radiological examination, incisional soft tissue biopsy was done with ameloblastoma, odontogenic myxoma or CEOT prediagnosis.

Histopathological Examination

Showed epithelial polyhedral cell islands with pleomorphism and prominent intercellular bridges. Areas of homogenous eosinophilic amyloid-like material are present. Concentric lamellar calcifications which are characteristic of this lesion were seen among epithelial polyhedral cell islands.

Surgical Treatment

After general anesthesia using nasal intubation, the intraorally crevicular incision was given from the 33 to 37 region with the vertical relieving incision in 32 region. Extraction was done irt #34 , 35 & 36. Enucleation of the tumour was done aggressively with a clear margin. The gross specimen measured 3*3 cm and was well-encapsulated round to ovoid cystic mass. It revealed regular borders, a smooth surface and firm to hard inconsistency. The reconstruction plate was carefully fixed in position. The postoperative course showed no evidence of recurrence after the surgical procedure.

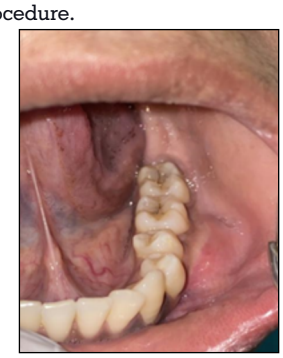

Figure 1 Intraoral Mucosal Swelling

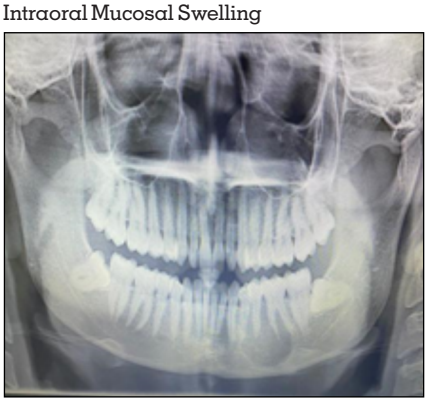

Figure 2 OPG

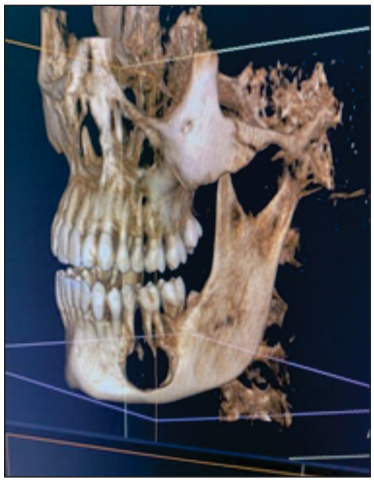

Figure 3 Cbct

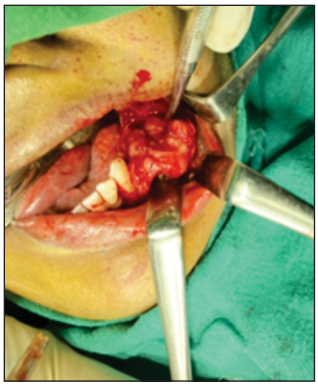

Figure 4 Surgical Excision

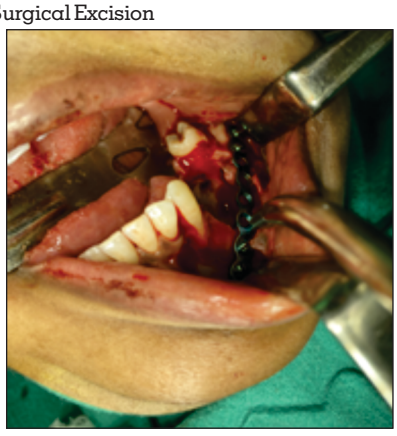

Figure 5 Reconstruction Of Plate

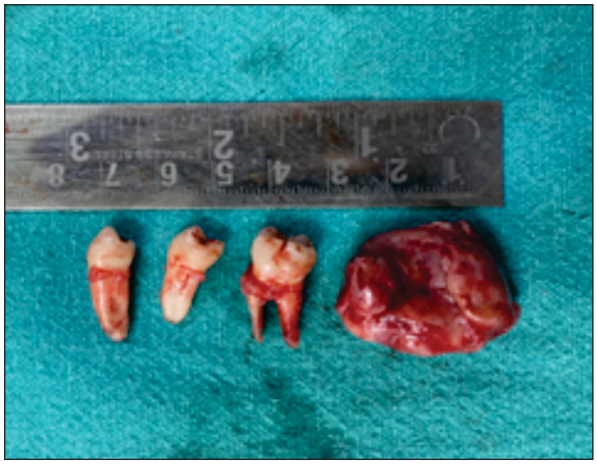

Figure 6: Excision Specimen

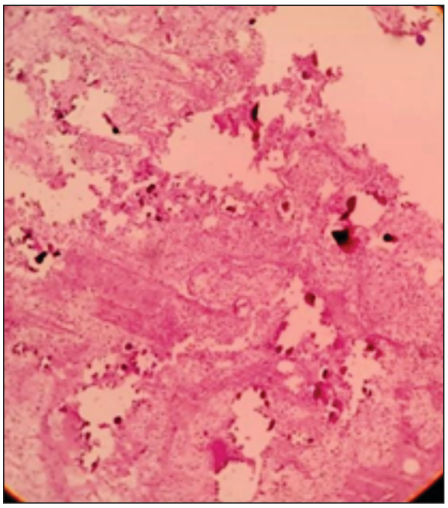

Figure 7 Histological Specimen

DISCUSSION

CEOT is a rare benign epithelial odontogenic neoplasm that was first described by Pindborg in 1955. Though the tumour is benign, a few cases have been reported as locally aggressive, invading surrounding soft tissues and bone. It accounts for less than 1% of all odontogenic tumours (Bouckaert, M. M. R.,2000) Most investigators believe that the tumour cells originate from the reduced enamel epithelium, but today they believe they originate from the stratum intermedium as cellular morphology is similar to tumour cells and they agree that the central type is usually located in the premolar and molar regions with a mandibular to the maxillary ratio of 2:1(Houston, G. D 1997).

They tend to occur over a wide age range .Predominate in the 3rd to 6th decades of life. With almost equal sex predilection. CEOTs are slow-growing, expansile, painless masses that cause expansion of the cortical plates but occasionally patients may report pain, epistaxis, nasal stuffiness etc (Neville 2002). The extraosseous variant usually presents as nodular swelling. The intraosseous variant is often easily enucleated and varies in size from 1 to 4 cm in diameter (Karabit, Z. 2017). The mass is usually greyish-white in colour, bisection of which reveals multiple calcified particles which produce a crunching sound on cutting. The tumour may be solid or contain minute cystic spaces with the associated unerupted tooth being present within the tumour mass (Houston, G. D 1997).

The lesion becomes multilocular with a honeycomb patternin some cases. In others, multiple radio opacities are seen within the radiolucent area, giving rise to the term "driven snow appearance"(Sedghizadeh, P. P 2007).

The histopathology is unique consisting of sheets, nests and masses of polyhedral epithelial cells with abundant eosinophilic cytoplasm and prominent intercellular bridges. The cytomorphology of the cells may suggest malignancy as they exhibit significant cellular and nuclear pleomorphism, prominent nucleoli and scattered giant cells. Nevertheless, mitotic figures are rare. A characteristic feature is the presence of homogenous eosinophilic 'amyloid like' material interspersed between the cells; which stains positively with Congo red and exhibit apple-green birefringence under polarized microscopy. This material undergoes calcification in the form of concentric 'Leisgang rings' that are pathognomic of this tumors. Occasionally, extensive clear cell differentiation has also been reported (Houston, G. D 1997).

Recently, newer variants of CEOT like non-calcifying CEOT with Langerhans cells, CEOT displaying cementum and bone-

like material and a combined adenomatoid odontogenic tumour and CEOT have also been described. The clear-cell CEOT variant is more aggressive with a higher recurrence rate (22%), and some would consider this form to be a low-grade odontogenic carcinoma (Bouckaert, M. M. R,2000).

Treatment options for CEOT have ranged from simple enucleation to radical and extensive resection (Nelson 1992). Several authors initially advocated aggressive treatment, but increasingly, histologic information shows that this tumour does not appear to extend into the intertrabecular bony spaces as does ameloblastoma; therefore, a more conservative approach is warranted. Sadeghi and Hopper believe the surgical treatment of CEOT should be guided by the site, size, and histologic features of the lesion (Bouckaert, M. M. R,2000).

The prognosis of the CEOT is good with infrequent recurrence(9). Although malignant behaviour is extremely rare, 5 years follow up of the operated patients should be recommended to assess the healing of this tumour (Philipsen, H. P., 2000).

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