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Primary Bladder Osteosarcoma: A Rare Entity with an Unfavorable Prognosis

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Abstract

Urothelial carcinoma is the most common malignant neoplasm of the bladder. Malignant mesenchymal tumors comprise <0.04% of bladder malignancies, and with osteosarcoma being extremely rare < 40 cases have been reported till now to our knowledge.1-2 This is a case of 63-year-old female who presented with abdominal pain, hematuria and inability to void with a large necrotic pelvic mass involving the bladder, lower uterus, and cecum. Histomorphological evaluation confirmed the diagnosis of osteosarcoma.

Introduction

Primary osteosarcoma of bladder is extremely rare with an aggressive course and poor prognosis [1]. There is believed to be a slight male preponderance with male to female ratio of 2:1 [2]. The treatment can vary from localized medication to cystectomy with chemoradiation depending upon size and depth of invasion [1]. The presented case is a locally aggressive tumor initially diagnosed as pleomorphic sarcoma on transurethral resection specimen; patient later presented with a necrotic mass and bladder rupture and was subsequently diagnosed osteosarcoma.

Clinical history and microscopic description:

We present a case of a 63-year-old female presented with abdominal pain, hematuria, and inability to void. Imaging study showed lobulated mass measuring 12.5 x 7.9 x 4.1 cm arising from bladder, involving the posterior aspect with abutting and effacement of vagina. Transurethral resection of mass was performed. On microscopy tumor cells showed severe pleomorphism with extensive necrosis and involvement of muscularis propria. Immunohistochemistry was positive for vimentin (figure D) and negative for AE1/AE3, CK5/6. Diagnosis of undifferentiated pleomorphic sarcoma was made, and a gene panel was performed which revealed PIK3CA, FGFR3, TP53, KDM6A, STAG2, and TERT somatic mutation. The patient underwent chemotherapy, but after 3 months she presented with bladder rupture. Imaging showed large necrotic pelvic mass involving bladder and lower uterus and cecum. We received a specimen with 12.8 x 11.7 x 8.6 cm, exophytic, circumferential bladder mass invading perivesical fat, anterior vaginal wall, and cecum (Figure 1A). On microscopy the tumor was composed of spindle cells arranged in fascicles with moderate to severe pleomorphism and multinucleated giant cells with abundant eosinophilic intercellular lace like osteoid with 2-3 mitosis/high power field (Figure 1B, C). Extraskeletal osteosarcoma needs to be differentiated from other bone forming tumors which have better prognosis.

Discussion

Primary extraosseous osteosarcomas are a rare occurrence that can affect the kidneys, brain, lungs, heart, and breasts [3,4]. In order to diagnose primary extraosseous osteosarcoma three factors must be met: a consistent morphological structure of sarcomatous tissue is evident and rules out a mixed malignant mesenchymal tumor, the production of malignant osteoid by sarcomatoid tissue, and the elimination of the possibility of an osseous source [3,4].

Primary urinary bladder osteosarcoma is an extremely rare variant of osteosarcoma, having less than 40 recorded cases and accounting for <0.05% of urinary bladder cancers [2]. This malignancy has been noted primarily in males; however, due to limited data, the ratio for male-to-female prevalence has been reported to be anywhere from 2:1 - 4:1 [2,7]. Analysis of recorded cases showed a reported age range anywhere from 24-83 years old at the time of diagnosis [5-14]. Upon presentation, patients had a wide array of symptoms, but common symptoms seen amongst the cases included: dysuria, hematuria, and flank pain [2,7-10]. Additionally, a majority of individuals diagnosed with primary urinary bladder osteosarcoma were noted to have comorbidities; however, cyclophosphamide usage and a previous history of transitional cell carcinoma were comorbidities that have raised suspicion of increasing susceptibility to primary urinary bladder osteosarcoma [9,11]. Currently, treatment of primary urinary bladder osteosarcoma varies based on patients' age, comorbidities, personal preferences, and extent of the cancer. Most cases have been treated with segmental or complete cystectomy with pelvic/abdominal lymphadenectomy and resection of surrounding affected structures – sometimes with adjunctive chemotherapy and radiation [2,6-11]. Differential diagnosis of primary urinary bladder osteosarcoma includes stromal osseous metaplasia of urothelial metastasis, carcinosarcoma of the bladder, and urothelial cell carcinoma of the urinary bladder [15-18]. The listed malignancies have similar clinical presentations consisting of hematuria, dysuria, issues with voiding, and abdominal/pelvic pain. Additionally, computed tomography imaging of the cancers are quite similar as well showing pelvic masses with heterogeneous areas of necrosis and hypoattenuation [16-18]. However, upon histological analysis, primary urinary bladder osteosarcoma yields a unique pattern of spindle shaped cell bundles within a lace-like osteoid matrix [2,7-8]. Stromal osseous metaplasia of urothelial metastasis and urothelial cell carcinoma of the urinary bladder show matured osseous metaplasia, while carcinosarcoma of the bladder shows sarcomatoid and epithelial features [16,17]. Given the rarity of primary osteosarcoma of the urinary bladder, this case lends itself to further adding to the information collected from the past case studies. According to Oka et al., there have been a total of 38 reported cases of primary bladder osteosarcoma, making this study the 39th case according to the analyzed data [1]. The patient in this case fell within the age range at time of diagnosis when compared to the previous studies, and had similar microscopy to previous stud-

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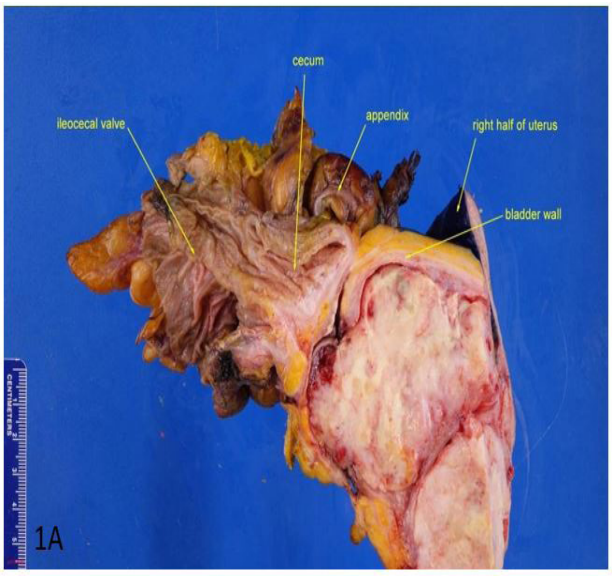


***Figure 1A**: Gross microphotography showing exophytic, circumferential bladder mass invading perivesical fat and cecum.*

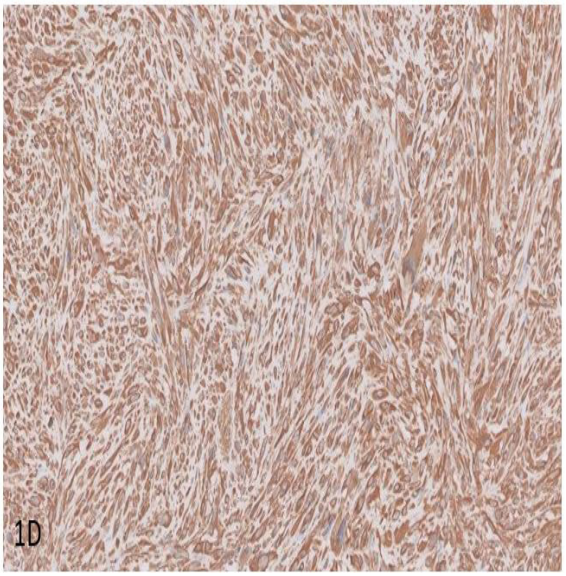
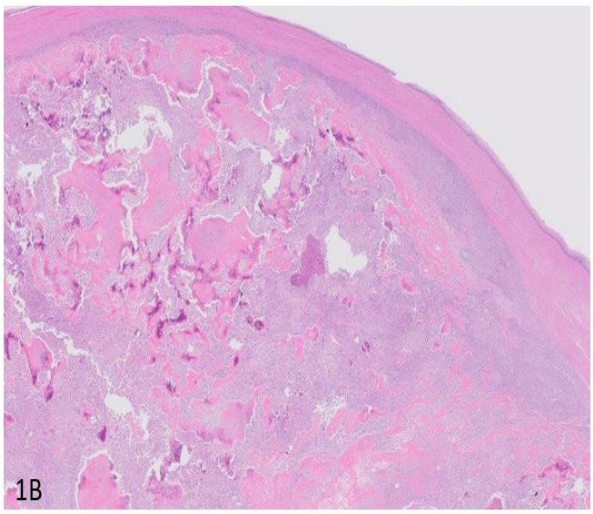


***Figure 1D:** Immunohistochemical staining of vimentin shows diffuse cytoplasmic expression.*



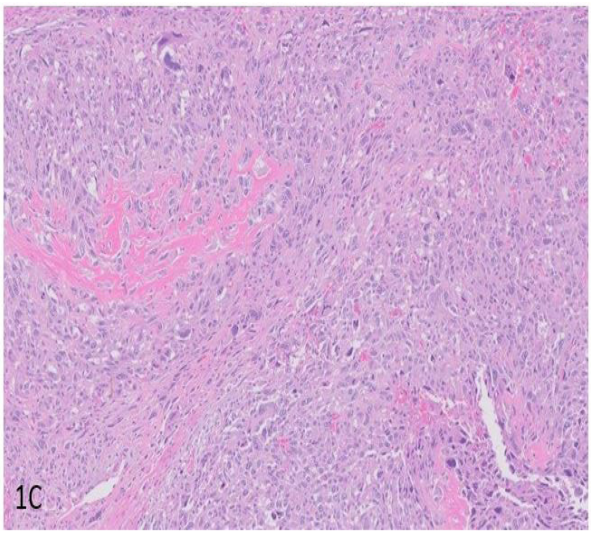


***Figure 1B & 1C**: Hematoxylin and Eosin-stained section showed spindle cells arranged in fascicles with moderate to severe pleomorphism and multinucleated giant cells with abundant eosinophilic intercellular lace like osteoid with 2-3 mitosis/high power.*

ies. Also, this individual's symptoms were aligned with most previous cases – including flank pain and gross hematuria. Where this case differs from previous cases is both in the macroscopy of gene paneling. In comparison to previous cases, the presented case showed a much higher degree of local invasion by the cancer warranting a complete cystectomy of the bladder with ureteroileal conduit/sigmoid bladder pelvic lymphadenectomy along with robotic total abdominal hysterectomy and partial vaginectomy due to extraperitoneal bladder perforation as well as possible cecum-bladder fistula (Figure 2). Also, in comparison to previous research, extensive gene paneling was conducted on the pleomorphic sarcoma, which showed PIK3CA, FGFR3, TP53, KDM6A, STAG2, and TERT somatic mutation.

Conclusion

While primary osteosarcoma of the urinary bladder is extremely elusive, research into this malignancy will result in earlier diagnosis and better prognosis. Additionally, if further exploration into primary extraskeletal osteosarcomas and their linked gene mutations yields pertinent data, it may be possible to see if certain individuals are susceptible to developing such a cancer. Such discoveries can influence the development of treatments and interventions as well.

Reference

1. Oka T, Matsuzaki K, Izumi H, Nonomura D, Mori K, Nishimura K. Osteosarcoma of the bladder: a case report. Journal of Medical Case Reports. 2022;16(1). doi:https://doi.org/10.1186/s13256-022-03346-2.
2. Mremi A, Pyuza JJ, Bartholomeo N, Andongolile AA, Yahaya JJ. Diagnosis of primary osteosarcoma of the urinary bladder in a resource-limited setting: a case report and review of the literature. Journal of Surgical Case Reports. 2020;2020(7). doi:https://doi.org/10.1093/jscr/rjaa236.
3. Dekkers IA, Cleven A, Lamb HJ, Kroon HM. Primary Osteosarcoma of the Breast. RadioGraphics. 2019;39(3):626-629. doi:https://doi.org/10.1148/rg.2019180181.
4. Namdari F, Nazar Pour MJ, Behzadi A, Akbari M, Moradi H, Dialameh H. Primary renal osteosarcoma: A case report and review of literature. Clinical Case Reports. 2022;10(6). doi:https://doi.org/10.1002/ccr3.5957.
5. Allan CJ, Solle EH. Osteogenic sarcoma of the somatic soft tissues.Clinicopathologic study of 26 cases and review of literature. Cancer. 1971;27(5):1121-1133. doi:https://doi.org/10.1002/1097-0142(197105)27:5%3C1121::aid-cncr2820270519%3E3.0.co;2-3.
6. Chung EB, Enzinger FM. Extraskeletal osteosarcoma. Cancer. 1987;60(5):1132- 1142. doi:https://doi.org/10.1002/1097-0142(19870901)60:5%3C1132::aid- cncr2820600536%3E3.0.co;2-l.
7. Elwy A, Soliman A, Maher M, et al. Primary Osteosarcoma of the

Urinary Bladder Metastatic to Lung. Journal of Medical Cases. 2021;12(7):280-283. doi:https://doi.org/10.14740/jmc3613.

8. Siddappa JK, Singla S, Jain A, Kumar A. A Rare Case of Primary osteosarcoma of urinary bladder. Journal of Clinical Imaging Science. 2012;2:82. doi:https://doi.org/10.4103/2156-7514.105145.

9. Baydar DE, Himmetoglu C, Yazici S, Kiziloz H, Ozen H. Primary osteosarcoma of the urinary bladder following cyclophosphamide therapy for systemic lupus erythematosus: a case report. Journal of Medical Case Reports. 2009;3(1). doi:https://doi.org/10.1186/1752-1947-3-39.

10. Grubišić I, Leniček T, Tomas D, et al. Primary osteosarcoma of bladder diverticulum mimicking intradiverticular calculus: a case report. Diagnostic Pathology. 2011;6(1). doi:https://doi.org/10.1186/1746-1596-6-37.

11. Berenson RJ, Flynn S, Freiha FS, Kempson RL, Torti FM. Primary osteogenic sarcoma of the bladder. Case report and review of the literature. Cancer. 1986;57(2):350-355. doi:https://doi.org/10.1002/1097-0142(19860115)57:2%3C350::aid-cncr2820570227%3E3.0.co;2-s.

12. Chitiyo ME. Primary osteogenic sarcoma of the urinary bladder. The Journal of Pathology. 1973;111(1):53-56. doi:https://doi.org/10.1002/path.1711110109.

13. Phan CN, Wilkinson M, Cohen MB, Dunn S, Carroll P. Primary osteogenic sarcoma of the urinary bladder successfully treated with combination therapy. Urology. 1994;44(5):771-774. doi:https://doi.org/10.1016/s0090-4295(94)80228-9.

14. Nourse MH. Primary Osteogenic Sarcoma of the Bladder. Journal of Urology. 1957;77(4):634-638. doi:https://doi.org/10.1016/s0022-5347(17)66610-2.

15. Powers JH, Van Zandt Hawn C, Carter RD. Osteogenic Sarcoma and Transitional Cell Carcinoma Occurring Simultaneously in the Urinary Bladder: Report of a Case. Journal of Urology. 1956;76(3):263-269. doi:https://doi.org/10.1016/s0022-5347(17)66691-6.

16. Atılgan D, Gençten Y. Carcinosarcoma of the Bladder: A Case Report and Review of Literature. Case Reports in Urology. 2013; 2013:1-3. doi:https://doi.org/10.1155/2013/716704.

17. Razafimahefa J, Gosset C, Mongiat-Artus P, Andriamampionona TF, Verine J. Stromal osseous metaplasia in urothelial carcinoma of the bladder: a rare case report and literature review. Diagnostic Pathology. 2019;14(1). doi:https://doi.org/10.1186/s13000-019-0851-z.

18. Dudley AG, Tomaszewski JJ, Hughes AH, Davies BJ. Incidentally Discovered Osseous Metaplasia Within High-grade Urothelial Carcinoma of the Bladder. Urology. 2012;79(4): e59-e60. doi:https://doi.org/10.1016/j.urology.2011.06.043.