Intraosseous Mucoepidermoid Carcinoma Radiographically Mimicking a Cystic Lesion – A Case Report

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Abstract

A 42-year-old man presented with pain and swelling in the left lower back tooth region. He underwent uneventful extraction of impacted tooth 1 year before. There was a well-defined dome shaped swelling of size 1.5 cm × 2.5 cm in the left side posterior alveolar ridge. Lesional aspiration revealed a thick mucus aspirate. Computed tomography revealed significant buccal and lingual cortex destruction with an isodense mass similar in density to adjacent soft tissues was noticed. CECT revealed an ill-defined heterogeneously enhancing soft tissue lesion measuring 1.8 cm × 2 cm, in the left retromolar trigone causing expansion and cortical thinning of the left mandibular ramus. Incisional biopsy of the lesion confirmed the diagnosis of the lesion as MEC of mandible. To date, only one case series and two case reports have focused strictly on the diagnostic imaging characteristics of IMC. This case report focuses on the diagnostic imaging characteristics of IMC.

Keywords: Salivary gland neoplasm; Mucoepidermoid carcinoma; Intraosseous; Diagnostic imaging

Introduction

Mucoepidermoid carcinoma is a malignant epithelial tumor, first studied and described as a separate entity by Stewart, Foote and Becker in 1945. Mucoepidermoid carcinoma represents 20% to 34% of malignant tumors originating in both major and minor salivary glands. This carcinoma of salivary glands accounts for 5% of all salivary gland tumors [1]. Eversole reviewed 815 cases and found that of the major salivary gland tumors, 89.6% involved the parotid, 8.4% submandibular and 0.4% sublingual gland. In 1991, after a systematic review of its histology and degree of differentiation the WHO classification recommended that the term “mucoepidermoid tumor” be changed to “mucoepidermoid carcinoma” [2].

Case Report

A 42-year-old male reported with complaints of pain and swelling in the left lower back tooth region for 5 months, 5 months back patient noted a small swelling in the left lower back tooth region which has gradually increased to the present size. He also complained of pain over the swelling for past 4 months, which was continuous, dull, relieved on medication. There was no history of trauma or numbness and he underwent uneventful extraction of impacted tooth 1 year before. In medical history, patient was known case of hypothyroid and bronchial asthma for past 4 years.

Extra oral examination showed no gross facial asymmetry and there was no evidence of swelling (Figure 1). On palpation of lymph nodes, left side single palpable submandibular lymph node of size 1.5 cm × 1.5 cm in diameter, soft to firm, mobile and non-tender. Intraoral examination showed a well-defined dome shaped swelling of size 1.5 cm × 2.5 cm present in relation to left side posterior alveolar ridge extending from distal of 37 to retromolar region (Figure 2). Buccally, lingually from buccal vestibule to 1 cm short of floor of the mouth. The surface covering the swelling showed no secondary changes like sinus opening or pus discharge and no visible pulsations. On palpation, inspexitory findings were confirmed. The selling was soft to firm in consistency, compressible, non-fluctuant and tender. Hard tissue examination revealed missing 38, no tooth discoloration or hypoplasia seen.

Based on above history and clinical findings, provisional diagnosis

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was given as residual odontogenic cyst most probably dentigerous cyst in relation to extracted 38. The differential diagnosis was radicular cyst, paradental cyst, odontogenic keratocyst, calcifying epithelial odontogenic cyst, simple bone cyst, pleomorphic adenoma and its variants. Ameloblastoma, ameloblastic fibroma, odontogenic myxoma, salivary gland tumors like mucoepidermoid carcinoma, adenoid cystic carcinoma, intraosseous squamous cell carcinoma, metastatic tumors to jaws from lungs, kidney, prostate.

Vitality test revealed immediate response in relation to 36, 37. Lesional aspiration was attempted using 23-gauge needle and 2 drops of thick mucus aspirate was obtained. Routine haemogram showed normal range of all the blood cells. Intraoral periapical radiograph in relation to 36, 37, edentulous 38 revealed a well-defined, corticated, homogenous radiolucency extending from distal aspect of 37 and the posterior extent was not covered. Orthopantogram revealed a well-defined, corticated homogenous radiolucency extending from distal aspect of 37 to level of ascending ramus. The inferior alveolar nerve canal was traceable and not displaced (Figure 3). Lateral occlusal radiograph revealed no evidence of cortical expansion.

Computed tomogram axial view bone window revealed significant buccal and lingual cortex destruction. An isodense mass similar in density to adjacent soft tissues was noticed. No evidence of impacted tooth. Computed tomogram coronal section bone window (Figure 4) revealed buccal and lingual cortical destruction extending from left angle to ramus of the mandible. Computed tomogram 3D reconstruction (Figure 5) revealed through and through buccal and lingual cortical expansion. Inferior border of mandible is intact without any destruction/expansion. CECT (Figure 6) revealed an ill-defined heterogeneously enhancing soft tissue lesion measuring 1.8 cm × 2 cm is seen in the left retromolar trigone causing expansion and cortical thinning of the left mandibular ramus surrounding the apices of 2nd and 3rd molars. Few discrete enhancing level 1a bilateral level 1b and 2 cervical nodes noted.

Incisional biopsy was done and the section (Figure 7) basically consist of mucous and epidermoid type of cells. Low cuboidal type of cells representing intermediate kind and few mitotic changes were also noticed in solid area. Medially there is focal loss of fat plane with the medial pterygoid muscle and extension to masticator space.

Incisional biopsy was done and the section (Figure 7) basically consist of mucous and epidermoid type of cells. Low cuboidal type of cells representing intermediate kind of cells but scanty in nature. But some areas stromal components and clear cell changes too. Few mitotic changes noticed in solid area. There is not much of inflammatory changes noticed suggestive of intermediate grade mucoepidermoid carcinoma. The patient was referred to a regional cancer center for further treatment where radical excision of the lesion with adjuvant radiotherapy was carried out.
Discussion

Mucoepidermoid carcinomas arising within the jaws as primary central bony lesions are termed central mucoepidermoid carcinomas and are extremely rare, comprising 2% to 3% of all mucoepidermoid carcinomas. The origin of these lesions in the jaws is thought to be due to neoplastic transformation of the sinus epithelium; entrapped retromolar mucous glands and developmental embryonic remnants of the submandibular gland within the mandible or neoplastic transformation of the mucous-secreting cells common to the pluripotential epithelial lining of dentigerous cysts associated with impacted third molars [3]. The origin of the lesion in our case could be from entrapped retromolar mucous glands since the lesion was more involving the retromolar region. In 1974, Alexander et al. introduced the following criteria for diagnosing intraosseous MEC:

1. Absence of any primary lesion in the salivary glands
2. Absence of any odontogenic tumors
3. Radiographic evidence of bone destruction
4. Retention of cortical plate integrity
5. Positive mucin staining and
6. Microscopic confirmation of the diagnosis

This case fulfilled all of these criteria. In the study by He et al. [4] the average age range of the 24 patients was 40 years to 50 years with male-female ratio of 1.67:1 which was consistent with the present case. These intraosseous MEC had 1.18:1 predilection for the maxilla, and in the mandible, whereas in our case the lesion was in mandibular body. To better characterize this tumor, a literature search was conducted in the PubMed database to survey the published case reports of intraosseous mucoepidermoid carcinoma in the last 3 years, as described in (Table 1).

Based on the history, age, site of occurrence provisional diagnosis was given as residual odontogenic cyst most probably dentigerous cyst in relation to extracted 38. The differential diagnosis of unicystic/multicystic lesions in the mandible or maxilla usually includes ameloblastoma and keratocystic odontogenic tumor; however, it should not exclude less common, but more serious conditions, as metastatic tumors; malignant osseous tumors; primary intraosseous carcinoma and malignant salivary glands tumors (Table 1).

Brookstone and Huvos [5] have proposed a staging classification for intraosseous MEC.

1. Stage I: Lesions with intact cortical plates with no evidence of bone expansion
2. Stage II: Neoplasms with intact plates, but intrabony expansion seen.
3. Stage III: Lesions associated with cortical perforation or nodal disease.

According to these categories, the current case can be fitted in stage III. Radiographic features are diverse and not exclusive of CMC. Usually, it appears as a unilocular or multilocular radiolucent lesion with sclerotic and well-defined margins. Da Silva et al. [6] reported mixed radiopaque-radiolucent lesion in mandible, Chundru et al. [7], Kechgias et al. [8], reported multilocular radiolucent lesion in maxilla and Nallamilli et al. [9] reported unilocular radiolucent lesion in mandible; whereas our case present unicellular radiolucent lesion.

In the classification proposed by Waldron and Mustoe [10] (Table 2) our case was a type-4 PIOC, based on the representative histological findings. Common diagnostic imaging features of intraosseous mucoepidermoid carcinoma are well-defined sclerotic periphery, amorphous sclerotic bone internally, multiple small loculations internally, loculations with and without peripheral septa, expansion and perforation of the outer cortex with extension into surrounding soft tissues [11]. In the present case the Figure 8: Cropped a. panoramic, b. CT bone window and c. CECT shows a well defined unilocular radiolucency with sclerotic border (straight arrows) in the left mandible. The buccal cortex exhibits considerable cystic expansion and perforation (curved arrows).

The MEC are usually graded as low grade/well differentiated (tumor exhibiting greater than 50% of mucous elements), intermediate grade (10% to 50% of mucous elements and high grade (less than 10% of mucous elements). Present case showed 10% to 50% of mucous elements and fits under intermediate grade of mucoepidermoid carcinoma. The histopathologic grading is usually used as the main prognostic indicator. Presence of mucous cells, cells with foamy/clear cytoplasm, intermediate cells and lymphocytes in a mucinous background are diagnostic indicators of MEC [12]. Radical resection of the primary tumor should be used for patients with high-grade or intermediate-grade

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**Table 1:** Features of intraosseous mucoepidermoid carcinomas described worldwide in the last three years (2015-2017).

| Authors            | Age | Gender | Symptoms | Anatomical site | Radiographic findings | Diagnosis hypotheses |
|--------------------|-----|--------|----------|----------------|-----------------------|----------------------|
| Chundru et al. [10]| 37  | Male   | Asymptomatic | Mandible       | Multilocular radiolucent | Ameloblastoma/ KCCOT |
| Kechgias et al. [11]| 37  | Female | Pain     | Mandible       | Multilocular radiolucent | Ameloblastoma |
| Nallamilli et al. [12]| 36  | Male   | Pain     | Maxilla        | Unilocular radiolucent | Odontogenic tumor |
| Da Silva et al. [9]| 28  | Male   | Pain     | Mandible       | Multilocular radiolucent | Ameloblastoma |

**Present case (2017)**

| Age | Gender | Symptoms | Anatomical site | Radiographic findings | Diagnosis hypotheses |
|-----|--------|----------|----------------|-----------------------|----------------------|
| 42  | Male   | Pain     | Mandible       | Unilocular radiolucent | Cystic lesion |

**Table 2:** Classification of primary intraosseous carcinoma (PIOC) according to Waldron and Mustoe.
tumors, and adjuvant therapy after surgery is necessary to consolidate the therapeutic effect. The survival rate of patients with radiotherapy was 72.7%; therefore, radiotherapy should be recommended as routine treatment during the postoperative period [13].

Conclusion

Intraosseous mucoepidermoid carcinoma are rare in the jaws, and the clinical presentation of this tumor varies. Therefore, symptoms, such as swelling and paresthesia, and radiographic examination, including computerized tomography, magnetic resonance imaging, or even fine-needle aspiration cytology, are important to clinically confirm the diagnosis. In this case the short duration was also uncommon for a malignant lesion. Clinically and radiographically, since the lesion's behavior was different, it is important that the clinician be aware of the various clinical presentations of a particular disease process. This case report adds a new dimension to the features revealed by a central mucoepidermoid carcinoma.

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