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Case Report

Dentistry Section

Osteosarcoma of the Jaws: Case Report on Synchronous Multicentric Osteosarcomas

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ABSTRACT

Research has shown that osteosarcomas display high potential for metastasis to the lungs, pleurae and bones. Mandible, on the other hand, is an uncommon site for metastatic tumour cell colonization. Nevertheless, a metastatic tumour to mandible might be the first indication of an undiscovered malignancy at a distant site. This case report presents a case of a 61-year-old female patient. An osteosarcoma metastasized to her mandible shortly after the curettage of her jaw cyst. Both the metastatic osteosarcoma and the jaw cyst were confirmed by pathology. Initially, bilateral well-defined radiolucent lesions were shown in her panoramic X-ray image. Also, the diagnosis of a dentigerous cyst was made, based on histology. Two months later, a mixed radiolucent-radio opaque mass, which was confirmed as an osteosarcoma by pathology later, occupied the site of the previously enucleated dentigerous cyst, in her right mandible. Then, an identical osteosarcoma was found in the left pelvis on further doing overall radiological and pathological examinations. The pathologic hypotheses, treatment modality and follow-up of this case have also been presented.

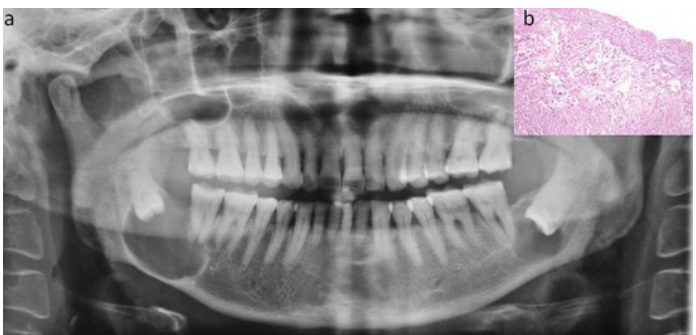
Keywords: Dentigerous cyst, Osteosarcoma, Ilium, Metastasis, Jaw, Clinicopathology

CASE REPORT

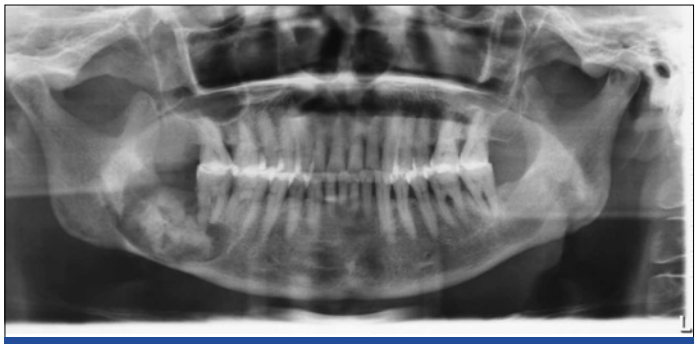
A 61-year-old female who had bilateral cystic lesions in her mandible was diagnosed ten months ago, at a local hospital. She complained that she had felt no pain or numbness in her teeth and mandible. Also, she claimed that she had suffered from no tumours or systemic diseases before. Afterwards, a panoramic X-ray was taken, which revealed some radiolucent lesions that were located bilaterally in her posterior mandible. The result showed that the cystic expansion was surrounded by well-defined cortical margins Each cystic lesion contained an un-erupted mandibular third molar. There was no evidence of root-restoration in the adjacent molar teeth [Table/Fig-1a].

According to the first panoramic X-ray examination, the presumptive result of clinical diagnosis was a dentigerous cyst. Then, the patient was initially treated with bilateral curettage in the local hospital. Meanwhile, an incisional biopsy was taken during the first surgery. The histology of the first biopsy showed a piece of cyst-like tissue, which was lined by non-keratinizing, squamous epithelium [Table/Fig-1b]. But the stromal necrosis was apparent. The histologic diagnosis of the first biopsy was a cystic lesion which was lined by non-keratinizing, squamous epithelium.

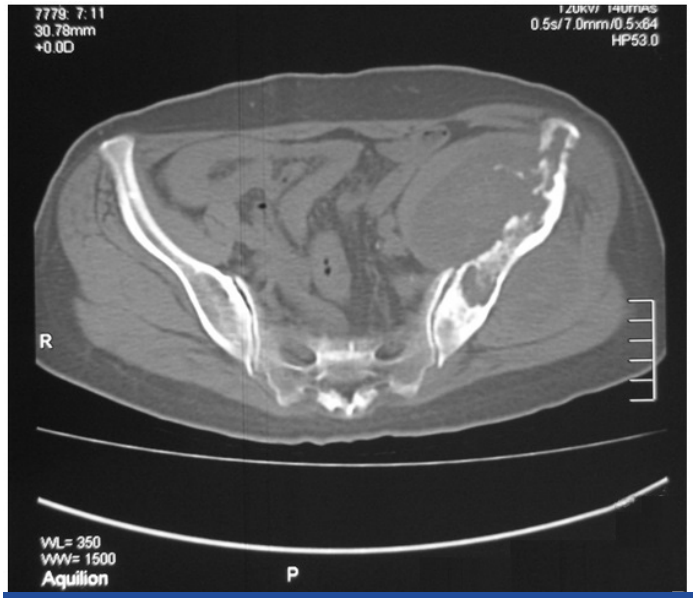
Two months after the curettage, the patient was referred to Peking University School and Hospital of Stomatology, after she complained of a mass on the right inferior gingiva that bled painlessly. Another panoramic X-ray was taken, which demonstrated a well-healed left lesion while the right lesion had become a mixed, radiolucent-radio opaque mass with a diffuse and irregular borderline [Table/Fig-2]. Therefore, a second incisional biopsy was performed. The second biopsy specimen was a piece of bone-like hard tissue. Histopathology showed many atypical cells which were embedded in poorly calcified region and the nuclear fission had increased. This time, a histologic diagnosis of a low-differentiated sarcoma was made preoperatively. In order to investigate the original site of the sarcoma, further Positron Emission Computed Tomography (PET/CT) and Emission Computed Tomography (ECT) were performed. PET/CT demonstrated an enhancing red mass, both in the right mandible and in the left pelvis. The left pelvis had multiple abnormal masses in the left ilium, pubis, ischial and acetabular regions [Table/Fig-3]. ECT showed that an imaging agent was deposited in the right mandible and in the left pelvis. Both the PET/CT and the ECT indicated that the patient had a malignant tumour in her left pelvis. After the general physical checkup, the patient recalled that she had been suffering from hip pain since past two years. This "indisposition" had been ignored by her earlier, as she had been apprehensive that it had been post-menopausal osteoporosis. However, it could have been reasonably attributable to the tumour which had been identified. The radical operation of hemi-dilectomy was performed [Table/Fig-4a,4b]. The specimen showed that the tumour had originated from the bone. It was invasive, pale and expanded to the surface mucosa, with lingual section having same manifestation as buccal section. Microscopically, the specimen had many tumour-like calcification regions and abundant blood vessels. Nuclear atypia, focal clear cytoplasm, marked increased mitotic figures, and focal haemorrhage were also noted [Table/Fig-4c]. A diagnosis of a metastatic osteosarcoma was made postoperatively. One month after the radical mandibular surgery, the patient underwent an operation of half pelvis excision in another bone-specific hospital. The diagnosis was the same as that of the mandible [Table/Fig-4d]. Unfortunately, this patient died of metastatic tumours of the lung and kidney, nine-months after the hemi-dilectomy surgery of the right mandible was done.



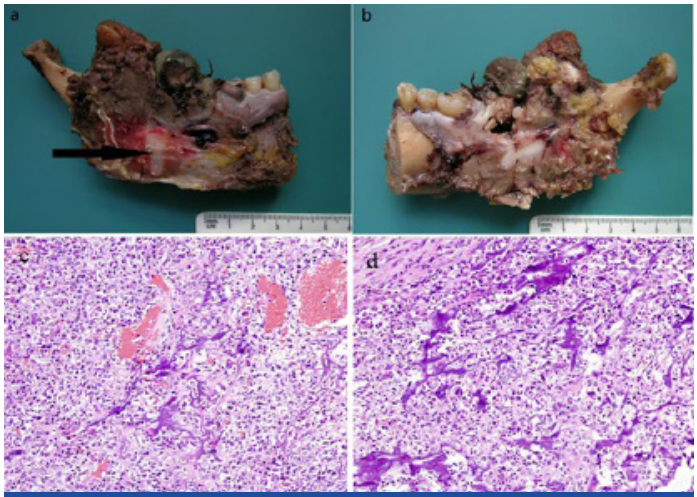
[Table/Fig-1a,b]: The initial panoramic radiograph taken in the local hospital. First biopsy after curettage of bilateral. (haematoxylin-eosin stain, ×20)



[Table/Fig-2]: The second panoramic radiograph taken two months after the curettage



[Table/Fig-3]: Overall PET/CT examination after the second biopsy showed an invasive mass in the left pelvis



[Table/Fig-4a,b,c,d]: Morphology of right mandibular mass after radical hemisection. (a) Macroscopic view of gross specimen of the mandible from the buccal direction. The black arrow suggested the tumor. (b) Macroscopic view of gross specimen of the mandible from the lingual direction. (c) Histological examination of the mandible. (d) Microscopic image of the ilium. c, d appeared nuclear atypia, focal clear cytoplasm, marked increased mitotic figures, and focal hemorrhage. (haematoxylin-eosin stain, ×20)

DISCUSSION

Osteosarcomas of the jaws are rare lesions which represent only 2 to 10% of all osteosarcomas. However, osteosarcomas which occur in the head and neck region are the most common primary malignant bone tumours which represent 23% of total head and neck malignancies [1]. Osteosarcomas which occur in the long bones display high potential for metastasis to the lungs and pleurae, and not the mandible. Nevertheless, the clinical presentation of a metastatic lesion in oral and maxillofacial region could be deceptive, leading to a misdiagnosis of a benign process [2]. Moreover, this situation will become more complex when there is a primary benign disease, followed by a metastatic lesion. The case report which has been presented is of a patient with a metastatic osteosarcoma of mandible, that had developed after patient had undergone a bilateral curettage for a dentigerous cyst. Due to its rarity, the diagnosis of a metastatic lesion in mandible is challenging, for both clinicians as well as the pathologists, in recognizing it and in determining its original site.

Tumours which originate from the prostate, breast and adrenal prefer jawbones as their metastatic targets [3]. In this case, patient's clinical history clearly showed that she was initially diagnosed with a "dentigerous cyst". Then, an "osteosarcoma" was observed in the mandible and finally an "osteosarcoma" was observed in the pelvis. Therefore, it is essential to evaluate as to whether the original diagnosis of a cyst was correct, so that one can identify the origin of this osteosarcoma correctly with confirmity. If the local hospital had misdiagnosed the lesion as a cyst, then the osteosarcoma had to be considered as the recrudescence of the primary osteosarcoma when the patient had come to Peking University School and Hospital of Stomatology.

Due to the histology of her first biopsy, which demonstrated a piece of cyst-like tissue which was lined by a non-keratinizing squamous epithelium, the initial diagnosis of the cyst could then be considered to be reasonable. Simultaneously, the diagnosis of the sarcoma, based on the second histology, was also credible. Moreover, considering the patient's medical history (left-hip pain since two years and a massive lesion in left-hip)it was also reasonable to assume that the mandibular osteosarcoma represented a metastatic focus from a primary osteosarcoma that had originated in the ilium.

The clinical symptoms of a metastatic lesion in jawbones, including swelling, pain and paresthaesia, etc. can cause the metastatic lesion to be misdiagnosed as chronic pulpitis or chronic periodontitis [4]. However, the radiographic description of the metastatic lesion is defined as a lytic radiolucent lesion with ill-defined margins, which may favour the pre-operative diagnosis of metastasis in patients with a known malignant disease. Nevertheless, Hirshberg et al., stated that in 24% of all the 673 patients whom they analyzed, a metastatic lesion in the oral region was the first indication of an undiscovered malignancy at a distant site [5]. These clinical presentations could be easily ignored by both clinicians and patients themselves, as it was in this reported case. Thus, Chorost et al., suggested that a complete history should be taken and that a physical examination should be performed for making a correct diagnosis, with special attention to the breast, rectal and pelvic regions [6].

The exact sequences of the cystic event and the micrometastasis of pelvic osteosarcoma in the right mandible, are still left to be unpredictable. But on the other hand, it was claimed that in the skeletal bones, the frequency of micrometastasis was higher than was found by bone scans and radiographs [7]. Hashimoto et al., reported that, in the mandible, there exist the microscopic deposits of metastatic tumour cells which were not identified in routine radiographic examinations. These deposits of tumour cells were found in 16% of autopsied carcinoma cases according to Hashimoto's study [8].

The pathogenesis of the metastatic process in the jawbones remains unclear. As with the other tumours, the bone metastasis of osteosarcoma consists of many steps, including the migration of cancer cells in the circulation and the colonization of cancer cells. Although the jawbones in the old age are poor in active marrow, the remnants of haematopoietic marrow can still be detected in edentulous mandibles. Therefore, considering the possible pathogenesis of this patient's mandible osteosarcoma, one can

presume that the local blood supply was abundant after the curettage surgery. This could cause the tumour cells to be more likely to be planted in the operated region. Moreover, the sufficient blood supply can provide enough nutrition to the tumour cells as well.

CONCLUSION

This is the first case report that described an osteosarcoma in the pelvis, which had metastasized to the opposite jaw bone. On studying this case, it can be suggested that due to the risk of neoplasm metastasis, before a clinician performs cyst curettage or scrapes a teeth extraction socket, a comprehensive physical examination and history taking should be strongly considered.

REFERENCES

[1] Nthumba PM. Osteosarcoma of the jaws: a review of literature and a case report on synchronous multicentric osteosarcomas. *World J Surg Oncol*. 2012; 10:240.
[2] Forteza G, Colmenero B, Lopez-Barea F. Osteogenic sarcoma of maxilla and mandible. *Oral Surg Oral Med Oral Pathol*. 1986;62:179-84.
[3] Rodan GA. The development and function of the skeleton and bone metastases. *Cancer*. 2003;97:726–32.
[4] Granowski-LeCornu M, Chuang SK, Kaban LB, et al. Osteosarcoma of the jaws: factors influencing prognosis. *J Oral Maxillofac Surg*. 2011;69:2368-75.
[5] Hirshberg A, Shnaiderman-Shapiro A, Kaplan I, et al. Metastatic tumours to the oral cavity-pathogenesis and analysis of 673 cases. *Oral Oncol*. 2008;44:743-52.
[6] Chorost MI, Lee MC, Yeoh CB, et al. Unknown primary. *J Surg Oncol*. 2004;87:191–203.
[7] Cotta CV, Konoplev S, Medeiros LJ, et al. Metastatic tumors in bone marrow: histopathology and advances in the biology of the tumor cells and bone marrow environment. *Ann Diag Pathol*. 2006;10:169–92.
[8] Hashimoto N, Kwrihara K, Yamasaki H, et al. Pathological characteristics of metastatic carcinoma in the human mandible. *J Oral Pathol Med*. 1987;16:362–7.

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