CLEAR CELL ODONTOGENIC CARCINOMA OF MANDIBLE

ABSTRACT

Clear cell odontogenic carcinoma (CCOC) a benign but locally invasive odontogenic tumour composed predominantly of clear cells is a rare neoplasm of the jaw. This paper reports the surgical management of a Clear cell odontogenic carcinoma of mandible in a 50 year old female patient.

Keywords: Clear Cell Odontogenic Carcinoma; Odontogenic Tumors; Segmental Osteotomy.

Introduction

Clear cell odontogenic carcinoma (CCOC) is one of the rare benign but locally invasive odontogenic tumours in oral cavity. Literature review suggests that it is more common in anterior region of mandible and has a predilection for females with 45 cases reported in literature. In 1992 WHO defined Clear Cell Odontogenic Tumor as “A benign but locally invasive neoplasm originating from odontogenic epithelium and characterized by sheets and islands of uniform, vacuolated and clear cells.” Subsequent reports of their aggressive behavior, predilection for local recurrence, evidence of pulmonary and lymph node metastases and tumor-related deaths necessitated a change in their classification and nomenclature and is now called CCOC. This paper reports the surgical management of a Clear cell odontogenic carcinoma of mandible in a 50 year old female patient.

Case Report

A 50 year old female was reported to PMS College of Dental Science and Research, Vattappara, Trivandrum, Kerala, India with a chief complaint of swelling of the right lower jaw for the last six months. Swelling was gradually increasing in size. On inspection it appeared diffuse and was roughly oval in shape (Figure 1). The swelling measured approximate size 3cm x 4cm which was firm, tender and not mobile. Patient had moderate pain which was intermittent in nature and was located to the right side of the mandible. The overlying skin appeared normal in colour and was stressed. Right submandibular lymph node was tender and palpable. On intraoral examination the swelling was extending from 33–46 area with obliteration of buccal and lingual vestibule. It appeared as a pink, bulging and fleshy mass (Figure 2).

Radiographic investigation by orthopantomomogram revealed a multilocular radiolucency with irregular borders involving the lower border of right parasympysis of mandible and displacement of anterior teeth. Occulusal radiograph shows lingual cortical plate expansion (Figure 3, 4). Routine blood investigations, ECG and chest radiograph were taken and anesthetist fitness obtained. Medical history was non-contributory. FNAC result was inconclusive. Incisional biopsy was done under local anesthesia. Microscopic examination of histopathological specimen shows large and irregular sheets or cords of neoplastic cells in a richly cellular, collagenous stroma (Figure 5, 6). The neoplastic cells were cuboidal or polyhedral, with centrally placed, rounded nuclei. Some of the neoplastic clusters showed a peripheral rim of cells with abundant eosinophilic cytoplasm and centrally located clear cells. Each cell had a single nucleus with fine chromatin and prominent eosinophilic nucleolus. The cells at the periphery of the nests occasionally demonstrated nuclear palisading away from the basement membrane, i.e., reverse nuclear polarity. Based on the findings diagnosis of clear cell odontogenic carcinoma was entertained. Under general anesthesia segmental osteotomy of mandible with surgical reconstruction using stainless steel plate was planned.

Procedure: Under nasotracheal intubation general anesthesia was established. Intraoral vestibular incision was placed (Figure 7). Segmental mandibular osteotomy was done from 35 to 46 and tumour excision was done with 1cm safety margin (Figure 8, 9). Reconstruction was done with 2mm 20 hole stainless steel plate (Figure 10). The patient was alive with no evidence of recurrence after the initial diagnosis.

Discussion

Clear cell odontogenic carcinoma (CCOC) a benign but locally invasive odontogenic tumour composed predominantly of clear cells is a rare neoplasm of the jaw. This paper reports the surgical management of a Clear cell odontogenic carcinoma of mandible in a 50 year old female with prolonged disease-free survival after relatively conservative surgical treatment. A review of the literature revealed 45 cases of neoplasms with similar clinicopathological features.
The peak incidence of these tumors is in the fifth to seventh decades (mean age, 56.5 years; range, 17-89 years), with a female preponderance (M/F ratio, 10:17) and a prevalent localization in the anterior segments of the jaws; the mandible is affected much more commonly than the maxilla, which was similar to our observation. The overall recurrence rate for these tumors was 55% and local recurrence rates were higher (80%) for curettage alone than for resection alone (43%). Lymph node metastasis on initial presentation was rare (10%) but rapidly increased in those with recurrent disease (33%). Factors such as size of the lesion, soft tissue involvement, lymph node metastasis and most importantly, the presence or absence of positive surgical margins should be considered when developing the treatment strategy.

On the basis of literature review, surgical control of CCOC with an en-bloc resection of bone and soft tissue involvement decreases the risk of recurrence. It is imperative that the surgical margins are free of tumor. If positive margins are noted on permanent section – the treatment will be re-resection to attain tumor-free margins. A regional lymph node dissection can be performed for staging and treatment of regional disease. Adjuvant radiation therapy may contribute to local control in patients with extensive soft tissue invasion which tumor-free margins are not possible or in patients with positive nodes and/or extracapsular spread.

In this case the patient underwent right segmental osteotomy of mandible with surgical reconstruction using stainless steel plate. Postoperative histopathological examination showed tumor morphology similar to incisional biopsy and lymph nodes were negative for the tumor metastasis without any extranodal spread. Even though the patient was referred for adjuvant radiotherapy she refused to undergo the same. At the time of discharge the patient was accepting soft diet and the suture line on the neck and chest was healthy. Subsequent to the discharge, the patient was lost to follow-up only to re-appear after 1 year and at that time had no signs of local or regional spread. Since then patient is again lost to follow-up.

Conclusion
In conclusion, even though CCOC are rare, it should be considered in the differential diagnosis of jaw tumors with prominent clear cell component. As per the literature review the main objective of any treatment plan is to achieve a tumor-free margins by wide surgical resection and loco-regional control by lymph node resection. Local radiation is advised in cases with extensive soft tissue invasion, perineural spread, lymph node metastasis with extra nodal involvement or in cases where tumor-free margins are not possible. A long-term follow-up is essential as these tumors may recur locally or present with late distant metastases.

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