World Journal of Advanced Research and Reviews
eISSN: 2581-9615 CODEN (USA): WJARAI
Cross Ref DOI: 10.30574/wjarr
Journal homepage: https://wjarr.com/

(CASE REPORT)

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Dentinogenic ghost cell tumor: A case report with review of literature

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World Journal of Advanced Research and Reviews, 2022, 16(03), 193–197

Publication history: Received on 06 October 2022; revised on 17 November 2022; accepted on 20 November 2022

Article DOI: https://doi.org/10.30574/wjarr.2022.16.3.1237

Abstract

Dentinogenic ghost cell tumor is a rare odontogenic neoplasm first described by Praetorious et al in 1981. It is a solid variant of calcifying odontogenic cyst. This tumor shows no gender predilection and is seen more in mandibular anterior and molar region. Clinically, this tumor presents as two forms, Intraosseous & Extraosseous. Ghost cells are pathognomonic in diagnosing this tumor. Very few cases of this rare dentinogenic ghost cell tumor have been reported in the literature till date.

Keywords: Dentinogenic; Ghost cells; Tumors; Calcifying odontogenic cyst

1. Introduction

Dentinogenic ghost cell tumor (DGCT) is a rare, but locally invasive odontogenic tumor. Buchner et al in their study reported that only 2 to 14 % of all calcifying odontogenic cysts were solid in nature and were named DGCT [1]. DGCT comprises of only 11.5% of all calcifying odontogenic cysts indicating its rarity [2]. DGCT effects a wide age range and both the gender and jaws equally. Two forms of DGCT are seen depending on the site, intraosseous {central} and extraosseous {peripheral}. Combined radiolucent, radiopaque picture is common x ray finding for DGCT. Histologically, DGCT is characterized by ameloblastomatous odontogenic epithelium, ghost cells and dentinoid material.

2. Case Report

A 60 year old lady reported to the Department of Oral and Maxillofacial Surgery, College of Dental Science and Hospital, Rau, Indore, Madhya Pradesh, India, with a chief complaint of swelling in the lower right side of the jaw since 2 months (Figure 1). The swelling was associated with intermittent dull type of pain. The lady was moderately built and her medical history was non contributory. A tender firm swelling was seen extra orally extending from midline to right symphysis and body region. Intra oral examination showed vestibular obliteration from mandibular right lateral incisor till right first molar. Oral hygiene was poor with generalized attrition of teeth. None of the teeth in the area of the pathology were mobile.

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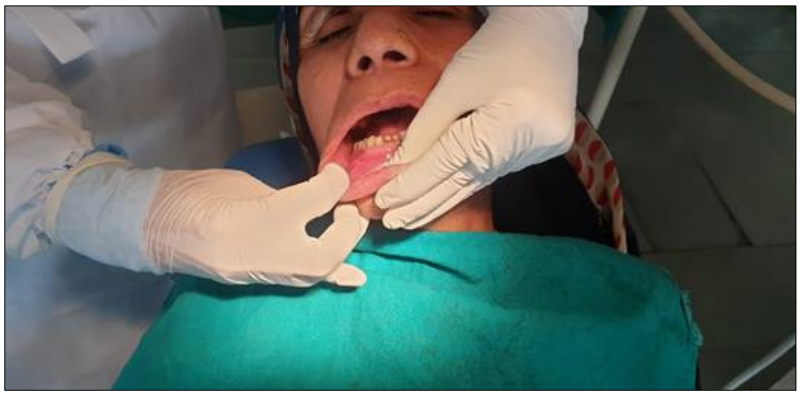

Figure 1 Pathology with Vestibular obliteration

Cone beam computed tomography (CBCT) (Figure 2A & 2B) showed well defined radiolucency with bit of radio opaque areas (Figure 3) from midline till mandibular right first molar and resorption of right first premolar root.

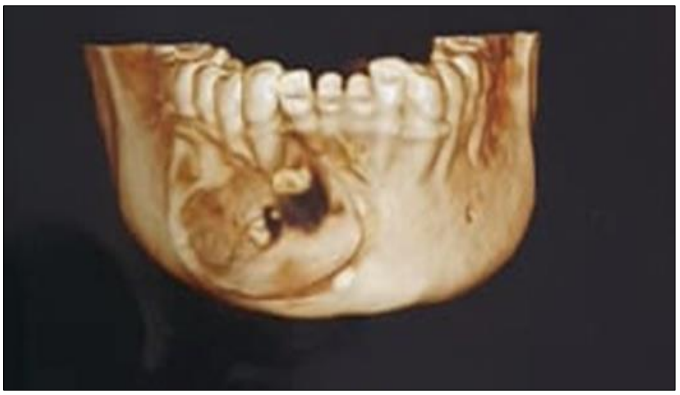

Figure 2A CBCT showing Lingual Cortex Involvement

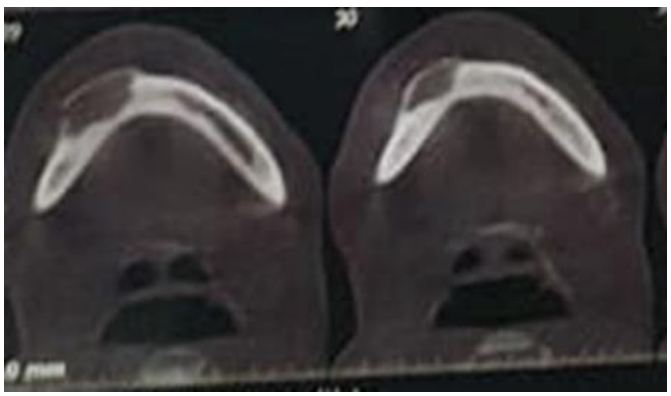

Figure 3 CBCT showing Bone Loss

The need for surgical excision was explained to the patient. A well informed written consent was obtained. Complete surgical excision with enucleation was done(Figure 4). The specimen was sent for histopathologic examination.

Figure 4 Intra-Operative picture showing Bony Defect

The microscopic examination showed presence of dentinoid material in abundance with keratinization of ghost cells and its presence in proliferative odontogenic epithelium(Figure 5).

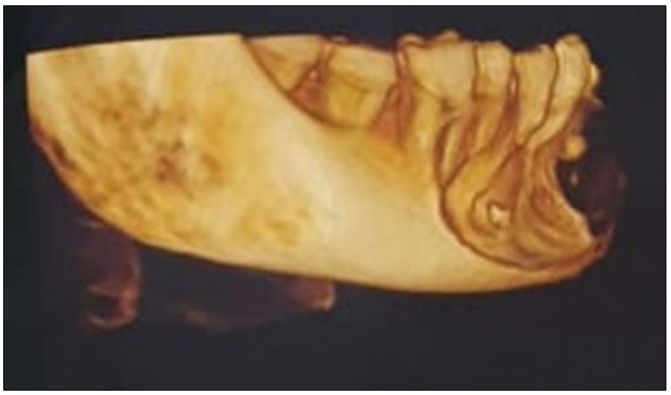

Figure 2B CBCT showing Extent of Bone Loss

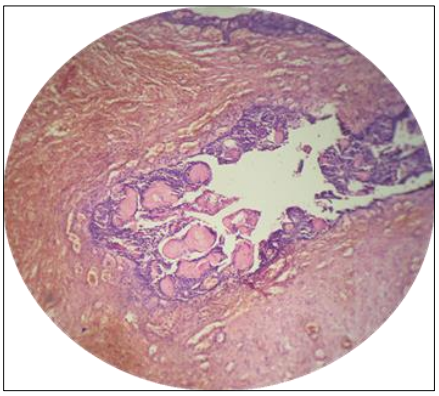

Figure 5 Microscopic Slide depicting Ghost Cells and Dentinoid material

3. Results and discussion

The term dentinogenic ghost cell tumor (DGCT) was first described by Gorlin et al in 1962 as a solid clinicopathologic variant of calcifying cystic odontogenic tumor [3]. Later in 1981, Praetorious et al suggested the term DGCT, because of presence of ghost cells & dentinoid material [4]. WHO in 2005, defined DGCT, as a locally invasive neoplasm characterized by ameloblastoma like islands of epithelial cells in the mature connective tissue stroma. Aberrant keratinization may be found in the form of ghost cells in association with varying amounts of dysplastic dentin [5]. Etiology of DGCT is still unknown but it has been suggested that missense mutation in B catenin in wingless integrated pathway plays a crucial role in the development of DGCT [6].

Other names for DGCT include calcifying ghost cell odontogenic tumor, Odontogenic ghost cell tumor & dentinoameloblastoma [7]. Buchner et al in their study of 215 cases, stated that only 1 to 2 % of all odontogenic tumors were calcifying odontogenic cysts out of which only 2 to 14% were solid tumors and were named DGCT. A wide age range of population can be effected by DGCT, which can vary from 12 to 70 years. Few studies reported that DGCT exhibits male predilection [8] but equal gender predilection has also been reported [9]. De Arruda et al in their study claim that 65% of DGCT are seen in mandibular incisor to canine region [10]. While few other studies show both the jaws being equally affected by DGCT [11]. Clinically DGCT might show symptoms as pain, swelling, teeth mobility, while other few lesions might remain entirely asymptomatic & get diagnosed only on routine dental examination.

DGCT has two variants. One is the central (intraosseous) and other is peripheral (extraosseous). The intra osseous or the central variant comprises of 83% of all the DGCT, while the peripheral type forms only 17% of all [12]. Among these the central or the intra osseous is said to be more aggressive in nature & are more commonly seen in mandibular canine to molar region [13]. Buchner in a study of single case report of DGCT, into comprehensive analysis of clinical & radiographic features, reported that he could find only 45 well documented cases of central DGCT in the literature from 1972-2014[14]. The peripheral variant might mimic other soft tissue lesions as epulis, peripheral giant cell granuloma arising in the gingival soft tissues. DGCT on radiographs show, well or ill-defined radiolucency with scattered radiopaque calcifications, with unilocular or multilocular picture [15]. In some cases, DGCT might be found associated with root resorption, odontomes, impacted teeth etc. Histologically DGCT is composed of ameloblastoma like area & odontogenic epithelial islands. Varying amount of keratinizing ghost cells are a peculiar picture & pathognomonic for diagnosis of DGCT [16]. Ghost cells are swollen, ellipsoid keratinized epithelial cells characterized by loss of nuclei with preservation of basic cell outline, induction of foreign body granuloma and potential to calcify [17]. Masses of dentinoid material are also found abundant with ghost cells, which actually represents inflammatory response of body tissues towards masses of ghost cells. Ghost cells are most important feature in diagnosing DGCT. But ghost cells are prominent feature seen in few other lesions also. These include, ameloblastomas, ameloblastic fibroodontoma, craniopharyngioma, odontomas & others. So these lesions need to be differentiated before any surgical intervention of DGCT.

Enucleation with complete surgical excision is the treatment of choice for management of DGCT. Among both the clinical forms, the intra osseous or the central variant requires a more radical approach may be with segmental resection as these are more aggressive in nature & tend to recur. Soluk Tekkesin et al even suggested a long term follow up of mainly the central variant [18]. Few studies have reported the recurrence rate of 33 to 73% within one to twenty year span [19]. One case of very early recurrence of DGCT has also been reported which required extensive hemi maxillectomy & reconstruction [20]

4. Conclusion

Dentinogenic ghost cell tumor is rare among all the other odontogenic tumors. Successful management of intraosseous dentinogenic ghost cell tumor requires a carefully planned radical approach.

Compliance with ethical standards

Acknowledgments

Acknowledgments The authors thank the reviewers for their insightful suggestions.

Disclosure of conflict of interest

No conflict of interest.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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