

Extraskelatal Primary Osteosarcoma of the Gallbladder: A Rare Occurrence—A Case Report

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ABSTRACT

Extraskelatal osteosarcoma (EOS) is a highly aggressive and rare mesenchymal tumor. We present a case of 50-year-old woman who underwent laparoscopic cholecystectomy for symptomatic gallstone disease. Histopathological examination confirmed high-grade osteosarcoma of the gallbladder.

Keywords: Case report, Extraskelatal, Gallbladder, Osteosarcoma.

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INTRODUCTION

Extraskelatal osteosarcoma (EOS) accounts for about 1% of malignant soft tissue tumors which arise from connective tissue embryologically derived from mesenchyme. Extraskelatal osteosarcoma is a high-grade neoplasm occurring most commonly in the extremities, the thorax, and the abdomen. To the best of our knowledge, there have been only four reported cases of EOS occurring in the gallbladder reported till date.¹⁻⁴

CASE DESCRIPTION

A 50-year-old female presented in the outpatient department with history of intermittent right upper abdominal pain, and flatulent dyspepsia of 6 months duration. There was no history of exposure to carcinogenic agents or radiation. Past history was insignificant. General physical and systemic examinations were normal as were the routine blood investigations. Ultrasonography of the abdomen revealed features consistent with cholelithiasis with chronic cholecystitis. The patient was subjected to laparoscopic cholecystectomy (four port). Intra-operative findings were inconclusive. The specimen was sent for histopathological examination. Cut sections revealed a globular mass measuring 3.5 cm × 1.5 cm within the lumen, which was attached to the fundal mucosa of the gallbladder by a narrow base measuring about 1.5 cm. The rest of the mucosa appeared atrophic. Microscopic examination of the sections from the globular lesion revealed features of high-grade sarcoma with numerous giant cells and areas of malignant osteoid formation (Fig. 1). Large areas of cartilaginous differentiation with areas of calcifications were also observed. The lesion was limited to the upper muscular layer of the gallbladder wall. The rest of the gallbladder sections revealed foci of xanthogranulomatous inflammation (Fig. 2). The resection margins and serosa were free and there was no perineural or vascular invasion noticed. The final histological diagnosis of high-grade osteosarcoma of the gallbladder was confirmed. Postoperatively, the patient was subjected to CECT abdomen, chest, and bone scintigraphy, all of which were negative and no further treatment was advised. The patient has been on regular monthly follow-up for last 6 months and has not had any evidence of recurrence or metastasis to date.

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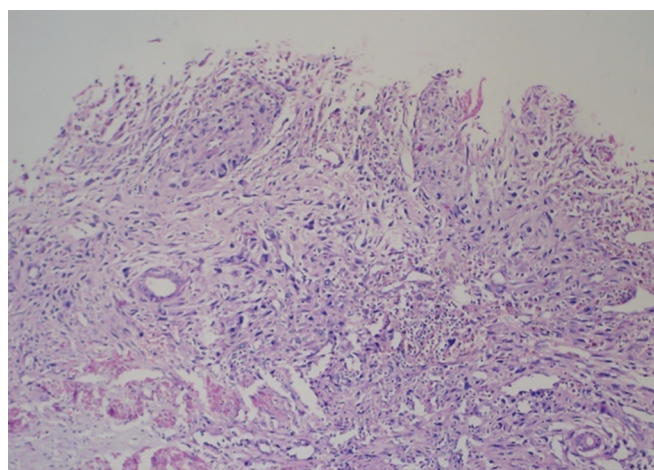


Fig. 1: Sections of the gallbladder show a focus in mucosa with pleomorphic malignant cells infiltrating the superficial muscular layer suggestive of features of high-grade sarcoma with numerous giant cells and areas of malignant osteoid formation

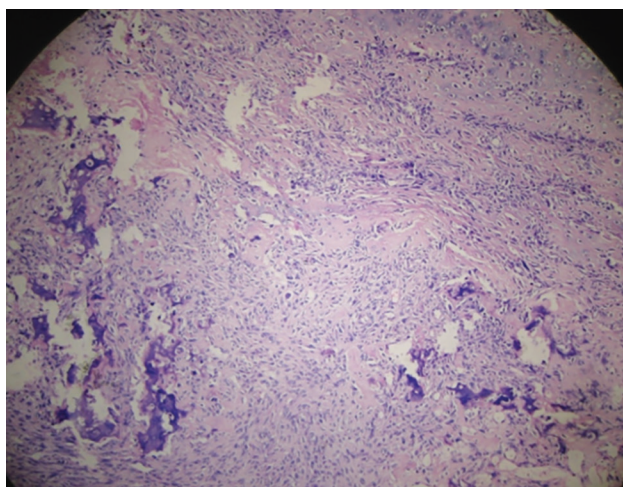


Fig. 2: Sections of the gallbladder revealing large areas of cartilaginous differentiation with areas of calcifications. The lesion is limited to the upper muscular layer of the gallbladder wall. The rest of the gallbladder sections revealed foci of xanthogranulomatous inflammation

DISCUSSION

Extraskeletal osteosarcoma is defined as a malignant, highly aggressive mesenchymal tumor characterized by the production of osteoid in soft tissues without (or with minimal) attachment to the bone or periosteum.⁵ The first case of extraskeletal osteosarcoma was reported by Wilson in 1941.⁶ Extraskeletal osteosarcoma accounts for <1% of all soft tissue sarcomas and approximately 4% of all osteosarcomas.^{7,8} Sites of involvement commonly include deep soft tissue of the thigh, gluteal region, upper extremities, and retroperitoneum, but can occur in any part of the body.⁹ Primary extraskeletal osteosarcoma has also been reported to involve kidneys, urinary bladder, ureter, breast, mesentery, omentum, and liver.^{10–16} Only four cases of extraskeletal osteosarcoma of the gallbladder have been reported in the literature.^{1–4} Olgay G et al.¹ documented the lesion on the serosal aspect, while in our case, the lesion is present on the mucosal surface infiltrating superficial portions of muscularis propria.

Most of the patients with EOS are more than 50 years of age.¹⁵ The exact etiology of extraskeletal osteosarcoma is essentially unknown, although it has been seen in patients treated by radiotherapy for some other malignant lesion. Radiation-induced EOS can develop years following high-dose radiation.¹⁴ The diagnosis is generally delayed as the symptoms of the disease are often vague or even absent for considerable periods. Microscopically, the tumor contains varying amounts of neoplastic osteoid, bone, and cartilaginous tissue in variable proportions with osteo-fibro-chondroblastic or giant (osteoclast) cells.¹

Despite an adequate surgical excision of the tumor with adjuvant chemotherapy which is the best available form of treatment for patients with primary extraskeletal osteogenic sarcoma, they are known to have a poor prognosis.¹² The most common sites of metastases are the lung, regional lymph nodes, and the bone. There is no survival difference between the three main tumor subtypes and the bad prognostic factors include the tumor size and the proliferation index.⁹ More than 80–90% of patients develop either local recurrence or/and metastasis to lungs

and bone. Median survival time is 24 months, and the cause-specific survival rate at 5 years is less than 25%.¹⁷

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