Undifferentiated sarcoma of bladder with sarcomatoid carcinoma: A case report

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1. Introduction

Undifferentiated sarcoma of the bladder refers to the shape of tumor cells and normal tissue cells. Malignant tumors with a high malignancy vary widely and have highly high genomic complexity.\textsuperscript{1} Sarcomatoid carcinomas are some poorly differentiated carcinomas in which the cancer cells appear spindled or pleomorphic. It is considered an aggressive variant in bladder malignancies. It is a more specific and rare tumor type of urothelial variant origin, accounting for only 0.1–0.3% of bladder cancers of all histologic types.\textsuperscript{2} Currently, the diagnosis of these two diseases mainly depends on the results of the pathological biopsy. As for the treatment plan, most researchers recognized the early diagnosis and active surgical treatment as the treatment plan, but it seems that postoperative patients still have tumor recurrence and multiple metastases. This article reported a case of a pathological diagnosis of undifferentiated sarcoma with sarcomatoid carcinoma. To the best of our knowledge, this is the first report that two types of diseases co-occur in one patient’s bladder. The tumor recurrence and metastasis progressed rapidly, and the patient’s survival time was significantly shortened.

2. Case information

This is a female patient the age of 62 years old. She was admitted to the hospital on April 15, 2021, due to “intermittent gross hematuria.” The patient presented with gross hematuria throughout the course, accompanied by abdominal pain, frequent urination, urgency, and dysuria. The ultrasound suggested possible bladder cancer. Physical examination: With percussion pain (+) in the bilateral kidney area, no tenderness in the double ureteral area. The diagnosis considered bladder space-occupying lesions.

Further improvement of the CT scan and enhancement showed an irregularly thickened bladder wall, a formed mass of about 6.4×5.0cm (Fig. 1). The growth broke through the bladder wall. On April 20, 2021, the patient underwent transurethral resection of bladder tumor biopsy.

Postoperative pathological diagnosis: undifferentiated bladder sarcoma combined with sarcomatoid carcinoma should be mainly considered. Immunohistochemical results showed: CK-pan (−), UroplakincI\textsubscript{II} (−), CK7 (−), CK20 (−), p40 (−), GATA3 (−), HMB45 (−), S-100 (−), p63 (−), INI1 (−), CD34 (−), Syn (−), CgA (−), CD56 (−), Vimentin (−), SMA (individual +), desmin (−), LCA (−), CD30 (−), ALK (−), Ki-67 (+, ∼60%) (Fig. 2A–C). On May 10, 2021, radical cystectomy + ileal catheterization was performed. Postoperative pathological diagnosis: The tumors in the body of the bladder were all malignant tumors with necrosis, with a size of about 5.5×5.0×3.2cm, and invaded the adipose tissue around the bladder; invaded the submucosa; tumor thrombus was found in the blood vessels in the surrounding adipose tissue, but no definite nerve invasion. Fifteen days after the operation, the CT showed (Fig. 3A): No residual tumor was found. The patient improved and was discharged. The discharged diagnosis was undifferentiated bladder sarcoma with sarcomatoid carcinoma.

One month after, the patient was admitted to the hospital due to “oliguria with fever.” The CT scans showed: The possibility of tumor recurrence and metastasis progressed rapidly, and the patient’s survival time was significantly shortened.

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bone metastasis and pleural metastasis should be considered. Multiple non-uniformly enhanced soft tissue density masses were newly added to the pelvis (Fig. 3B). Bone destruction of the left occipital bone with a noticeable enhancement of soft tissue mass inside and outside the skull. Diagnosis considers “Bladder tumor with multiple metastases.” Finally, the patient improved and was discharged. Telephone follow-up: the patient eventually died one week after discharge. It was considered to be caused by multiple metastases of the tumor throughout the body.

3. Discuss

Sarcoma is a rare mesenchymal cancer, accounting for less than 1% of all malignant tumors, but there are still sarcomas that cannot be distinguished. Pathologists classified these sarcomas as undifferentiated sarcomas. The lack of objective diagnostic and classification standards has led to slow progress or even stagnation of research in this area. In undifferentiated sarcoma, the cancerous component is positive for α-1-Antichymotrypsin and macrophage-derived CD68, and the mesenchymal marker Vimentin is positive. So, it is relatively recognized to diagnose it based on immunohistochemistry.

Sarcomatoid carcinoma is a malignant tumor with bidirectional differentiation of epithelial and mesenchyme. In immunohistochemistry, the cancerous component is positive for the epithelial markers pan-keratin and epithelial membrane antigen while positive for the mesenchymal marker vimentin in approximately 80%–90%. We know two intermediate filament proteins, keratin and vimentin, distinguish epithelial and mesenchymal phenotypes, respectively. Therefore, the diagnosis mainly relies on immunohistochemistry. There is currently no specific treatment guideline. So some researchers recommend giving up transurethral resection of bladder tumor for its treatment plan and directly performing radical total cystectomy. Other researchers advocated the use of neoadjuvant chemotherapy and radiotherapy combined with surgery. In this case, the patient was diagnosed with undifferentiated sarcoma with sarcomatoid carcinoma based on immunohistochemistry.

Moreover, further radical cystectomy was performed. One month after the operation, she died due to multiple tumor metastases throughout the body. Relevant studies have proved that the median survival time of patients with undifferentiated sarcoma is about 12 months, while the median survival time of patients with sarcomatoid carcinoma is only 11 months. However, in this case, the two types of tumors were concentrated in the bladder of the same patient. Only one month, metastasis and recurrence occurred. The rate of disease progression is speedy, and the postoperative survival is only one month.

Compared with bladder urothelial cell carcinoma, there is no treatment plan for this type of tumor. Therefore, for such diseases that progress so rapidly, whether the administration of adjuvant chemoradiotherapy and the choice of treatment timing can improve the survival rate of patients needs further research.
4. Conclusion

In conclusion, developing new and more effective treatments for improving survival in this group of patients requires multicenter and large-sample studies. Relevant statistics showed that cases with undifferentiated bladder sarcoma combined with sarcomatoid carcinoma are rarely reported. Therefore, this paper reviewed the diagnosis and treatment process of a patient with an undifferentiated bladder sarcoma combined with sarcomatoid carcinoma, aiming to improve the understanding of the disease, and can provide appropriate ideas and basis for early diagnosis, treatment, and timing of treatment of the disease.

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Ethics declarations

Ethics approval and consent to participate.

Written consent to participate was obtained from the subject discussed in this article.

Author contribution statement

XT drafted the article. XQ, BL, and YYG performed the surgery. XC participation in Diagnosis and Treatment of Patients. ZY made critical revisions for important intellectual content. All authors read and approved the final article.

Declaration of competing interest

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Not applicable.

Abbreviations

Not applicable.

References

1. Steele CD, Pillay N. The genomics of undifferentiated sarcoma of soft tissue: progress, challenges, and opportunities. Semin Cancer Biol. 2020;61:42–55. https://doi.org/10.1016/j.semcancer.2019.11.009.
2. Wright JL, Black PC, Brown GA, et al. Differences in survival among patients with sarcomatoid carcinoma, carcinosarcoma and urothelial carcinoma of the bladder. J Urol. 2007;178(6):2302–2307. https://doi.org/10.1016/j.juro.2007.08.038.
3. Cheng L, Zhang S, Alexander R, et al. Sarcomatoid carcinoma of the urinary bladder: the final common pathway of urothelial carcinoma dedifferentiation. Am J Surg Pathol. 2011;35(5):c34–e46. https://doi.org/10.1097/PAS.0b013e3182159dec.
4. Urrea YR, Epstein JI. Sarcomatoid carcinoma associated with small cell carcinoma of the urinary bladder: a series of 28 cases. Hum Pathol. 2017;67:169–175. https://doi.org/10.1016/j.humpath.2017.08.008.
5. Wang J, Wang FW, LaGrange CA, et al. Clinical features of sarcomatoid carcinoma (carcinosarcoma) of the urinary bladder: analysis of 221 cases. Sarcoma. 2010;2010:1–7. https://doi.org/10.1155/2010/454792.

Fig. 3. A. 15d after operation, abdominal CT showed no obvious recurrence and new organisms in the pelvis. B. Abdominal CT at one month after operation showed that there was a new mass in the pelvis, and the larger section was.