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Case Report

Clear cell odontogenic carcinoma of maxilla: A rare case in a rarer presentation



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ABSTRACT

Clear cell odontogenic carcinoma (CCOC) is a rare odontogenic tumor occurring predominantly in the mandible, most commonly in the 5th to 7th decades. We describe a case of CCOC of maxilla, in a 28-year-old female presenting with right-sided facial swelling and nasal obstruction. The patient underwent a right-sided extended total maxillectomy with orbital exenteration with local forehead rotation flap repair followed by postoperative radiotherapy. Histological and immunohistochemical features were consistent with CCOC. This case exemplifies the need for reviewing ultrastructural details and immunohistochemistry in a clear cell tumor of the jaw.

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1. Introduction

Clear cell odontogenic carcinoma (CCOC) is a rare neoplasm, with 79 cases reported in English literature till date [1–3]. It commonly occurs in the mandible, most common age group belong between 5th and 7th decades [4]. CCOC was first described in 1985 by Hansen et al. as a benign tumor with locally invasive properties [5]. However, in 2005, WHO reclassified it as malignant odontogenic neoplasm owing to its high incidence of local recurrence with nodal and pulmonary metastasis [6]. This report describes a case of CCOC presenting atypically in a young female, at an uncommon site with review of previous cases in the literature with discussion on the management of such a case.

2. Case report

A 28-year-old female presented to the Otorhinolaryngology Department with complaints of a swelling on the right side of face and nasal obstruction for 3 months. On examination, the swelling was 6 cm × 8 cm in size, extending from just below the right infraorbital margin superiorly, till the level of angle of mouth inferiorly, medially obliterating the nasomaxillary groove, with a lateral extension up to 1.5 cm medial to the tragus. The overlying skin was erythematous and tender (Fig. 1A). Anterior rhinoscopy revealed a lateral wall bulge on the right side with a pink polypoidal mass at the level of middle turbinate.

Nasal endoscopy identified the mass arising from the middle meatus (Fig. 1B). A punch biopsy was taken which, on histopathological examination, was suggestive of CCOC. Contrast enhanced computed tomographic scan (CECT) showed a heterogeneously enhancing mass, measuring 6 cm × 8 cm, involving the right maxillary sinus with destruction of the superior, anterior, posterior and medial wall of the maxilla and extension into the right orbit, involving the periorbital tissue and extraocular muscles (Fig. 2A). Involvement of subcutaneous tissue of cheek anteriorly was also noted (Fig. 2B). There was no cervical lymphadenopathy.

An ultrasound abdomen was performed to rule out metastasis from the kidney or ovary, and was normal. Chest X-ray ruled out any apparent pulmonary metastasis.

The patient underwent a right extended total maxillectomy with orbital exenteration with lateral forehead local rotation flap repair

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Fig. 1. (A) Clinical photograph of the patient showing right-sided cheek swelling with associated blood stained nasal discharge. (B) Nasal endoscopic picture showing tumor in the middle meatus and nasal cavity.

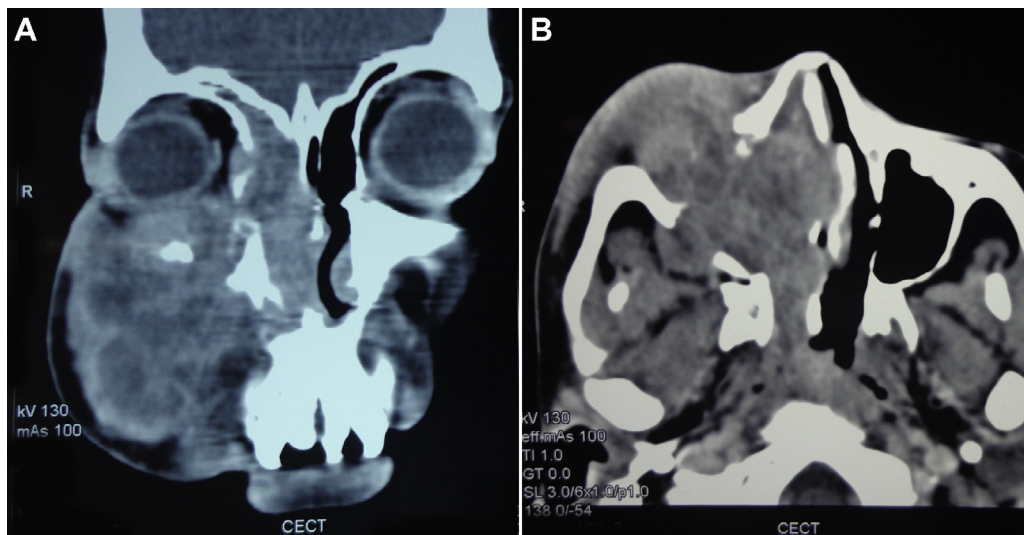


Fig. 2. Contrast enhanced computed tomographic scan of paranasal sinuses showing a large heterogeneously enhancing mass involving the right maxillary sinus causing destruction of the superior, anterior, posterior and medial wall of the maxilla with extension into right orbit involving the periorbital tissue (A: coronal section; B: axial section).

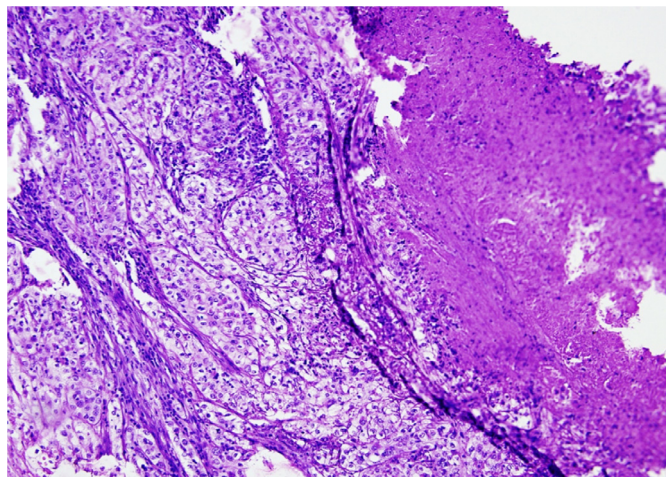


Fig. 3. Sheets of clear tumor cells surrounded by fibrovascular stroma, with areas of necrosis (H&E, 200 \times).

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