Case Report

Synchronous osteosarcoma of the bladder and adenocarcinoma of the colon: a case report

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Abstract: Introduction: Extraskeletal osteosarcomas are rare mesenchymal tumors. Primary osteosarcomas may arise from parenchymal organs like bladder, liver and heart. The number of cases with osteosarcoma primarily arising from the bladder found to be less than 35 in a literature review. We describe the second patient reported in the English literature with primary bladder osteosarcoma and colon adenocarcinoma. Case report: An 83-year-old male patient presented with complaints of abdominal pain, hematuria, constipation, and weight loss. Computed tomography of the abdomen and pelvis demonstrated a heterogeneous giant mass with lobulated contour measuring 5×3.5 cm at the level of the right ureter orifice in the bladder lumen. The transurethral bladder biopsy showed pleomorphic tumor cells with hyperchromatic nuclei, localized between eosinophilic homogeneous osteoids and fibroblastic malignant cells producing lace-like osteoid matrix. A polyp measuring 2 cm in diameter detected in the colon in the colonoscopy performed for preoperative evaluation. Adenocarcinoma reported as the result of biopsy. Cystectomy and left hemicolectomy in the same session decided. However, the patient refused all surgical and medical interventions. The patient died due to urosepsis 8 months after initial diagnosis. Conclusion: Osteosarcoma of the bladder is a rare tumor with a very poor prognosis. The differential diagnosis gains a significant degree of importance. It would be beneficial to investigate cases like this with regard to synchronous colon and bladder tumors.

Keywords: Osteosarcoma, bladder, synchronous, rare

Introduction

Extraskeletal osteosarcomas are a group of rare mesenchymal tumors. Even though osteosarcomas frequently localized in the lower extremities (47%) and upper extremities (20%), bladder osteosarcomas, which are associated with extraskeletal involvement, observed in only 0.04% of all cases [1]. They have the ability to develop osseous and cartilaginous matrix without bone or periostal tissue involvement. The number of cases with a rather poor prognosis found to be not more than 35 in a literature search. A single case reported in which synchronous osteosarcoma of the bladder and adenocarcinoma of the colon observed [2]. In this report, we describe the second patient reported in the English literature who presented to our clinic with complaints of abdominal pain, weight loss, constipation, and hematuria; a calcified mass noted in computed tomography images and diagnosed as primary bladder osteosarcoma and colon adenocarcinoma.

Case

An 83-year-old male patient presented to the Urology Clinic of Pamukkale University with complaints of abdominal pain, macroscopic hematuria, constipation, and weight loss. His medical history contained nothing of note except a smoking habit of 60 packs/year and coronary artery disease. Upon computed tomography of the patient at the time of diagnosis, a heterogeneous giant mass with lobulated contour measuring 5×3.5 cm at the level of the right ureter orifice in the bladder lumen was detected (Figure 1). Based on this finding, a hematoma and tumor exclusion recommended. Biopsy with transurethral resection was performed in the mass of bladder. On macroscopic examination of the biopsy specimen showed multiple irregular grey white firm tumor bits including necrosis, that measured 15 cc volume and weighed 14 grams. Microscopic examination revealed spindle and pleomorphic appearing tumor cells with bizarre hyperchro-
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Figure 1. Computed tomography image shows a 5×3.5 cm hypodense papillary mass lesion (black arrows) protruding from the wall into the lumen of the urinary bladder that was filled retrograde via a Foley catheter.

Figure 2. Tumor composed of spindle and pleomorphic appearing neoplastic cells with abundant eosinophilic intercellular material (osteoid) and neoplastic bone in the urinary bladder (Hematoxylin-eosin, ×200).

Figure 3. Computed tomography image shows a 2×1 cm pedunculated polyp (white arrows) isodense to the muscles, arising from the wall of the sigmoid colon and protruding into the lumen.

Figure 4. Moderately differentiated adenocarcinoma of the colon (Hematoxylin-eosin, ×100).

Malignant nuclei, localized between eosinophilic homogeneous osteoids, showing chondrosarcomatous differentiation in some regions and involving focal necrotic areas in the lamina propria (Figure 2). Urothelial carcinoma and/or other epithelial tumor components have not observed in any of the sections. Immunohistochemical examination revealed a profile where vimentin was positive and the proliferation index of the tumor cells detected to be 80% as indicated with Ki-67. Pancytokeratin, high molecular weight cytokeratin, CK5/6, p63, ALK, desmin, smooth muscle actin, and S100 markers were negative. Computed tomography has also shown a 2×1 cm pedunculated polyp, arising from the wall of the sigmoid colon and protruding into the lumen (Figure 3) during preoperative assessment. Colonic polypectomy was the removed of polyp which measuring 2 cm in diameter was detected in the rectosigmoid colon in the colonoscopy. Microscopic examination of polypectomy specimen reported as a moderately differentiated adenocarcinoma of the colon (Figure 4). Laboratory parameters of the patient revealed iron deficiency driven anemia and elevated creatinine levels. In the oncology council, cystectomy and left hemicolectomy in the same session have decided. However, the patient refused all interventions, namely surgery, chemotherapy, and radiotherapy and stated that he wanted the best supportive care. The patient died due to uropsepsis 8 months after initial diagnosis.

Discussion

Primary osteosarcomas may arise from parenchymal organs like bladder, liver and heart [2-4]. The incidence of bladder osteosarcoma,
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which is a malignant tumor formed by osteoid tissue, is 4 times higher in men than in women and peaks in the seventh decade of life. In some cases, a history of radiotherapy dating back 10-12 years prior to diagnosis was identified [5, 6]. It has also reported in the literature that three cases had concurrent bladder schistosomiasis [7]. Two cases developed in the background of bladder diverticulitis [8, 9]. Most bladder osteosarcoma cases are localized in the trigon, but there are also cases with pelvic or ureter origins. Solitary, large, polypoid, deeply invasive, and hemorrhagic masses observed. Osteosarcoma is one of the rarest tumors of the urinary bladder; only 5 cases are well documented in the English literature at the last three decades (Table 1).

Four diagnostic criterias for primary extraskeletal osteosarcoma set by Allan [10]. These three criteria are the exclusion of all possible bone primers, malignant osteoid production by the sarcomatous tissue, and the presence of uniform sarcomatous tissue that will exclude the diagnosis of a malignant mesenchymal tumor. Immunohistochemical analysis is negative for pancytokeratin 7 and 20, epithelial membrane antigen, smooth muscle actin, desmin, CD34 and 68; and vimentin and p53 are strongly expressed in >95% of tumor cells [6].

In differential diagnosis, sarcomatoid urothelial carcinoma [11], urothelial carcinoma coexisting with osseous metaplasia [12], and bladder carcinomas should considered [13]. In these types of tumor osteoid formation observed in a small area of the tumor and is generally of a well-differentiated character unlike sarcoma. In addition, irregular pattern and cellular and nuclear polymorphisms are not observed as in osteosarcoma [2]. Bladder osteosarcoma characterized by lace-like osteoid tissue formed by spindle sarcomatous cells. As the prognosis of osteosarcoma is rather poor compared with the other two tumors identified in this study, the differential diagnosis gains a significant degree of importance [14].

Specifically, bladder osteosarcoma has a poor prognosis with most patients dying within 6 months of diagnosis. The tumor has a tendency to behave in a locally aggressive manner. Distant metastases are rare and tumors generally metastasize to the lungs. Additionally, metastasis to the pleura, liver, heart, and lymph nodes may observed [8]. Even though radical cystectomy followed by radiotherapy and chemotherapy recommended, patients die due to obstruction frequently developing after local spread in a short period independent of the treatment approach and due to secondary infection [15].

Conclusion

In conclusion, osteosarcoma of the bladder is a rare tumor with a very poor prognosis. Carcinosarcoma and urothelial carcinoma coexisting with osseous metaplasia, which have better survival rates and share differential diagnosis criteria as two of the three bladder tumors associated with bone formation, should considered. It would be beneficial to investigate cases like this with regard to synchronous colon and bladder tumors.

Disclosure of conflict of interest

None.

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