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Case Report

Extra skeletal osteosarcoma of gall bladder: A case report

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KEYWORDS
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Abstract Extraskeletal osteosarcoma is a rare malignant soft tissue tumor. At open cholecystectomy performed for gallstones, a 45-year-old woman was found to have extraskeletal osteosarcoma on histopathological examination. 1 year after surgery, the patient is symptom free and all imaging studies are normal. After multidisciplinary discussion it was decided to give no further treatment. The patient was asked to follow up three monthly. Although osteosarcoma has rarely been reported at other extraskeletal sites, this appears to be the third case of a primary tumor in the gallbladder.

Introduction

Extraskeletal osteosarcoma (ESOS) is a rare malignant soft tissue sarcoma with histologic similarities to primary bone osteosarcoma but without attachment to the bone or periosteum. ESOS accounts for 1% of all soft tissue sarcomas and 4% of osteogenic osteosarcomas [1]. Two cases of gall bladder osteosarcoma have been reported previously; the first by Shin et al. [2] and the second by Olgyai et al. [3]. Although osteosarcoma has rarely been reported at other extraskeletal sites, this appears to be the third case of a primary tumor in the gallbladder.

Case history

A 45-year "old" postmenopausal lady with no co-morbidities presented in January, 2014 with a 1-month history of right upper abdominal discomfort with no nausea or vomiting, no loss of weight or appetite. For this complaint, abdominal sonographic examination was done and reported as acute acalcular cholecystitis with grade 1 fatty liver. Serum amylase and lipase were 310 U/L and 267 U/L respectively. Rest of the blood investigations were normal. In January 2014, elective open cholecystectomy was done for this patient. The patient recovered uneventfully and was discharged in good condition.

Microscopic examination of the gall bladder revealed diffuse ulceration of the surface epithelium. The lamina propria showed infiltration by a malignant neoplasm composed

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of polygonal to spindled cells with marked pleomorphism, hyperchromatic nuclei and frequent mitoses. Presence of malignant osteoid was noted, both as fine lacy deposits and as broad trabeculae. Neoplastic cartilage was also identified. Focally, mature-appearing bone was also seen, as were groups of osteoclastic multinucleated giant cells near areas of hemorrhage. The tumor was infiltrating into the muscularis propria of the gall bladder, but not through it. Post operative CECT chest and abdomen showed compression fracture of D12 vertebral body with D5 vertebral body hemangioma. Whole body bone scan was done for lower backache in February, 2014 which showed uptake in L1 vertebra suggestive of healing compression fracture and uptake in other vertebrae impressive of degenerative changes. No focus of primary tumor or metastasis was seen in any other part of the body on postoperative imaging (see Figs. 1 and 2).

In July, 2014, the patient presented to our institute for further treatment. After multidisciplinary discussion it was decided to go for histopathology review as an institutional protocol and CECT chest, abdomen and pelvis to assess post operative disease status. The patient did not report to hospital for follow up and presented in December, 2014 with no symptoms. CECT chest, abdomen and pelvis showed collapse of D12 vertebra which appeared to be degenerative. Pre operative serum alkaline phosphatase was not done. Post operative serum alkaline phosphatise was 180 U/L and 100 U/L in February, 2014 and April, 2015 respectively. After multidisciplinary discussion it was decided to give no further treatment. The patient is symptom-free and was asked to follow up three monthly (see Fig. 3).

Discussion

Extraskeletal osteosarcoma is defined as the malignant tumor originating from the soft tissues without involvement of the bone and composed of malignant cells of an osteoblastic phenotype producing an osseous matrix [4]. Approximately half the cases of this tumor occur in the lower extremity, and the other half frequently occurs in the upper extremity, retroperitoneum, and the trunk [5]. ESOS is a tumor primarily occurring in older age with a mean age of 47.5–61 years [1]. The majority of case series describe a male predominance [1,4,5]

From a histological point of view, the tumor consists of neoplastic osteoid, bone and cartilage in varying proportions, with or without multinucleated osteoclastic giant cells [6–7]. A reverse zonation pattern may be seen at times. This phenomenon was evident to some extent in our case, with the cellular sarcomatous areas being located directly beneath the surface epithelium, and osteoid and cartilaginous areas being located deeper in the gall bladder wall.

No specific etiological factor is reported to be associated with its occurrence [8,9]. The diagnosis is generally delayed because symptoms are often absent or vague. Histopathological evaluation remains fundamental in the diagnosis of primary extraskeletal osteosarcomas. The histological patterns are varied as similar to osteosarcomas, which includes osteoblastic, fibroblastic/pleomorphic malignant fibrous histiocytoma like, chondroblastic, giant cell MFH like, small cell, mixed and telangiectatic pattern. Nevertheless, the major predictor of clinical outcome was found to be the tumor size (>5 cm) according to the study by Bane et al. [10,11]. In this study, size of the presented tumor was $5\,\text{cm} \times 1.5\,\text{cm}$. Radical resection of the tumor has been considered the appropriate initial treatment because the tumor is not particularly chemosensitive or radiosensitive and resection of a pulmonary metastasis will occasionally produce a cure [11,12]. Radiotherapy may provide temporary palliation, while adjuvant chemotherapy is usually ineffective [13].

An early report described an overall survival rate of 38% at 5 years [14]. However, more recent groups that have used multiagent chemotherapy and wide resection during surgical procedures have described overall survival rates of 66–77%,

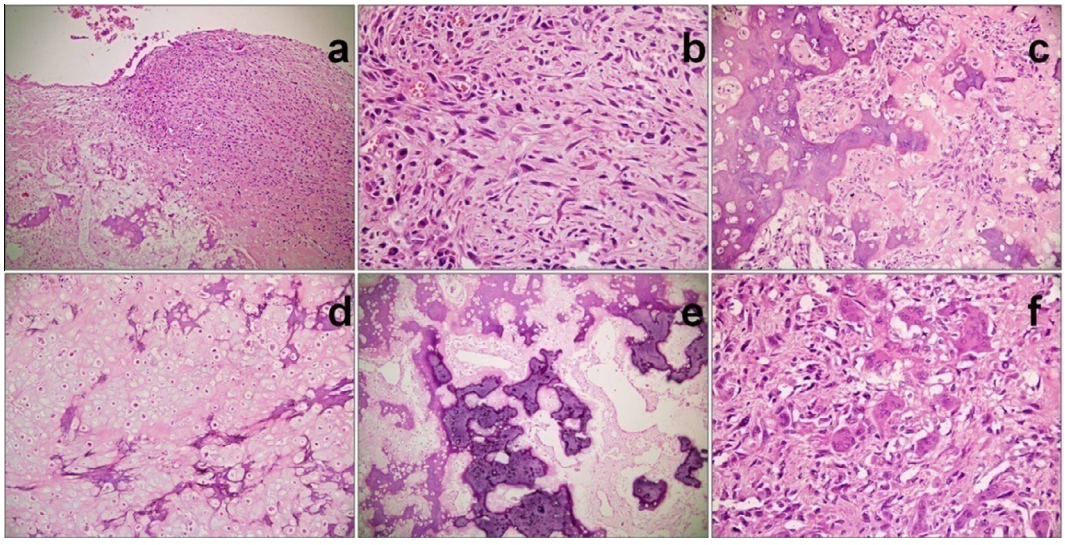

Figure 1 Photomicrographs show a malignant tumor (a; HE, 200×) composed of spindle to polygonal cells with nuclear hyperchromasia and pleomorphism (b; HE, 400×); areas with osteoid (c; HE, 400×), cartilage (d; HE, 200×), bone (e; HE, 200×) and giant cells (f; HE, 400×) are seen.