

Intraosseous verrucous carcinoma originating from an odontogenic cyst: a case report

Nooshin Mohtasham¹⁾, Faramarz Babazadeh²⁾ and Hamid Jafarzadeh³⁾

¹⁾Department of Oral and Maxillofacial Pathology, Faculty of Dentistry and Dental Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

²⁾Department of Oral and Maxillofacial Surgery, Faculty of Dentistry and Dental Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

³⁾Department of Endodontics, Faculty of Dentistry and Dental Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

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Abstract: Carcinoma originating in bone is uncommon; most of them apparently arise in odontogenic cysts. In this paper, we report an extremely rare case in which, verrucous carcinoma originated from an odontogenic cyst. This lesion was firm and non-tender on palpation and had a white-pink appearance. It was encased in the anterior aspect of the maxilla and exhibited microscopic features of verrucous carcinoma of the oral mucosa. After surgical enucleation, no recurrence or metastasis has been observed up to now. It is mandatory to correlate the clinical and histopathologic findings to establish a true diagnosis. (J. Oral Sci. 50, 91-94, 2008)

Keywords: maxillary bone; odontogenic cyst; verrucous carcinoma.

Introduction

Carcinoma arising in bone is an extremely rare condition that is essentially limited to the jaws; most of them arise in odontogenic cysts. Residual periapical cyst, dentigerous cyst, and odontogenic keratocyst are the most common types associated with carcinomatous transformation. Most carcinomas arising in cysts are well-differentiated squamous

cell carcinomas (1). However, only a few cases have been described in the literature (2). Scheer et al. (3) reported a primary intraosseous carcinoma arising from an odontogenic cyst in a 64-year-old woman. Aboul-hosn Centenero et al. (2) presented three cases of carcinoma that had originated from a previous cystic lesion. Chaisuparat et al. (4) reported 6 cases affecting 4 female and 2 male patients with a mean age of 56.2 years, whereas Gonzalez-Garcia et al. (5) reported 3 cases of primary intraosseous carcinoma of the jaws that had developed within an odontogenic cyst, ameloblastoma, and de novo origin.

Verrucous carcinoma, which was first reported by Ackerman in 1948 as a spit tobacco-associated malignancy (1,6,7), represents 1% to 10% of all oral squamous cell carcinomas. It is observed predominantly in men older than 55 years of age and most commonly in the mandibular vestibule, buccal mucosa, and hard palate (1,6). In fact, it is a rare variant of squamous cell carcinoma with a characteristic morphology and specific behavior. It presents a potential for local recurrence that should be considered when planning for surgical removal of this neoplasm in oral cavity (8). Brush biopsies of such neoplastic oral lesions showing DNA-aneuploidy with tetraploid stemlines should be used for diagnosis and follow-up examination of these patients (9).

Primary intraosseous verrucous carcinoma arising from an odontogenic cyst is an extremely rare lesion (7,10). To the best of our knowledge, only two cases of this phenomenon have been reported up to now. In the present paper, we report a case of intraosseous verrucous carcinoma which originated from an odontogenic cyst.

Correspondence to Dr. Hamid Jafarzadeh, Faculty of Dentistry and Dental Research Center, Vakilabad Blvd, Mashhad, P.O. Box: 91735-984, Iran

Tel: +98-511-8829501

Fax: +98-511-7626058

E-mail: hamid_j365@yahoo.com & JafarzadehBH@mums.ac.ir

Case Report and Results

A 58-year-old male patient was admitted to the Faculty of Dentistry, Mashhad University of Medical Sciences in December 2004 for evaluation of an exophytic polypoid lesion on the labial and palatal aspects of the right maxillary alveolus. (Fig. 1) The lesion was firm and non-tender on palpation and had a white-pink appearance. The right anterior teeth (central incisor to the first premolar) were extracted two months earlier. The painless mass was evident in the right anterior maxilla for the past four years, according to his dental history. The patient reported that this exophytic lesion had appeared in the extraction site only few weeks after the extraction. The patient's medical history was non-contributory.

Radiographic investigation revealed a well-defined radiolucency measuring 35 × 25 mm in the anterior aspect

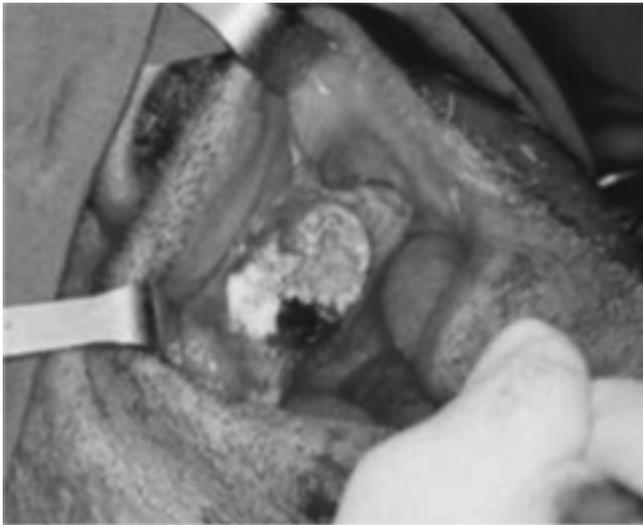


Fig. 1 Clinical view of the exophytic lesion.

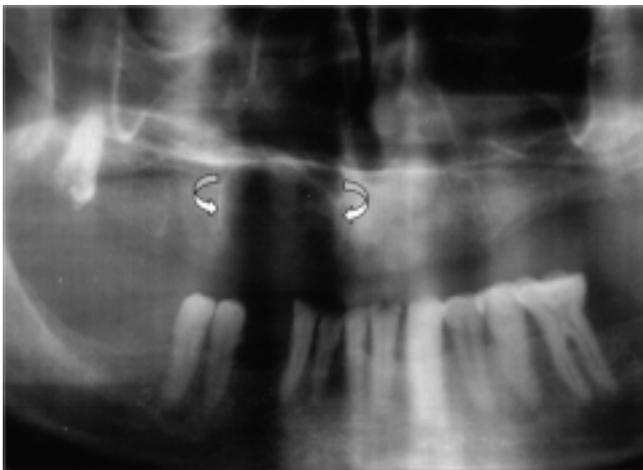


Fig. 2 Dental panoramic radiograph indicating a unilocular radiolucency in the right maxilla.

of the maxilla. (Fig. 2) During the operation, a cystic lesion with verrucous proliferations and cheesy material was found within the anterior maxilla. Incisional biopsy was performed and the tissue was submitted for histopathologic examination but the biopsy was not enough for a true diagnosis. It was evidently a pseudo-epitheliomatous hyperplasia. Therefore, in June 2005, an excisional biopsy via enucleation surgery was performed and resubmitted for histopathologic examination.

Pathologic findings

Gross:

A soft tissue lesion of size 4 × 4 × 3 cm attached to the alveolar bone of maxilla and bone loss in the lateral and medial walls of the bone were observed.

Microscopic:

Finger-like projections of well-differentiated squamous epithelium with papillomatosis and marked hyperkeratosis in the lining of an odontogenic keratocystic tumor were observed. (Figs. 3 and 4) Acanthosis consisting of downward growth of numerous thickened, bulbous rete ridges of squamous cells with mild atypia and parakeratin plugging between the surface projections were also present. (Figs. 5 and 6) The lumen contained eosinophilic stained keratin and chronic inflammation was noted in the corium of the lesion.

Taking together all the features, a diagnosis of verrucous carcinoma with odontogenic cyst was established.

Discussion

Verrucous carcinoma is a low-grade variant of squamous cell carcinoma with unique clinical characteristics and histopathologic features (1). Poor oral hygiene and tobacco abuse are the common etiologic factors for verrucous carcinoma in the oral mucosa (1,11-13). Numerous investigators have reported that verrucous carcinoma arises in areas other than oral mucosa including larynx, esophagus, glans penis, and vulva (10).

Primary intraosseous verrucous carcinoma arising from an odontogenic cyst is extremely rare (7). Enrique et al. (10) in 1980 reported a case with the history of tobacco and alcohol abuse in which a large, firm, nontender mass in the parotid area, with several draining fistulous tracts was observed. In contrast, there was no history of tobacco or alcohol abuse in the present case. Pomatto et al. (7) reported a case of verrucous carcinoma in the lining of a maxillary odontogenic cyst. Although Aldred et al. (13) reported an odontogenic cyst with verrucous proliferation in the maxillary alveolus, it did not represent any carcinomatous transformation. Consequently, the present case would be the third of such cases.

The gross and microscopic findings as well as the benign histopathologic features of verrucous carcinoma can result in misdiagnosis (10). In the present case, the initial biopsy did not provide an exact diagnosis. Therefore, an excisional biopsy was performed. The patient had an uneventful post-surgical course. Moreover, after more than twenty months, no recurrence or metastasis was observed.

It should be remembered that although verrucous carcinoma arising in an odontogenic cyst is very rare, it can be expected. In fact, in most cases, it would be found in the soft tissues. It has been proposed that because of

the excellent prognosis of verrucous carcinoma following surgical resection, it is important to identify this lesion properly. Metastasis is extremely rare in verrucous carcinoma, thus the treatment of choice is surgical excision without radical neck dissection. A history of a localized, painless, slow-growing mass appearing clinically as a friable exophytic papillary lesion would suggest verrucous carcinoma, so it is mandatory to correlate the clinical and histopathologic findings when establishing a diagnosis (10).

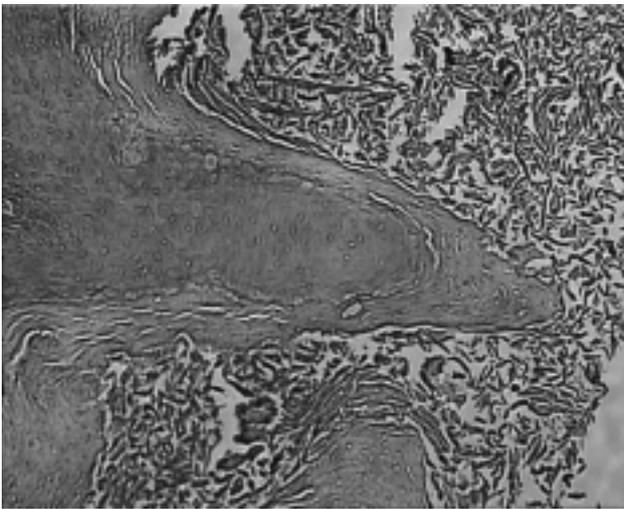


Fig. 3 Papillomatosis and hyperkeratosis of well-differentiated squamous epithelium with keratin plugging ($\times 40$ magnification-Hematoxylin & Eosin staining).

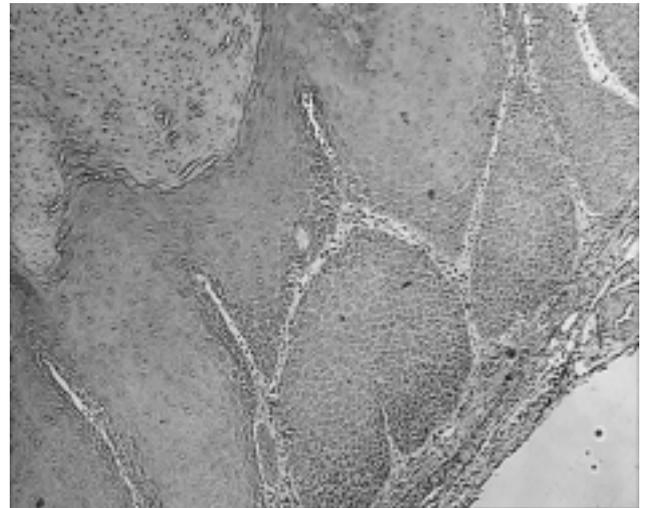


Fig. 5 Downward growth of numerous thickened, bulbous rete ridges of squamous cells with mild atypia ($\times 40$ magnification-Hematoxylin & Eosin staining).

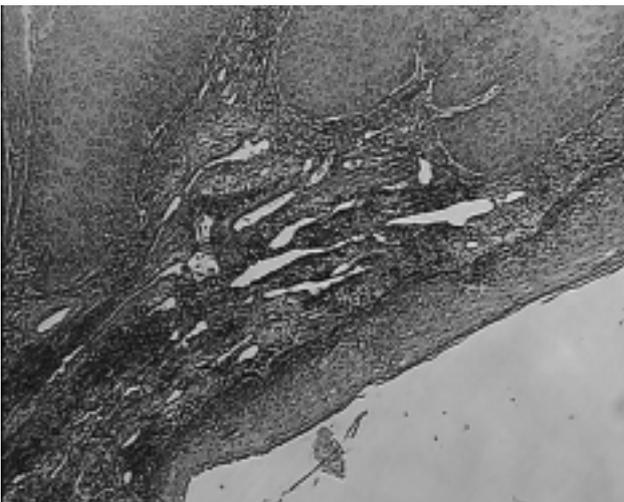


Fig. 4 Finger-like projections of squamous epithelium and hyperkeratosis in the lining of an odontogenic keratocystic tumor ($\times 40$ magnification-Hematoxylin & Eosin staining).

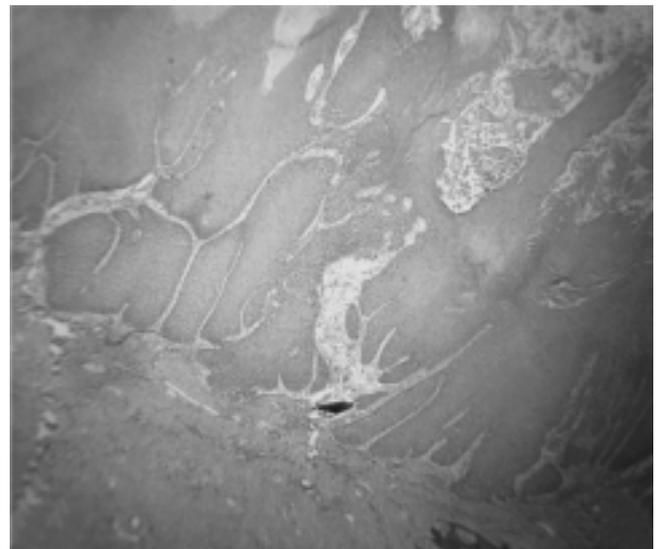


Fig. 6 Low power photomicrograph of the morphological view of the lesion.

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