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Case Report

Aggressive Recurrent Odontogenic Ghost Tumor with Cranial Invasion.

A. Hossein Zadeh,* P. Deihimy**

Abstract

Calcifying odontogenic cyst (COC) is a unique odontogenic lesion with combined features of a cyst and neoplasm. This lesion shows a great diversity in its clinicopathological behavior. It may become aggressive and rarely malignant. This case is an aggressive COC of the maxilla in a young male patient which recurred after several radical surgical excisions. Eventually invading the cranium, the lesion neither responded to surgical excision nor to radiotherapy.
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Keywords • Calcifying odontogenic • cyst Odontogenic ghost cell tumor • Cranial invasion.

Introduction

Calcifying odontogenic cyst (COC) is a well-established pathological entity. Harboring characteristics of both a cyst and a tumor, it was first described by Gorlin et al, in 1962.[1-17] Praetorius and his colleagues, classified COC [8] as type I (1. Unicystic (simple); 2. Odontoma producing; and 3. Ameloblastomatous proliferating), and Type II Neoplastic (Dentinogenic ghost cell tumor (DGCT), or epithelial odontogenic ghost cell tumor (EOGCT)). Praetorius classification has a dualistic concept. However WHO in 1992 classified COC as an odontogenic tumor and designated it as odontogenic ghost cell tumor (OGCT).[6-7] This is a monistic concept, suggesting that all COCs are neoplastic in nature although they might be cystic in architecture. Although few cases of aggressive or malignant form of this lesion have been reported before, this is the first case report of aggressive recurrent OGCT with cranial invasion in a young male patient.

Case report

In May 1999, a 21-year-old man was referred for a small swelling in the area of the right upper lateral and canine teeth. It was painless with intact mucosa and radiolucent with well defined borders between the above-mentioned teeth which resembled a cystic lesion on orthopantogram. The teeth were vital with pulp tester. There were no root resorption or discoloration and no luxation was identified. The patient had no history of systemic disease. The patient underwent an excisional biopsy three days later and returned a month later.(Fig 1). At this time the lesion

*Department of Oral and Maxillofacial, Surgery, **Department of Oral and Maxillofacial Pathology, Isfahan University of Medical Sciences, Hezar Jarib Ave, Isfahan, Iran.

Correspondence: A. Hossein Zadeh, MD, Isfahan University of Medical Sciences, Hezar Jarib Ave, Isfahan, Iran.

Tel: +98-311-6623339
E-mail: *a_hosseinzadeh@hotmail.com*

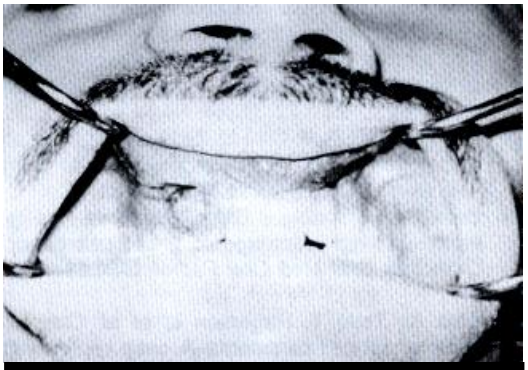

Fig 1: Small exfoliative lesion of maxilla buccal the upper lateral and canine teeth. Tumor recurrence after first operation.

had increased in size and ulceration with exfoliation was visible on the mucosa. Upper molar and canine teeth on right side had luxated. In the time period from June 1999 to Dec 2001 the patient underwent four operations starting with total excision to extensive surgeries including hemi-maxillectomy. Finally the lesion invaded cranium, (Fig 2) which responded neither to craniotomy nor to radiotherapy. Histopathologic examination revealed squamous and ovoid proliferative epithelial cells with a large number of ghost cells, palisading of marginal cells in epithelial islands and stellate-like tissues. Slight cellular pleomorphism with nuclear hyperchromatism were identified, however mitotic activity was rarely observed. So the tumor was diagnosed as COC or odontologic ghost cell tumor. (Fig 3) The patient died of the recurrency of his cranial tumor.

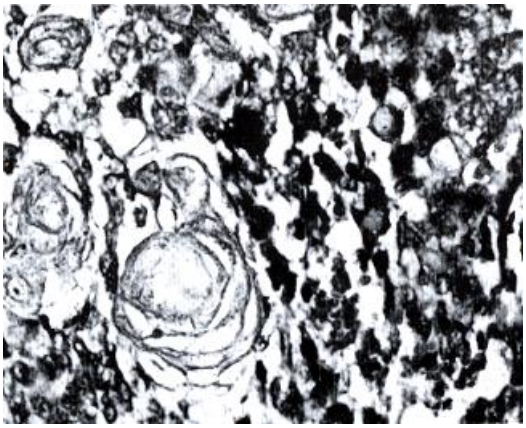

Fig 3: Slight cellular atypism in epithelial cells with ghost cells (H&E).

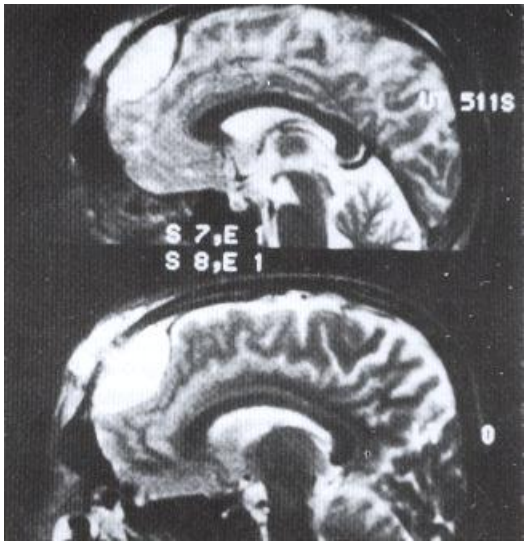

Fig 2: MRI (lateral view) invasion of tumor to the anterior part of brain.

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