CASE REPORT

RARE CALCIFYING EPITHELIAL ODONTOGENIC TUMOR IN A HIV POSITIVE INDIVIDUAL

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The calcifying epithelial odontogenic tumour (CEOT) is a rare but well recognized benign odontogenic tumour for any Pathologist. Histologically, it shows sheets of polyhedral epithelial cells with prominent intercellular bridges, hyperchromatic nuclei and globules of amyloid-like material among tumour cells which undergoes calcification. We present a case of CEOT with classical histopathological picture in a 37-year-old HIV positive patient with long evolution history. Its association with HIV positivity, long clinical history and delay in surgical treatment make it a challenging case to report.

Keywords: Calcifying Epithelial Odontogenic Tumour, HIV

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INTRODUCTION

Calcifying Epithelial Odontogenic Tumour (CEOT) or "Pindborg tumour"[1] is a painless slow growing locally aggressive tumour involving the premolar-molar area of the mandible, no gender predilection, and peak incidence in fourth decade of life.[2] Clinical characteristics like tumour occurrence age range, jaw sites and biological behaviour of CEOT are similar to ameloblastoma.[3] We present a classical CEOT in a HIV positive individual with a long clinical history and delayed treatment.

CASE REPORT

A 37-year-old male patient reported with a complaint of swelling in the right back region of face since last 3 years. During routine pre-operative blood investigations, he was found to be HIV positive. Patient refused to give any information regarding history of infection. On examination a hard firm swelling of right posterior mandible, size 5x4cm extending anteriorly from tooth 43 to angle of mandible, involving 48 was seen. Teeth 44, 45, 46 and 47 were missing (extracted due to mobility). (Figure-1) This slow growing swelling was present since last 18 years when he first underwent extraction of teeth 46, 47 along with incision and drainage. Three years back, the patient underwent his first incisional biopsy from right body of mandible. This was reported as secondarily infected odontogenic tumour.

His retrieved history corroborated that he was HIV positive at that time also. At this point no treatment was done as patient declined intervention and surgery. Six months back, at a different centre a second incisional biopsy was reported as ameloblastoma. (Table-1)

A detailed clinical history including occurrence of any opportunistic infections or increased bleeding tendencies were inquired for. He had no other obvious medical illness (cardiovascular disease, diabetes, hepatitis-induced liver disease) at the time of presentation or other signs of HIV-related disorders. Patient's present T lymphocyte count was 930 cells/cu.mm. After thorough pre-surgical medical and radiological evaluation [panoramic radiograph (Figure 2) and computed tomography (CT)], he underwent a right segmental mandibulectomy and reconstruction. The surgical specimen was submitted to the Department of Oral Pathology for histopathological diagnosis and confirmation. (Figure-1)

Haematoxylin eosin-stained sections showed epithelial (odontogenic) cell islands with prominent desmosomes interspersed with eosinophillic cell-free secretions most likely to be amyloid. Prominent Leisgang rings and dystrophic calcifications are also seen. Some of the epithelial islands show clear cell differentiation. (Figure 3) Post operatively there was slight delay in the healing of the surgical site and patient reported development of slight pus discharge from the area. Patient has been advised regular long-term follow-up and constant monitoring of blood counts for his HIV status.

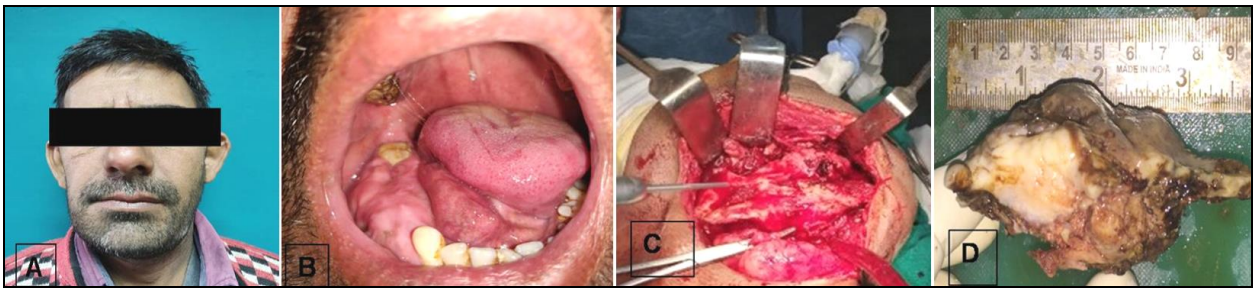

Figure-1: Clinical presentation of the case with an extra oral painless swelling of the right side of face [A] and intraoral photograph [B] of the swelling extending on the right body of mandible. Intra operative photograph of the right mandibular swelling [C] and gross picture of the right segmental mandibulectomy specimen submitted for histopathology [D].

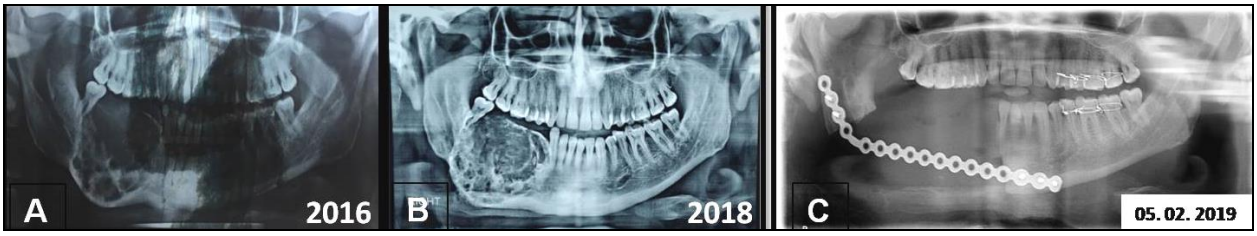

Figure-2: The panoramic radiographs taken in 2016 [A] and 2018 [B] of the pathology. The radiograph reveals a multilocular radiolucency with multiple radiopaque calcifications with expanded lower mandibular border, extending anteriorly from the tooth 43 and involving tooth 48 with opaque irregular borders. Figure 2[C] is a post operative panoramic radiograph with standard mandibular reconstruction plate following right segmental mandibular resection.

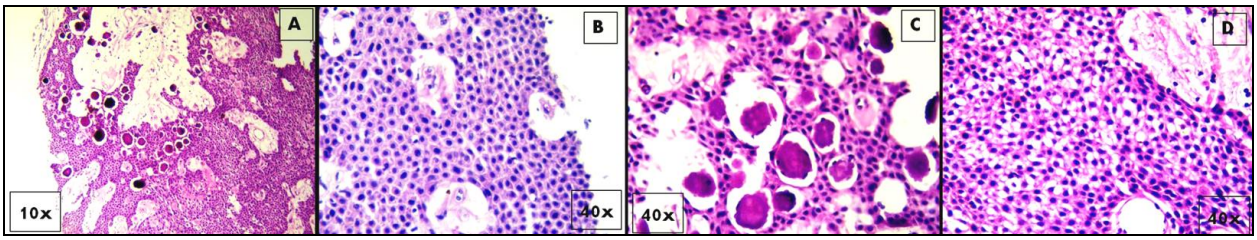

Figure-3: H & E stained sections showing epithelial (odontogenic) cell islands with prominent desmosomes interspersed with eosinophillic cell-free secretions most likely to be amyloid at magnifications of 10x [A] and 40x [B] Prominent Leisgang rings and dystrophic calcifications are also seen at 40x magnification [C]. Some of the epithelial islands show clear cell differentiation [D].

DISCUSSION

According to India HIV Estimation 2017 report, National adult (15–49 years) HIV prevalence in India is estimated at 0.22% (0.16–0.30%) which has continued its steady decline.[4] The incidence of oral mucosal lesions associated with HIV has decreased with the exception of human papillomavirus–associated oral lesions.[5] Using CDC classification, our patient is HIV positive with at least 3 years of positive infection history. Odontogenic tumour is an extraordinarily varied group of both benign and malignant tumours arising from similar sources of odontogenic epithelium. Odontogenic tumour as ameloblastoma and CEOT are generally locally destructive lesions with variable prognosis and thus are clinically challenging to manage. Different genetic and/or environmental etiological risk factors along with certain medically compromised conditions may play a role in prevalence and incidence of odontogenic tumour.[6]

Medical assessment as well of dental treatment of HIV-infected dental patients should not vary from that of any medically complex dental patient. Slight to no modification in the delivery of dental care is required in a medically-stable, asymptomatic HIV-infected patients. The HIV immunosuppression is least likely to affect the delivery of dental care than non–HIV-associated conditions. HIV associated stigma appears to be still prevalent in the age of reported successful HIV remissions as our patient was extremely circumspect or totally ignorant about revealing his past medical history, past infection and HIV status.[5]

Table-1: **Summary of patient's clinical history, radiographic and surgical history, histopathology reports and treatment in reference to the year of presentation. (*as told by the patient, # data retrieved from the archives of Dept. of Oral Pathology)**

Year of presentation	Clinical history	Radiographic findings	Surgical history	Treatment	Histopathology report	Immune status
2001*	Small swelling in the right posterior mandible.	NA	Extraction of teeth 46, 47.	Incision and drainage.*	NA	Not known
November 2016#	Slow growing swelling in the right posterior mandible.	OPG - Multilocular radiolucency with multiple radio opaque densities in the lower half involving anteriorly tooth 45 and posteriorly 48. Expanded lower border of body of mandible in that region. Figure 2[A] CT scan in axial view showed a single locular lesion of size 40x 36 x 42 mm with lingual and buccal cortical plate expansion. Teeth 48, 45 appear to be involved. Some radiopaque internal calcification seen.	FIRST Incisional biopsy	No treatment done (due to lack of patient follow)	Secondarily infected odontogenic tumor.# Second Oral Pathologist opinion (with same block) - inconclusive*	HIV positive
November 2018*	Osteophytes lesion in right mandibular body with expansion of both cortical plates.	None	SECOND Incisional biopsy	No treatment done	Ameloblastoma No evidence of fibro-osseous lesion.*	NA
December 2018- January 2019	Swelling in the right back region of face since last 3 years	OPG- Multilocular radiolucency with multiple radiopaque dentisties in the lower half involving anteriorly teeth 43 and posteriorly 48. Expanded lower border of body of mandible in that region. Figure 2 [B] CT Scan- 4.4x 4x4.3 cm expansile soft tissue density mass in body of mandible right side with internal calcification, destruction of inner & outer cortex and medulla of right side body of mandible bulging adjacent soft tissue of buccal cavity, floor of mouth.	Excision	Right segmental mandibular resection along with attached soft tissue.	Calcifying epithelial odontogenic tumour. (Figure 3)	HIV positive.

Till date only two odontogenic tumours have been reported occurring in HIV-positive patients. Ramanujapuram ML *et al.* reported unicystic ameloblastoma of both jaws in a 19-year-old female HIV-positive patient. They stated that HIV positive patients commonly develop Kaposi's sarcoma and non- Hodgkin's lymphoma as a result of immune-suppression.[7] Esteves CMD *et al.* reported a case of peripheral CEOT with clear cells occurring in a HIV positive patient as a conference abstract.[8]

Chrcanovic BR and Gomez RS reviewed 339 cases out of a total 362 reported CEOT lesions. The authors stressed that CEOT were not so often reported in the literature so their epidemiological study could help improve the diagnostic accuracy, refine treatment plan and optimize the clinical outcome.[9] But none of cases reviewed were associated with HIV positivity.

Histopathological confirmation is imperative in spite of recent advances in imaging-assisted surgical margin localization. Surgical management of odontogenic tumour is challenging due to their calcified nature, infiltrative nature and involvement of surrounding tissues thus, conservative approach risks the need for a second surgery and recurrence while aggressive treatment increases morbidity with extensive reconstructive surgery.[9] Both the scenarios can be remarkably devastating for medically compromised patients. Extensive and invasive CEOTs may need either marginal or segmental resection as in our case. Careful surgical considerations and decisions had been taken in our case owing its large size (greater than 4 cm)[6], late tumour diagnosis and medical condition. Philipsen HP and Reichart PA advocated that five years should be the absolute minimum follow-up necessary to assess the healing for CEOT.[2]

CONCLUSION

The dental management of the asymptomatic HIV-infected patient is the same as treatment of the non - HIV-positive patient.[6] Our case is only third case of odontogenic tumour in HIV positive patients. Whether a positive correlation exists between HIV positivity and odontogenic tumour occurrences is not known and needs to be explored. The connection between HIV/AIDS and certain tumour is not completely understood – but the relation probably relies on an impaired immune system. These reported cases at present appear to be a coincidence. The association with HIV positivity, associated stigmas, delay in treatment, an inconclusive incisional biopsy

report and definitive surgical treatment necessitate our case to be discussed. Further studies also need to be encouraged for exploration of co-relation between HIV and odontogenic tumour.

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