Oral metastasis of chondroblastic osteosarcoma

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Abstract

Osteosarcoma is the most common primary malignant mesenchymal tumor, accounting for approximately 20% of sarcomas, with 5% incidence in the jaws. They present various clinical and histological aspects as well as variable disease prognosis and outcome. About 50% of all osteosarcomas are osteoblastic, 25% fibroblastic, 25% chondroblastic. Metastasis of osteosarcoma in the oral cavity is rare, and very few cases have been described so far in the literature. This article presents a metastatic case of chondroblastic osteosarcoma in the mandibular right-attached gingiva arising from 4th rib. This case report further suggests that chondroblastic osteosarcoma has poor prognosis.

Keywords: 4th rib, buccal and lingual attached gingiva, chondroblastic osteosarcoma, mandible, metastasis

Introduction

Osteosarcoma or osteogenic sarcoma (OS) is defined as a primary intramedullary high-grade malignant tumor, in which the neoplastic cells produce osteoid.[1] OS represents the most common primary malignant tumor of bones, accounting for 40-60% of all malignant bone tumors.[2] OS of the maxillo-facial region (primary or metastatic) is rare,[3] which accounts for approximately 5%.[1] The mandible is more commonly involved than maxilla (1.5:1 to 2:1).[4] Metastases from osteosarcoma are most commonly observed in lungs or bone.[5]

Several epidemiological risk factors related to development of OS include- a history of ionizing radiation exposure; fibrous dysplasia; bone cysts; osteogenesis imperfecta; osteochondroma; trauma; hereditary retinoblastoma; or prior exposure to thorium oxide.[6] Mutation in tumor suppressor gene like p53 and retinoblastoma gene (Rb) are more likely to cause this tumor.[7]

Metastases in the mouth and jaw bone are rare, which accounts for 1%.[8] An extensive research has revealed only 3 well-documented cases of metastatic osteosarcoma of the oral mucosa.[8] Here, a fourth case is being reported, in which an osteosarcoma in the 4th rib metastasized to mandibular right buccal and lingual attached gingiva.

Case Report

A 24-year-old male patient attended the OPD of our college with a chief complaint of swelling in lower right back tooth region since 2 months. The swelling was non-tender and initially peanut size, which gradually increased to present size.

On extra-oral examination, a diffuse swelling was seen on the right side of the face, measuring approximately 1.5 × 2 cm extending superiorly 1 cm below the level of outer canthus of the eye and inferiorly 1 cm above the lower border of the mandible, anteriorly from the corner of the mouth to 1 cm in front of the tragus of ear [Figure 2]. Bilateral, sub-mandibular lymph nodes were palpable, which were non-tender, mobile, firm, round to ovoid in shape, measuring approximately 1 × 1 cm.

General physical examination revealed a solitary large nodular swelling on the left side of 4th rib region, measuring approximately 10 × 8 cm, which was well-circumscribed, non-tender, firm, oval in shape. Intra-oral examination revealed two well-defined pedunculated masses of approximately 3 × 2 cm arising from right buccal and lingual attached gingiva in relation to 44, 45, 46, and 47, causing obliteration of buccal and lingual vestibule. A solitary ulceration was present in the...
retromolar pad area covered by pseudo-membranous slough. The lesions were pink in color; smooth-surfaced with areas of indentation of teeth on superior surface, mild tender, and firm in consistency with indurated margins [Figure 3].

Orthopantamograph (OPG) revealed mild bone erosion in relation to 48 [Figure 4].

An incisional biopsy was performed. Histologically, the tumor showed hyperplastic stratified squamous epithelium with rete ridge formation and connective tissue composed of neoplastic cells infiltrating the stroma, with cellular pleomorphism, hyperchromatism and with few mitotic

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**Figure 1:** Chest Computed Tomography Scan Photograph

**Figure 2:** Extra-oral Photograph

**Figure 3:** Intra-oral Photograph

**Figure 4:** Orthopantamograph

**Figure 5:** 10x microscopic view showing areas of chondroid differentiation

**Figure 6:** 40x microscopic view showing tumor cells with cellular pleomorphism and nuclear hyperchromatism
Discussion

Metastatic tumors to the oral region are less common than primary oral lesions. In about 33% of patients, oral secondary tumors are the initial indicators of the existence of the primary tumors. 90% of all oral metastatic tumors occur in the jaws, of which 72% occur in the mandible.[3]

Osteosarcoma is the most common primary neoplasm of the bone, typically affecting the metaphysis of femur or tibia in children or young adults, and is the 3rd most common malignancy in adolescents.[6] In the jaw bones, osteosarcomas occur in older patients, mainly in the third and fourth decades.[9] Mandible molar region is affected in 55% of cases and the premolar region in 38%.[9]

Based on site of origin, osteosarcoma may be parosteal, periosteal, or multifocal varieties. The symptomology of pain, swelling, loosening of teeth, paresthesia, failure of extraction sites to heal, and enlargement or deformity of bone are commonly present.[5]

The present case showed two pedunculated, exophytic masses and an ulcerated region posterior to the pedunculated masses.[8]

Radiographic features of osteosarcoma shows areas of medullary destruction with osteosclerosis or osteolysis. “Sunray” spiculations radiating from the cortex into adjacent soft tissue may be a feature.[9]

Histologically, OS consist of a malignant undifferentiated stroma and neoplastic osteoid formation along with increased mitotic rate of proliferating stromal cells. The stromal component is characterized by dense cellularity and pleomorphism.[6] The 3 main histological types of osteosarcoma include osteoblastic, chondroblastic, and fibroblastic variants with the matrix composed of bone and osteoid, chondroid and collagen in each type, respectively.[6] Other rare subtypes of osteosarcoma include telangiectatic and small cell osteosarcoma, which show blood-filled or empty spaces and small cells with osteoid production, respectively.[10] Histologically, osteosarcoma may have to be distinguished from a malignant fibrous histiocytoma or a poorly-differentiated fibrosarcoma. Exceptionally, an osteosarcoma histologically mimics an osteoblastoma or an aneurysmal bone cyst.[10]

The optimal treatment for oral soft tissue metastases of osteosarcoma is surgical mandibular resection. This is because it remains possible that there are small metastatic deposits present in the jaw bone, although radiographic examinations show no pathological change.[8]

Five year survival rate for primary osteosarcomas of jaws varies from 30-40% with survival up to 80% reported for patients undergoing early radical resection.[3]

The factors associated with poor prognosis include neural sensory alteration as a presenting symptom, increasing age of patients, and surgical margins less than 5 mm.[7]

Conclusion

Osteosarcoma of the jaws is a malignant tumor with rare chances of metastasis. Metastases of primary osteosarcomas from the long bones are considered to be 6 months, but in our case, the mean time from the appearance of primary lesion is 4 years. Prognosis of oral metastatic osteosarcomas is very poor with the mean time from the appearance of metastasis to death being 7.3 months,[7] and in our case, the survival was 1 month after diagnosis of oral lesion.

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