

## CASE REPORT

# Mandibular Mesenchymal Chondrosarcoma: A Rare Case Presentation

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## Abstract

Mesenchymal chondrosarcoma (MCS) is one of the most unusual and rare malignant cartilaginous tumors accounting for less than 2% of all chondrosarcoma. They are aggressive tumours arising in unicentric or multicentric locations from both skeletal and extraskelatal tissues. They are associated with high metastatic and recurrence potential with poor survival. Among the facial skeletal region, jaws are most commonly involved with more preference to maxilla than mandible. In this case report, we describe a rare case of mesenchymal chondrosarcoma involving the mandible in 64-year-old women with detailed literature review of MCS.

## KEYWORDS

Mesenchymal chondrosarcoma, Mandible, Malignant bone tumor, paresthesia

## 1 | INTRODUCTION

MC is one of the most unusual rare malignant cartilaginous tumour of bone and soft tissues with distinct histopathological appearance and biological behavior.<sup>1</sup> Among all four types of Chondrosarcomas (Grade I, Grade II, mesenchymal and myxoid), mesenchymal type is the most aggressive form because of its tendency to grow in deeper tissues.<sup>2</sup> Among the facial skeletal region, jaws are most commonly involved with more preference to maxilla. In this case report we have described a case of MCS affecting mandible.<sup>3</sup> Due to its rarity, documentation of more number of cases will enhance the existing knowledge about the tumor.

## 2 | CASE PRESENTATION

A 64-year-old woman presented with painful swelling on the left lower back region of the jaw since 6 months. Swelling had gradually progressed to the present size and was associated difficulty in chewing and swallowing. Patient experienced paresthesia of lower lip since 2

months. Past medical history and personal history were not contributory. Extra oral examination revealed diffuse swelling in the left middle and lower third of the face measuring approximately 7 X 6 cms in size. Overlying skin appeared normal. On palpation the swelling was firm and tender. Left submandibular lymphnode was enlarged, palpable, tender & mobile. On examination of lower lip there was paresthesia on left side. Patient's mouth opening was restricted. Intraoral examination revealed a proliferative growth involving left mandibular alveolar region, extending anteriorly from the left mandibular premolar region to retromolar region. It extended to involve maxillary tuberosity and left buccal mucosa. The overlying surface was lobulated with presence of indentation of maxillary molars. There was lingual displacement of 37

On palpation, the growth was firm in consistency and tender, 37 and 36 were Grade III mobile 28 was grade II mobile. A provisional diagnosis of carcinoma of left lower alveolus and buccal mucosa was given. Differential diagnosis included squamous cell carcinoma, salivary gland carcinoma and soft tissue sarcoma. OPG revealed diffuse osteolytic lesion present on left ramus and body extending from 36 region to the sigmoid notch. There was destruction of the lower border of mandible,



**FIGURE 1** Proliferative growth in the Left mandibular alveolar and buccal mucosa. 37 is lingually displaced.

anterior and posterior border of ramus. There was destruction of maxillary tuberosity. Uniform widening of PDL space with 33, 34 and 35 was seen



**FIGURE 2** OPG showing osteolytic lesion on left ramus and body and maxillary tuberosity region

PA mandible showed osteolysis of entire body and ramus of mandible, destruction of buccal and lingual cortices with irregular spicules of trabeculae extending from the buccal cortex suggesting periosteal reaction

Submentovertex view showed destruction of buccal and lingual cortices

IOPA revealed ill-defined radiolucency in relation to 36, 37 region with flecks of calcifications, loss of lamina dura in relation to 36,37 and circumferential root resorption with 36, 37 leading to spiking of roots

. Bone and soft biopsy were performed at multiple sites. Histopathological sections revealed characteristic bimorphic pattern composed of sheets of undifferentiated round and spindle cells along with areas of chondroid matrix of variable cellularity

. Cells of chondroid differentiation exhibited both nuclear and cytoplasmic pleomorphism, hyperchromasia along with numerous abnormal mitotic figures

. Histopathologic picture was suggestive of mesenchymal chondrosarcoma. The patient was subsequently advised to undergo computed tomography (CT) of the jaws and chest radiography for further



**FIGURE 3** PA view showing osteolytic lesion in left mandible with periosteal reaction



**FIGURE 4** Submentovertex view showing destruction of left body and ramus of mandible with loss of buccal and lingual cortices

evaluation of the extent of the lesion and to assess for possible pulmonary metastasis and regional lymph node involvement. However, the patient did not return for further evaluation or treatment and succumbed to the disease seven months later.

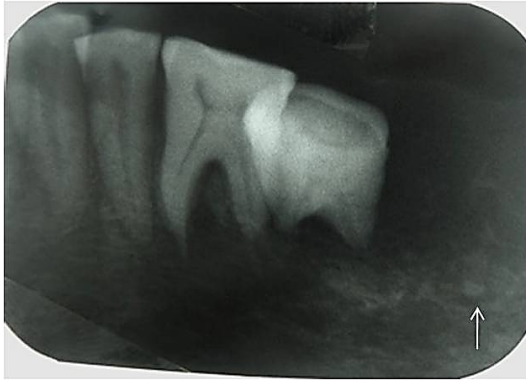


**FIGURE 5** IOPA shows ill-defined radiolucency in relation to 36, 37 region with flecks of calcifications distal to 37. Circumferential root resorption seen with 36, 37 leading to spiking of roots

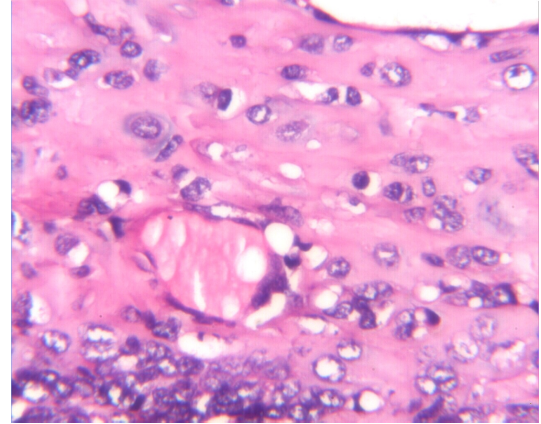


**FIGURE 7** Figure 7: histopathological section (H&E, 20X) showing atypical chondrocytes with hyperchromatic nuclei and hyaline cartilage.

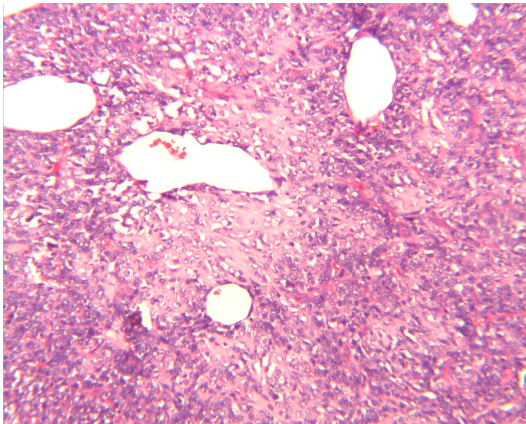


**FIGURE 6** Histopathological section (H&E, 10X) showing bimorphic pattern with round and spindle cells along with areas of chondroid matrix

### 3 | DISCUSSION

Mesenchymal chondrosarcoma is one of the most unusual, rare malignant cartilaginous tumors. It was first described by Lichtenstein and Bernstein (1958) as 'biphasic tumor, comprising of spindle cell mesenchyme interspersed with areas of chondroid differentiation. It accounts for 3% to 25% of all skeletal MCS occurring in the maxillofacial region<sup>2</sup>. Most chondrosarcoma of the head and neck region occur in the maxilla; others are found in descending order of frequency in the body of the mandible, the ramus, the nasal septum, and the paranasal sinuses<sup>4</sup>. This is a unique case of MCS that involved the body and ramus of mandible and extended to involve maxillary tuberosity. MCS variant occurs in 2nd-3rd decade of life in 70% of cases.<sup>5</sup> Very few cases have been reported to occur beyond the 6th decade.<sup>5,6</sup> In this paper we reported MC affecting 64-year-old female. There is no much gender predilection. There are some reports indicating equal prevalence between sexes while others showing a slight male prevalence.<sup>7</sup>

Nakashima Y reviewed series of 111 MCS, where the ages of patients ranged from 5 to 74 years, 60% were in the second and third decades of life. There was no significant sex predilection.<sup>8</sup> Unni KK reported that out of 11,000 primary neoplasms of bone at the Mayo Clinic; only 34 patients had MCS: 27 with skeleton involvement and 7 with soft tissue tumors. Of 27 tumors of bone, 6 were in the jawbones: 3 in mandible and 3 in maxilla.<sup>9</sup> Vencio EF et al reviewed 19 cases of MCS of jawbones among which 10 were males and 9 females with average age of 19 years. Eleven tumors involved mandible and 8 involved maxilla.<sup>10</sup> The most common clinical presentation of MCS of jaws is a painless mass or swelling in comparison to its occurrence elsewhere in the body where pain and swelling are the most common presenting symptoms.<sup>5</sup> Our reported case presented to us as painful swelling on the left side of the jaw associated with paresthesia of lower lip. The radiographic features of MCS are not characteristic. The tumor usually shows features of a malignancy, consisting of a radiolucent process with poorly defined borders. The radiolucent area often contains scattered foci, which are caused by calcification or ossification of the cartilage matrix. It often demonstrates extensive infiltration between the osseous trabeculae of the preexisting bone without causing appreciable resorption. The lesion may also cause symmetrical widening of the periodontal ligament space of the involved teeth.<sup>7</sup> The present case showed similar radiographic appearance of an ill-defined lytic lesion with radio-opaque sclerotic foci, circumferential root resorption and uniform widening of PDL space. The histologic appearance of MCS is characteristically biphasic pattern. Undifferentiated areas appear as sheets of primitive mesenchymal spindle/round cells similar to small cell anaplastic sarcoma. However, islands of relatively well-differentiated cartilaginous tumor help in making a specific diagnosis.<sup>5</sup> Similar histologic features were found in our case. Histologically the lesion must be differentiated from similar other lesions like hemangiopericytoma, Ewing's sarcoma, PNET, leukemia/lymphoma, rhabdomyosarcoma, malignant melanoma and small cell osteosarcoma.<sup>11</sup> Immunohistochemistry is advised for difficult cases. Chondroid areas are positive for S-100 protein, and neuron-specific enolase is focally positive for primitive mesenchymal cells. The most

effective therapeutic modality for MCS is wide surgical excision.<sup>12</sup> Post-operative radiotherapy and chemotherapy offer a good prognosis and eradicate micrometastases that have not been previously detected. Vencio EF reported that hemimandibulectomy and hemimaxillectomy was the main treatment in 16 patients out of 19.<sup>10</sup> The prognosis of MCS is poor because tumours have a tendency for late recurrence either local or via metastasis. Metastasis of MCS is hematogenous and the most common site is the lung and bone.<sup>4</sup> Vencio EF et al reported out of 16 patients treated by hemimandibulectomy, 7 patients had local recurrence, and 5 patients developed distant metastases. Six patients died of disease, and the 5-year and 10-year survival rates were 82% and 56%, respectively.<sup>10</sup> Takashi et al in his review of 41 cases of mesenchymal chondrosarcoma of the jaw found a 58% overall survival rate.<sup>13</sup> Lockhart R reported 4 cases of MCS, in which they followed two patients for more than 17 years, of whom one died of an intracranial extension and the other is still alive after surgical treatment of numerous recurrences and metastases. The two others patients are tumour-free with a follow up of 18 months and 11 years respectively.<sup>14</sup> In the present case patient did not turn up for further investigations and treatment and died after 7 months.

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### How to cite this article:

Patil P, Ramnarayan BK, Ramanand MD, Nayak DS. Mandibular Mesenchymal Chondrosarcoma: A Rare Case Presentation. *World J Dent Excell.* 2026;2(1):25.