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Chondrosarcoma of the mandibular ramus Extending to the infratemporal Space: A Case Report

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Abstract

Sarcomas represent a rare and diverse group of malignant neoplasms that derive histogenetically from tissues with mesenchymal origin (18). Chondrosarcoma is uncommon in the head and neck region, with only 5% to 10% of all chondrosarcomas, occurring within this region of the body. Additionally, chondrosarcomas constitute less than 10% of all head and neck sarcoma diagnoses overall (18). The head and neck Mesenchymal Chondrosarcoma usually emerges in the 3rd-6th decades of life. In the current study, a rare type of Mesenchymal Chondrosarcoma of infratemporal fossa is reported. This rare case of the tumor was cured via a tumor resection surgery, followed by reconstruction with condylar reconstruction plate. Subsequently, radiotherapy and chemotherapy were applied to the patient for 23 and 5 sessions.

Keywords: Mesenchymal Chondrosarcoma, Chondrosarcoma, Infratemporal Fossa, Bone Neoplasm

Introduction

Chondrosarcoma (CS) is known as a rare malignant neoplasm, which slowly enlarges and its origin is primitive cartilage, forming mesenchyme. Pure hyaline cartilage is generated, resulting in anomalous cartilage/ bone growth (1, 2). After osteosarcoma, CS is identified as the second most frequent bone cancer (2). Overall, 5–12 percent CS cases are observed in the head and neck, with 5–10 percent of total bone neoplasms happening in this location. The most prevalent origins of CS include the maxillo-nasal and larynx region (3). The rare cases of CSs include those in the Infratemporal Fossa, which are mainly observed in the mandibular symphyseal area (4-6). Despite the rareness of distant metastasis, its occurrence is in high grade, recurrent, advanced, or cases to the

sternum, vertebrae, and lungs (4). The CS prognosis in jaws is poor in comparison with the CS prognosis of long bones. The direct extension to the skull and via distant metastasis to bones and lungs is the death cause (2). CS has several variants, such as dedifferentiated, mesenchymal, clear cell, and myxoid (7). One of the most abnormal cartilaginous virulence is mesenchymal chondrosarcoma (MC) that occurs in soft tissues and bone. It has aggressive biological behavior and a distinct histopathological appearance. Bernstein and Lichtenstein (1959) originally described MC as the CS separate entity (8). The current study presents a rare case of MC of infratemporal fossa in a male.

Case Report

A 36-year-old male patient was referred to the Department of Oral and Maxillofacial Surgery of Guilan Medical University, in February 2018, by his neurosurgeon with a chief complaint of pain and swelling involving the right side of face for about 2 months.

In clinical examination, the patient suffered from pain and trismus. Extra orally, there was a slight swelling around the Tmj area (figure 1). Overlying skin appeared normal. Intraoral examination revealed the presence of firm, slightly tender swelling on coronoid process area while the overlying mucosa had a normal appearance. The medical and family histories were unremarkable.

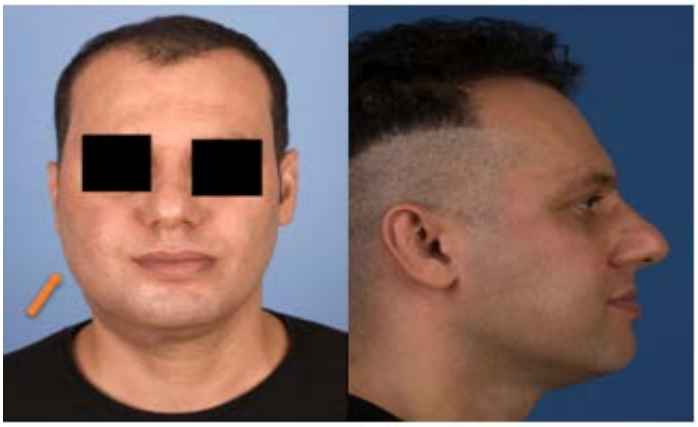

Fig. 1: Extra oral image of swelling

Brain and Face MRI and CT had been taken by the neurosurgeon before referral of the patient and the radiographic examination showed a well-defined solid mass about 70*45*30mm with heterogeneous signal intensity in right infratemporal region extending to masticator space and ptregomandibular fossa and ptregomaxillary fissure. Erosion was noted in ramus of mandible foci of calcification are noted within the tumor. The mass had heterogonous enhancement. No significant regional lymph node was seen. So these finding was suggestive for a chon droid sarcoma e.g. chondrosarcoma.(figure 2 and figure 3)

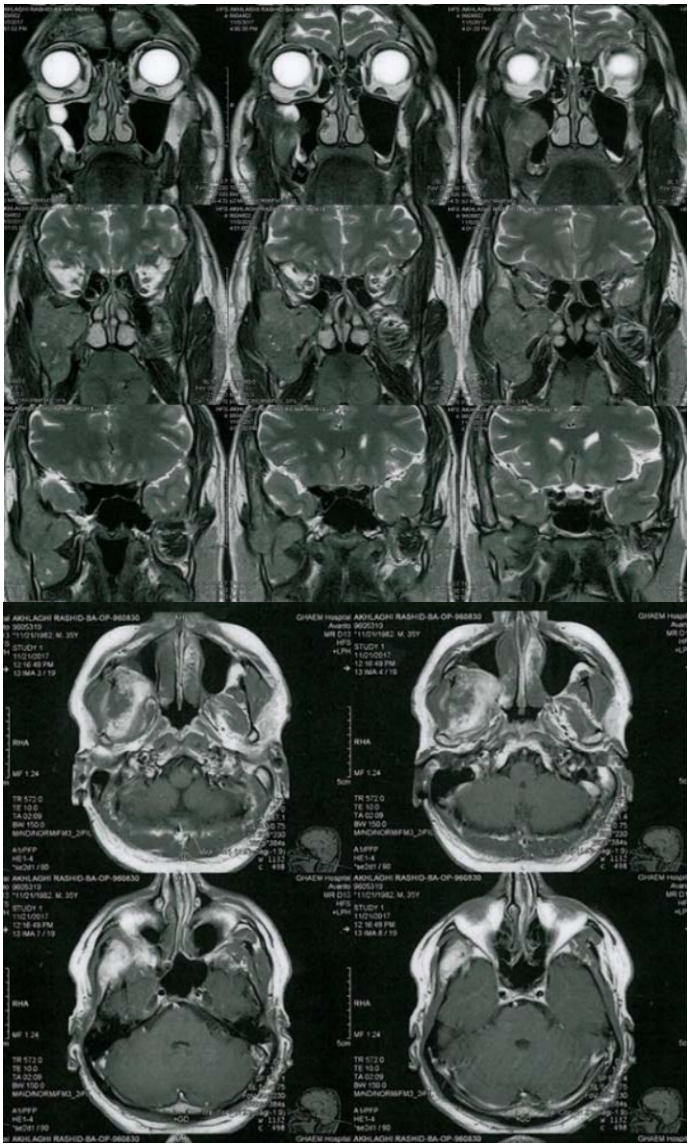

Fig. 2: Face MRI with and without contrast

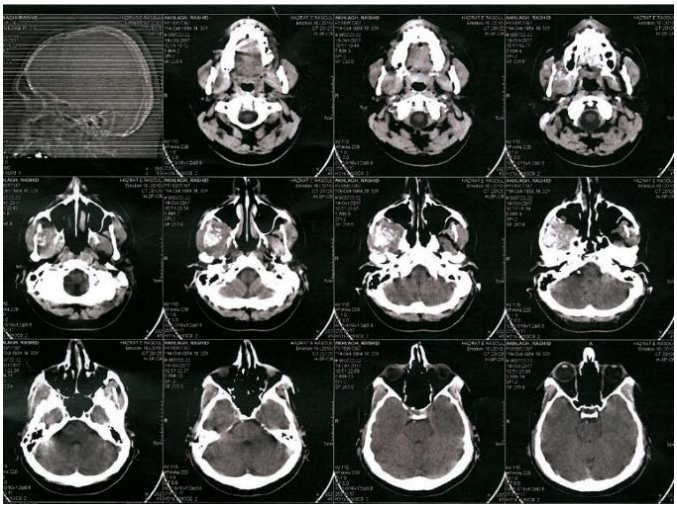

Fig. 3: CT scan showed soft tissue mass in right masticator space extending into sphenopalatine foramen, which contains several rings and crescents calcification as well as ramus erosion of right mandible and RT sphenoid bone and pressure impact at the posterior wall of right maxillary sinuses and Posterior wall maxillary sinus bowed anteriorly. It could suggest a neoplastic lesion like Ossifying fibroma or chondrosarcoma. Opacity was observed in right maxillary sinuses.

Incisional biopsy

Incisional biopsy via intraoral approach was performed under local anesthesia, and the specimen was subjected to histopathologic evaluation. (figure 4) Microscopic examination revealed a fragments of malignant cartilaginous tissue composed of malignant chondrocytes with moderate to severe atypia along with the focal osseous differentiation and telangectatic vessels. According to the histological findings, the pathologist suggested chondrosarcoma, also noting that chondroblastic osteosarcoma should be considered as an alternative diagnostic.

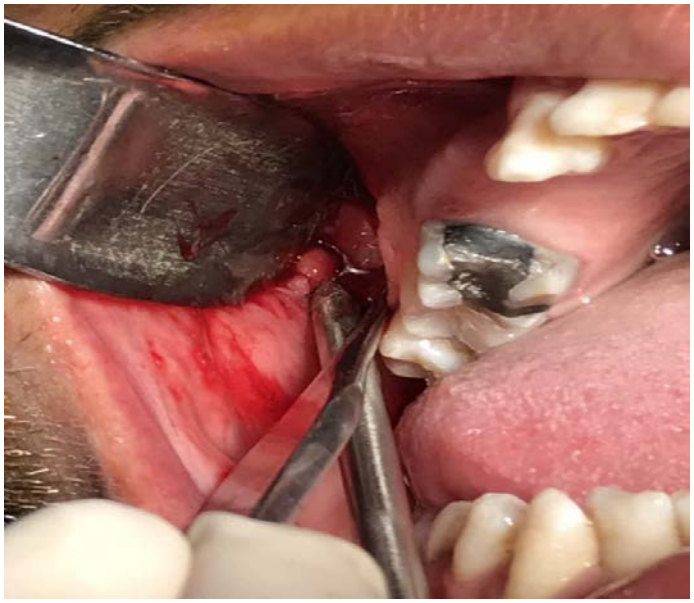

Figure 4: incisional biopsy

Other diagnostic and metastatic workups including, the whole body bone scan, chest x-ray, Para clinical examination like CBC and liver enzymes examination had been completed. And no evidence of metastasis had been showed.(figure 5)

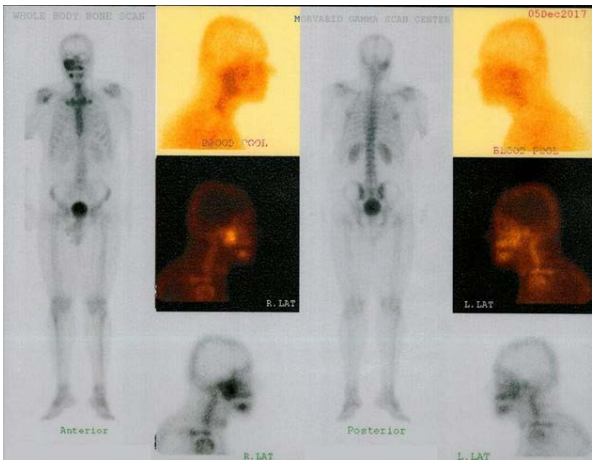

Fig. 5: The bone scan indicated an abnormal increase in uptake in the right maxilla on the static phases and blood pool. A focal increased uptake was observed in the right mandible, which is probably associated with dental disease. X-ray correlation is suggested. There is no significant abnormal uptake in other parts of the skeleton.

Surgery

After a complete diagnostic workup, the Patient prepared for a surgery. Under general anesthesia, submandibular incision, followed by periauricular incision, was made for accessing to the tumor side. The tumor was resected by a block resection of right mandibular ramal area and tumor mass was removed from infratemporal space after which the mandible was reconstructed by condylar reconstruction plate. (figure 6)

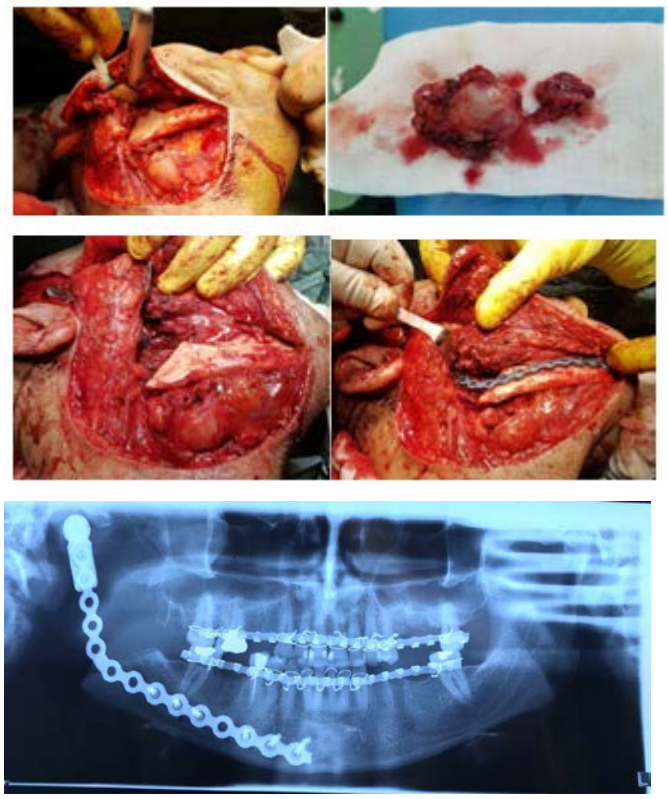

Fig.6: Tumor resection operation

Postoperative histopathological examination confirmed the diagnosis of mesenchymal type chondrosarcoma. (figure 7)

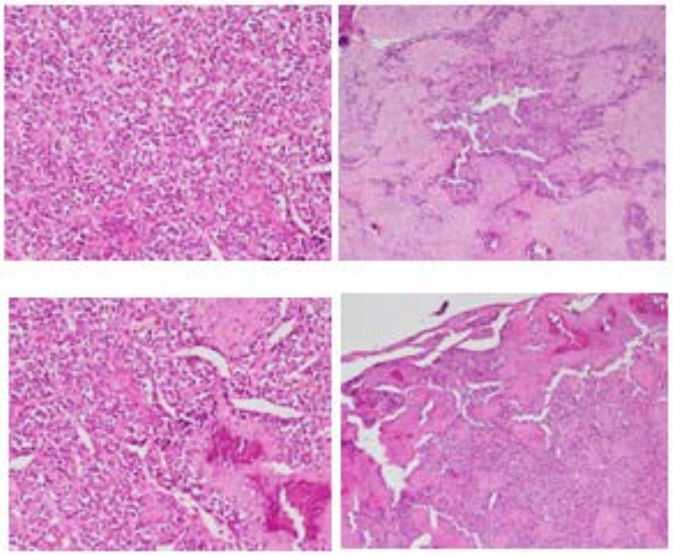

Fig. 7. As shown by the histologic sections, there are malignant cartilaginous tissue fragments that consist of malignant chondrocytes with mild to severe atypia and focal osseous differentiation and telangiectatic vessels, which suggest CS or more precisely MC.

Post-Operation

The patient was then subjected to postoperative radiotherapy because of the possibility of residual tumor cells in area and difficult access to the surgical site. Radiotherapy and chemotherapy were applied to the patient for 23 and 5 sessions and the patient was continuing to receive routine follow-up.(figure 8)

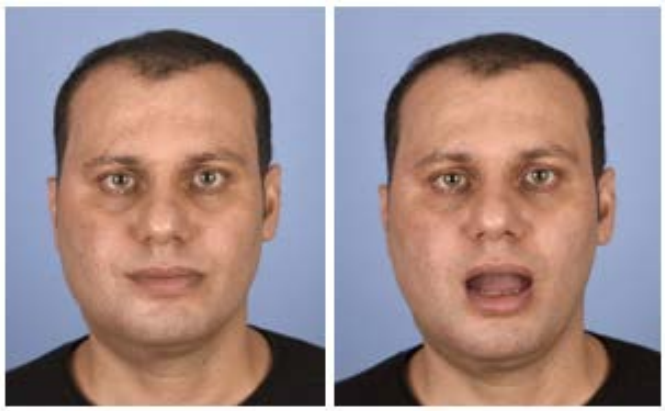

Figure 8: Post-operative images

Discussion

It is usually believed that CS results from embryonic cartilaginous rest or normal chondroid tissues (9). MC is known as one of the most abnormal neoplasms of CS (10). The previous studies have identified 4 CS types, including Grade I and II, myxoid, and mesenchymal. The most aggressive type is mesenchymal as it shows a tendency for growth in deeper tissues (11). The head and neck MC usually emerges in the 3rd-6th decades of life (12). In the present report, the patient was 36 years old. Irradiation may induce this malignant neoplasm in the head and neck, arising from already available Paget's disease in the bone or in relation to Fibrous Dysplasia, or resulting from vestigial cartilaginous rest (2). In the present case, the patient had no systemic disease. Nevertheless, his history of irradiation was not known.

CS of the mandible mainly appears as a painless swelling, or it can be presented as mass of long duration, along with paresthesia, pain, teeth loosening, and trismus. These symptoms imply the disease progress. Nevertheless, these are not pathognomonic findings (13). In the present report, the patient had a pain complaint in the right side of the face. He also reported difficulty in opening his mouth and his face had swelled in the right side. Singh A *et al.* studied a 60-year-old male patient who complained mostly about swelling in his face in lower right side for one month. His history showed that the swelling was small one month ago and it had a rapid reaching the present size (7). A 60-year-old female patient was reported in another study ,complaining about painless swelling at right side of the mandible for six months (14). The final diagnosis in both of these studies was CS of the mandible. According to patients' case presentation in other works, swelling in a side of the face is suggested to be the common major complaint of patients who present CS of infratemporal fossa or mandible (2, 8).

The appearance of the lesion radiographically varies from ill-defined radiolucent to obvious radiopaque shadow. Nevertheless, these are not pathognomonic findings (13). Ordinary radiograph, magnetic resonance imaging, and computed tomography scan are useful tools to determine the lesion's extent and nature (15). In the present case presentation, the brain CT scan and MRI results, as well as face MRI result, suggested CS. However, a histologically definitive diagnosis is required, which indicates that CS is made of hyaline cartilage and fulfill malignant cytologic criteria (16). Also, for differentiating MC from other small cell sarcomas, the Molecular pathology of mesenchymal CS has been evaluated. It has been indicated that MC expresses type II collagen which differentiates it from other small cell sarcomas such as Ewing's, synovial sarcoma and haemangiopericytoma (17). In the present case, according to the incisional biopsy before surgery, there were malignant cartilaginous tissue fragments consisted of malignant chondrocytes with mild to severe atypia with focal osseous differentiation, which suggested CS. The post-surgery pathology laboratory report showed MC.

The wide surgical excision is regarded as the most efficient treatment. It is suggested to apply wide local excision with a tumor-free margin of 2-3 cm. The recurrence rate is less in extensive resection and its survival rate is better ,compared to limited surgical resection (7). Moreover, chemotherapy and radiotherapy after operation provide an appropriate prognosis, eliminating possibility of micro metastases. The patient in the present report took tumor resection operation. He subsequently received 23 sessions of radiotherapy and 5 sessions of chemotherapy. The MC prognosis is poor as tumors tend to have late recurrence locally or as metastasis. MC shows the hematogenous metastasis, with

the lung as the most prevalent region. Five-year- survival rate is 40-60 percent for craniofacial MC (10).

Conclusion

An MC case was presented here as MC is an abnormal tumor, and its occurrence in the infratemporal fossa is rare. The surgical excision with wide margins is required for MCS. Radiation or chemotherapy before and after operation could be considered as a treatment choice, although effectiveness of these treatments is not still clear.

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