Cystadenocarcinoma of the Mandible

Abstract

Papillary cystadenocarcinoma of the salivary gland is a rare malignant tumor and occurs in major and minor salivary glands. Papillary cystadenocarcinoma of the mandible is exceptionally rare. It is usually a low-grade destructive tumor with a papillary and cystic architecture. This case describes a unique presentation, location, and radiographic appearance of this lesion.

Keywords: Cystadenocarcinoma, mandible, salivary gland, tumor

Introduction

Cystadenocarcinoma is a rare salivary gland tumor which commonly arises in the major salivary glands, mainly the parotid gland. It was first defined by the World Health Organization (WHO) in 1991 as a low-grade malignancy that rarely metastasizes to lymph nodes. It was included as cystadenocarcinoma in the recent 2005 WHO classification of head and neck tumors. They are characterized histologically by prominent cystic and frequently papillary growth but lacking features that characterize cystic variants of several more common salivary gland neoplasms. Epidemiologically, the lesion comprises roughly 2% of malignant salivary gland tumors. Salivary gland lesions that present intraosseously are also rare, only 4 cases being reported in the literature.

The authors describe this case of papillary cystadenocarcinoma arising from a rare location with unique radiographic findings.

Case Report

A 71-year-old male patient came to the Department of Oral Medicine and Radiology with a complaint of swelling in relation to the right lower back region of jaw for 2 weeks with a rapid increase in size. Extraoral examination was negative for swelling or tenderness. The patient had given a history of extraction of a tooth at the same site 6 years back due to mobility. Medical history revealed that the patient was diabetic but was not on any treatment.

Intraoral examination revealed a diffuse swelling involving the edentulous canine–molar region that extends up to lower mucobuccal sulcus obliterating the buccal sulcus buccally and up to lingual sulcus lingually. On palpation, swelling had a variable consistency and was nontender. Surface over the swelling showed an ulcerated area 0.5 cm × 0.5 cm probably due to impingement from the supraerupted canine from the opposing arch [Figure 1].

Panoramic [Figure 2] and lateral radiographs [Figure 3] showed radiolucent lesion with irregular margin extending up to lower border of mandible. Fine-needle aspiration yielded [Figure 4] thick fluid red in color mixed with blood and the smear showed chronic inflammatory cells intermingled with red blood cells.

From the clinical and radiographical examination, a diagnosis of intraosseous carcinoma was arrived, with a differential diagnosis of metastatic lesion. The patient was sent for biopsy for further evaluation.

Histopathology report showed a neoplasm composed of numerous cysts in a desmoplastic stroma lined by mucinous and columnar cells with varying degree of papillary proliferation. They also showed moderate nuclear pleomorphism with scanty mitoses and with no areas of necrosis [Figures 5-7] These histopathological features lead to the diagnosis of cystadenocarcinoma.

The patient was referred to the Oncology Department of Medical College. However, the patient passed away before any treatment could start.

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Cystadenocarcinoma is rarely mentioned in literature because of low incidence and the subestimation of the diagnosis. Cystadenocarcinoma is also termed as papillary cystadenocarcinoma, mucus-producing adenopapillary (nonepidermoid) carcinoma, malignant papillary cystadenoma, or low-grade papillary cystadenocarcinoma and papillary adenocarcinoma.\textsuperscript{[4]}

Cystadenocarcinoma of the salivary gland is a distinct group of epithelial malignancy characterized by cystic and solid areas with luminal papillary projections, lined by cuboidal, columnar, or mucus-secreting cells. Malignant nature of tumor is confirmed by nuclear pleomorphism, mitosis, and infiltrative growth pattern.\textsuperscript{[10]} Microscopically,
The appropriate treatment of the primary tumor is *en bloc* resection, including marginal mandibulectomy and segmental mandibulectomy based on preoperative findings and intraoperative evaluation.[7] Neck dissection is necessary when evidence of any metastatic cervical lymph node involvement is present. Adjuvant radiotherapy and chemotherapy have been reported in cases with perineural invasion, inadequate surgical margin, large tumor, rupture of cortical plate, and soft-tissue invasion.[7,15]

**Conclusion**

Central papillary cystadenocarcinoma of the jaw is an extremely rare tumor with only four previously reported cases in the literature. Clinical, radiographic, and histopathologic examinations should be carefully reviewed to differentiate this neoplasm from other possible malignant neoplasms. The tumor prognosis is still unsettled. Surgical excision with wide margins is the appropriate treatment, while adjuvant radiotherapy should be considered in histologically aggressive or high-stage tumor. Prolonged clinical observation is mandatory.

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**Conflicts of interest**

There are no conflicts of interest.

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