Case Report ISSN 2639-9490

Oral Health & Dental Science

Primary Intraosseous Squamous Cell Carcinoma Associated with an Odontogenic Cyst: Case Report and Review of the Literature

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Received: 30 June 2025; **Accepted:** 26 Jul 2025; **Published:** 05 Aug 2025

Citation: Meza-García Gerardo, Sibaja-Ponce Tania Atenea, Cruz-Nuñez Mariana. Primary Intraosseous Squamous Cell Carcinoma Associated with an Odontogenic Cyst: Case Report and Review of the Literature. Oral Health Dental Sci. 2025; 9(4); 1-4.

ABSTRACT

Primary intraosseous squamous cell carcinoma (PISCC) associated with an odontogenic cyst (OCC) is a malignant odontogenic tumor that occurs infrequently and exclusively in the maxillary bones. It affects middle-aged people, mainly men, and is usually located in the posterior area of the mandible. Clinically it can present the classic characteristics of a benign odontogenic tumor, although it can also be associated with painful symptomatology and alterations in sensitivity. A case is presented of 65-year-old man diagnosed with CCEIP associated with an OCC. The clinical, radiological and histological characteristics are described, taking into account the importance of taking a biopsy of a cystic lesion characterized by affecting areas of the maxillary bones.

Keywords

Odontogenic carcinoma, Primary intraosseous spinocellular carcinoma, Odontogenic cyst.

Introduction

Primary intraosseous squamous cell carcinoma (PISCC) is defined as a squamous cell carcinoma (SCC) that arises primarily within the jaws and has no connection to the oral mucosa; it is thought to originate in the odontogenic epithelium and is rare in occurrence [1]. Clinical symptoms are not readily apparent, which means that the diagnosis may be difficult to detect in the early stages, and the prognosis is generally not considered optimistic. Clinical symptoms include: pain in the affected area, which tends to be confused with an odontogenic infectious process, so that by the time an accurate diagnosis is made, in many cases, the tumor has already infiltrated [1]. These carcinomas can be aggressive and affect large areas of the jaws. The biopsy is what confirms the diagnosis. The following is a clinical case and its follow-up [2].

Case Report

A 65-year-old male patient presents for dental consultation due to slight pain (VAS 4-5/10) in the left lower retromolar area, with data of paresthesia in the area. His personal pathological history included smoking and occasional alcoholism. On clinical examination there was no increase in volume in the painful area. On intraoral examination there was no pain on palpation, edema or redness in the area. The radiographic study showed a lower left third molar with a unilocular radiolucent area with a poorly defined border without cortices (Figure 1).

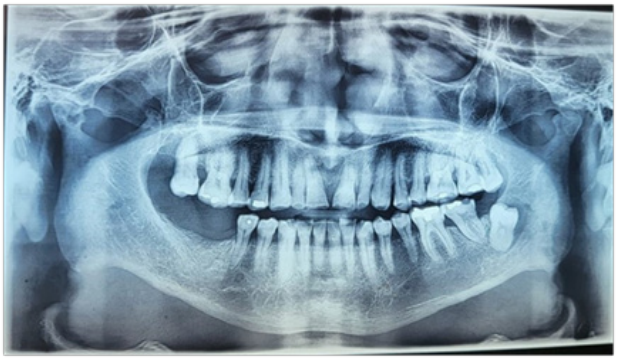


Figure 1: Lower left third molar with a unilocular radiolucent area with a poorly defined border.

Therefore, third molar surgery was performed where changes in

the anatomy of the bone were observed, as well as great bone loss and outflow of whitish-yellowish fat-like material (Figure 2).

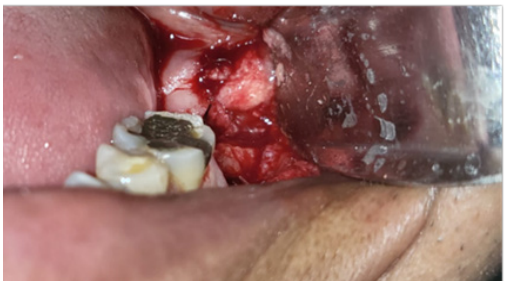


Figure 2: Third molar surgery where bone loss of the external cortical bone was observed.

It was decided to take an excisional biopsy and extraction of the involved dental organ (Figure 3); On histopathological study, a band of stratified parakeratinized squamous epithelium was found, continuous with masses of polygonal neoplastic cells with cellular and nuclear pleomorphism, hyperchromatism, dyskeratosis, individual and group keratinization, increase in the number of mitoses and aberrant aspect, infiltrating an underlying connective tissue capsule (Figure 4 and 5) which is well vascularized with a mixed type inflammatory infiltrate composed of lymphocytes and neutrophils, a large amount of desquamated keratin (Figure 6), irregular fragments of calcified tissue, A diagnosis of well-differentiated squamous cell carcinoma was made. Because it is thought to be primary to the dental follicle, it is considered a primary intraosseous squamous cell carcinoma associated with the lining of the odontogenic cyst.

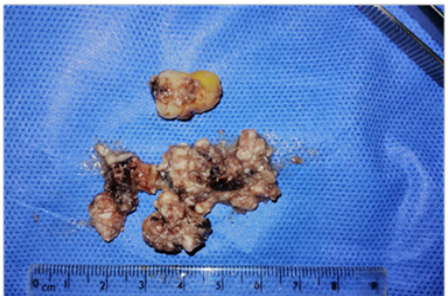


Figure 3: Surgical Piece.

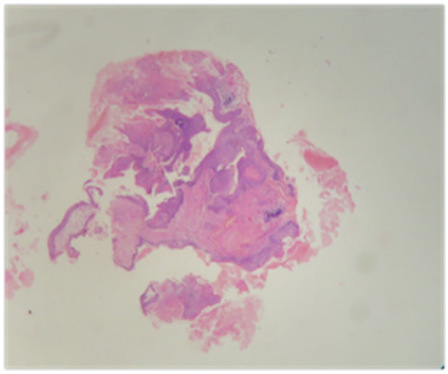


Figure 4: Stratified parakeratinized squamous epithelium continuous with masses of polygonal neoplastic cells with cellular and nuclear pleomorphism.

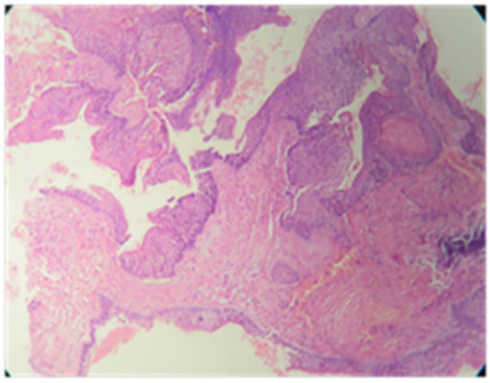


Figure 5: Infiltration to the connective tissue and keratin pearls can be seen.

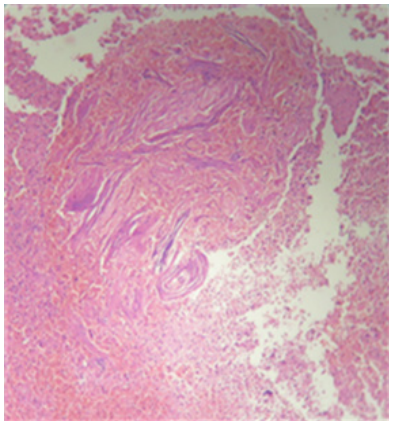


Figure 6: Desquamated keratin.

The patient was referred to oncology, presented with left adenopathy of less than 1 cm, treatment was started with induction chemotherapy with docetaxel, platinum and 5-fluorouracil for 4 cycles followed by chemotherapy. Subsequently he received radiotherapy. After 3 years of follow-up the patient does not show any recurrence of the disease, but as a consequence of the radiotherapy he presents a trismus that only allows an opening of the oral cavity of 2.5 cm. The surgical wound has not healed, a new biopsy was taken and reported as necrosis and bacterial infection, negative for malignancy (Figure 7).

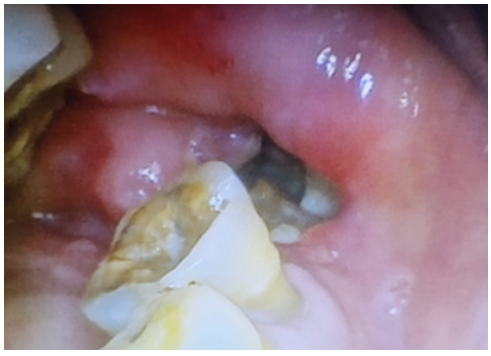
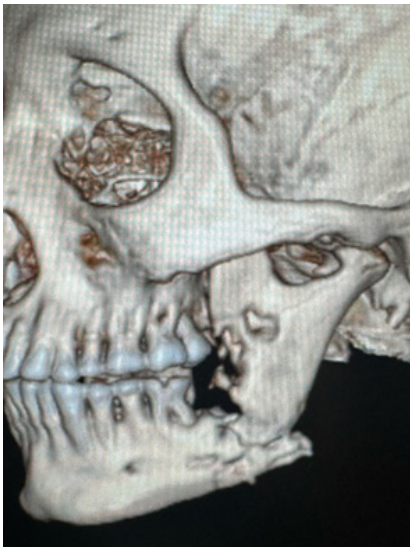


Figure 7: Follow-up photo 3 years after diagnosis, note the difficulty of opening and the unhealed wound.

Upon reviewing the CT scan, it showed a poorly defined lesion with a pathological fracture (Figure 8) in the angle of the mandible, so new surgical treatment is required.



Discussion

PIOSCC arises within the maxillary bones; without any initial connection to the oral mucosa or sinus mucosa and develops from remnants of the odontogenic epithelium. According to the latest WHO classification of tumors, there are three PIOSCC subtypes:

1. Solid tumor invading the medullary spaces and inducing bone resorption; 2. solid tumor invading the medullary spaces and inducing bone resorption.
2. PIOSCC arising from the lining of an odontogenic cyst, making a subdivision into carcinomas arising from the keratocyst and carcinomas arising from other odontogenic cysts.
3. PIOSCC in association with other benign epithelial odontogenic odontogenic tumors.

Malignant transformation of an odontogenic cyst into a PIOSCC is extremely rare; in an article describing about 386 cases in the literature, most of them arise from root/residual cysts (60%), although cases originating from dentigerous cysts (16%), keratocystic odontogenic tumor (14%) and lateral periodontal cysts (1%) [3] have also been reported; in the present case we suggest that it originated from a dentigerous cyst.

Malignization of dentigerous cysts to PIOSCC occurs mainly from the third decade of life onwards; the male gender predilection is 2:1, reflecting the higher pre-existing incidence of odontogenic cysts in men [4]; as was the case in our patient.

PIOSCC can be painless in most cases, however, in case of clinical manifestations, swelling is the most frequent sign (43.4%), followed by pain (13.3%) and paresthesia (10.0%); the mandible is involved in most cases (86.7%) [5]. The symptoms in our case were similar.

The expression of cytokeratins (CK) in the developing tooth germ may be useful in understanding the histogenesis of odontogenic cysts or benign and malignant tumors. CK 5, 7, 8, 14 and 19 are expressed in the enamel organ and CK 14 is the major intermediate filament found in the dental lamina, reduced enamel epithelium and enamel organ; however, this CK is also commonly expressed in other epithelia, including the oral mucosa. These findings may enhance the hypothesis of malignant transformation of the dentigerous cyst [6].

In a study between 1997 and 2023, 10 cases of PIOC were identified where immunohistochemical staining with CK 14, CK 19, p40 and p53 was performed. All 10 cases were associated with a cyst, showed strong diffuse cytoplasmic expression for CK14 and nuclear p40 expression in the cyst and tumor, CK19 was expressed in 7 of 9 cases, frequently confined to the dysplastic cyst lining and less differentiated tumor cells. 9 of 10 cases showed nuclear p53 expression; 2 cases diagnosed as arising in keratinizing cysts did not express CK19 [7].

The pathogenesis is not very clear but it is thought to be due to chronic inflammatory processes of the previous cyst infection [8]. Radiographically, PIOSCC may present as unilocular or multilocular lesions, with ill-defined or well-defined borders, but not corticated [9].

In terms of treatment, surgical excision is the first option and verify that the tumor is completely removed along with the causative tooth for a wide margin of safety [9]. For this case the use of chemotherapy was also recommended. Although cases have been described in which when the lesions are small, it is possible to be more conservative [10].

Cervical lymph node metastasis is seen in up to 50% of all cases of PIOSCC, and may also spread along the inferior dental nerve. Radiotherapy may be indicated depending on the extent of the tumor and regional lymph node involvement. In our case the patient presented with adenopathies so chemotherapy was recommended. The overall survival rate of patients with PIOSCC was 64.7% and 28.6% at 2 and 5 years, respectively, in our case it has been 2 years of survival without recurrence.

Conclusions

We presented a case of intraosseous carcinoma associated with a dentigerous cyst, it is a rare lesion arising from direct transformation of odontogenic epithelial debris in the mandible, including epithelial debris found within the alveolar bone of the enamel epithelium surrounding an impacted tooth.

All dental cysts and follicles should be evaluated histologically to determine if any may become malignant.

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