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Renal metastasis of osteosarcoma after multiple pulmonary metastases: A case report and review of literature

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ABSTRACT

Renal metastasis of osteosarcoma is a rare entity, with paucity of reported cases in the literature. We report a case of a 20-year-old gentleman who was diagnosed with right distal femur osteosarcoma, complicated with multiple pulmonary recurrences. At two-year-and-a-half interval post-treatment completion, the patient developed right flank pain and frank haematuria. Contrasted abdominal computed tomography revealed a right renal mass with calcification and perinephric haematoma. A right radical nephrectomy was undertaken and histopathological examination showed metastatic condroblastic osteosarcoma. A literature review on renal metastasis secondary to osteosarcoma was performed and we present a report and discussion of these cases.

1. Introduction

Osteosarcoma is the most common primary bone tumour, representing more than half of malignant bone tumours diagnosed among adolescents.[1] Renal metastases were found in autopsies of one-tenth of patients diagnosed with metastatic osteosarcoma, but were rarely diagnosed premortem because they were often clinically silent.[2] Literature review revealed 23 such cases reported worldwide. In this paper, we describe a case of renal metastasis of osteosarcoma and review reported cases.

2. Case presentation

A 17-year-old boy who presented with right knee pain and swelling was diagnosed with localized right distal femur osteosarcoma in March 2018. He underwent neo-adjuvant chemotherapy (high dose methotrexate + cisplatin + doxorubicin) as per the COG-CCG 7921 protocol. This was followed by wide local resection of the tumour with endoprosthetic reconstruction. The resected tissue showed poor tumour necrosis of 60% and lymphovascular permeation. A clean yet narrow lateral margin of 0.1cm from the tumour cells was achieved. Adjuvant chemotherapy was administered, and treatment was completed in January 2019.

In March 2020, he developed multiple lung nodules in the left upper and lower lobes. This was managed with a left thoracotomy and metastasectomy a month later. Computed tomography (CT) thorax done eight months after the surgery revealed a new right lower lobe lesion. In January 2021, he underwent a right thoracotomy and removal of right lower lobe nodule. Histopathology showed metastatic osteosarcoma. Bone scan done two months after the second thoracotomy revealed a Technetium-99m(^{99m}Tc)-methylene diphosphate (MDP) avid left upper lobe nodule. He then underwent a third thoracotomy and metastasectomy in March 2021, where the excision was complete. However, it was complicated with left empyema thoracis and a left lower lobectomy was performed.

In September 2021, the patient complained of right flank pain. Contrast-enhanced CT abdomen revealed a right renal mass with calcification and perinephric haematoma (Fig. 1). There was local mass effect onto the adjacent renal pelvis with no hydronephrosis. The patient's renal function test was normal. A month later, he developed frank haematuria.

Due to concerns of bleeding secondary to release of tamponade from the perinephric haematoma, a renal biopsy was not undertaken. However, radiological features of calcification within the mass supported the diagnosis of renal metastasis of osteosarcoma.

Right laparoscopic radical nephrectomy was carried out in

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November 2021. Histopathology of the resected specimen revealed metastatic chondroblastic osteosarcoma (Fig. 2). The patient was discharged without complications on post-operative day 6. At five months follow-up, the patient remains well and continues to be in remission.

3. Discussion

Even with surgery and chemotherapy, relapse occurred in over half of the patients with osteosarcoma.[3] The most reported site of relapse was the lung.[3] Patients who experience relapse had poor prognosis with a median survival of 14 months and a five-year post-recurrence survival rate of 18%.[3] Relapse without surgical intervention reported an astonishing zero survival rate at five years and ten years, while surgery significantly improved five-year and ten-year survival rate to 31% and 22.3% respectively.[4,5] Therefore, surgical resection remains the mainstay treatment in resectable recurrence.[5]

Based on the analysis of the 23 reported cases of metastatic osteosarcoma to the kidney, the mean interval to renal metastasis is 4.7 years from the primary diagnosis (Table 1). However, the longest interval of renal relapse documented by Hallet et al. was 14 years. 7 cases (32%) reported metastasis to kidney as the first site of relapse. However, patients with osteosarcoma tended to have one or more relapses before developing renal metastasis. Lung metastasis made up half of such relapses (n = 11/22, 50%).

In the review of literature, abdominal pain was the most common presentation of renal metastasis (n = 10/22, 45%). More than one-third of patients were asymptomatic (n = 8/22, 36%), where renal metastases were mostly picked up by follow-up bone scan. Other presentations include haematuria (n = 7/22, 32%) and abdominal mass (n = 4/22, 18%).

Slightly less than half of the CT revealed renal mass without calcification (n = 6/13, 46%). Thus, the absence of calcification does not rule out renal metastasis. Bone scan may be useful in the detection of renal metastases as it showed increased renal uptake in almost all cases reported (n = 6/7, 86%). In this case, the CT abdomen identified a renal mass with calcification and with perinephric haematoma. Although renal pelvis was seen compressed by the mass radiographically, there were no signs of hydronephrosis and patient's renal function remained normal. Raby et al. described a case of a renal metastasis with perinephric haematoma which compressed on the kidney, resulting in reduced renal perfusion and acute kidney injury.

Due to rarity of renal metastasis in osteosarcoma, no standard treatment has been established. In the literature, most patients were treated with nephrectomy only (n = 7/19, 37%), and only one patient survived beyond 9 months. The second most-common treatment modality was chemotherapy only (n = 5/19, 26%), with a median survival of 5 months. Three patients underwent nephrectomy followed by chemotherapy (methotrexate + leucovorin + doxorubicin), where two of them survived beyond 20 months. Radiofrequency ablation was done in one patient where the survival was 13 months. Lastly, percutaneous arterial embolization was described in one patient, where he passed away 6 months post-procedure due to worsening pulmonary metastases. Two patients were put on supportive care only as they were deemed unfit for intervention. Both passed away within 40 days after the diagnosis of renal metastasis.

Pneumoperitoneum induced during laparoscopic surgery is an anaesthetic concern for this patient with history of multiple lung surgeries. However, the reduced post-operative respiratory compromise by laparoscopic surgery outweighed the intra-operative anaesthetic risk. Patients with successful laparoscopic surgery also recover faster and have shorter hospital stay.

Angioembolization was offered as the alternative to nephrectomy as this patient has poor respiratory function and high anaesthetic risk. Nieh et al. described angioembolization suitable for poor-risk patients with symptomatic renal metastasis. One-month post-embolization, the reported patient achieved clear urine, had occasional flank pain, and survived up to 6 months.

4. Conclusion

Although rare, renal metastasis may occur in osteosarcoma. Due to the lack of standard guidance, different treatment modalities have been employed. Where possible, radical nephrectomy may be attempted to achieve complete resection.

Consent

Written informed consent was obtained from the patient for the publication of this report.

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Declarations of competing interest

None.

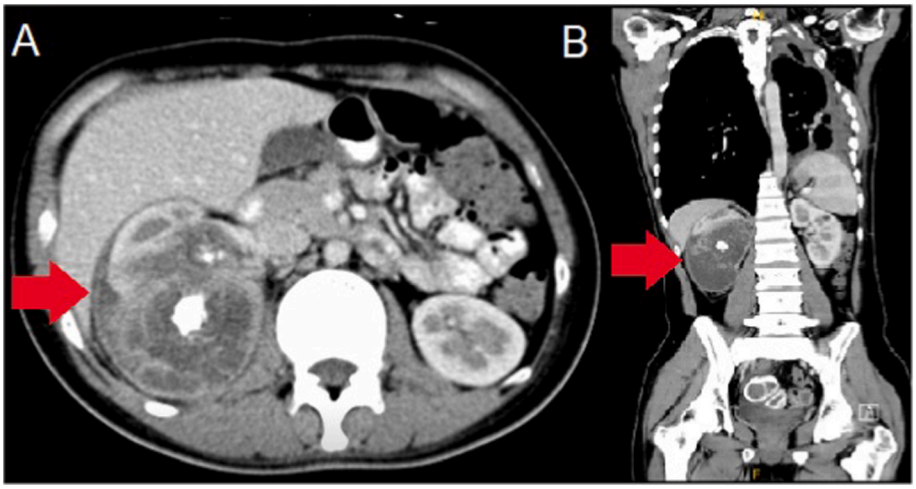

Fig. 1. Contrasted CT scan in axial (A) and coronal (B) plane showing right renal mass with intralesional calcification and perinephric haematoma (red arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

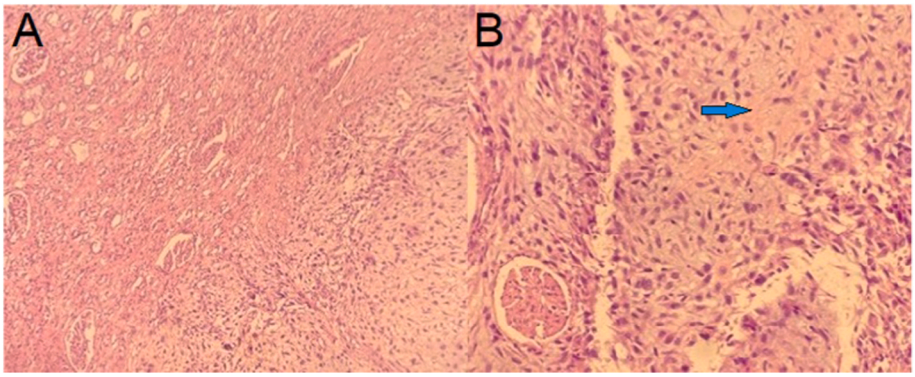

Fig. 2. Histology of the nephrectomy specimen (A) at 10× magnification showing kidney parenchyma (left side) infiltrated by tumour cells embedded within chondroid matrix (right side). (B) at 20× magnification showing the tumour cells are large, exhibiting enlarged and pleomorphic nuclei within moderate eosinophilic cytoplasm, resembling osteoblasts. The background is composed of chondroid matrix and osteoid material (blue arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

Table 1
Literature review of the 23 case reports on secondary renal metastases from osteosarcoma.

No	Study (Year)	Number of relapses before kidney relapse	Site of relapses before kidney relapse	Time from first diagnosis to renal relapse (year)	Presentation	Calcification in CT scan	Bone scan uptake	Treatment	Survival (month)
1	Marshall et al. (1949)	0	Nil	10	Abdominal pain	N/A	N/A	Nephrectomy	9+
2	Nelson et al. (1971)	0	Nil	0.33	N/A	N/A	N/A	Chemotherapy	4
3	Watson & Cubilla (1975)	N/A	N/A	2.5	Abdominal pain	N/A	N/A	N/A	N/A
4	Goldstein et al. (1977)	0	Nil	3	Asymptomatic	N/A	N/A	N/A	3
5	Nieh et al. (1977)	N/A	Lung	2	Abdominal pain, Haematuria	N/A	N/A	Embolization	6
6	Gilbert et al. (1983)	1	Lung	13	Asymptomatic	–	N/A	Nephrectomy	N/A
7	Hallet et al. (1984)	1	Lung	14	Asymptomatic	N/A	+	Nephrectomy + chemotherapy	24+
8	Ayres et al. (1985)	1	Bone	2	Asymptomatic	+	+	Chemotherapy	6
9	Lockhart et al. (1989)	2	Lung	2	Asymptomatic	–	+	Nephrectomy + chemotherapy	20+
10	Barnes et al. (1990)	1	Bone	4	Haematuria	N/A	N/A	Nephrectomy + chemotherapy	N/A
11	King et al. (1992)	2	Bone, lung	5	Haematuria, Abdominal mass	–	N/A	N/A	N/A
12	Kajikawa et al. (1993)	1	Bone	3	Asymptomatic	N/A	+	Nephrectomy	N/A
13	Balingit et al. (1994)	2	Lung	3	Asymptomatic	N/A	+	Nephrectomy	N/A
14	Williams et al. (1995)	1	Bone, lung	4	Abdominal pain	–	N/A	Nephrectomy	N/A
15	Raby et al. (1996)	1	Lung	3	Abdominal pain	–	–	Nephrectomy	0.5+
16	Ogose et al. (1999)	0	Nil	2.5	Haematuria	+	+	N/A	5
17	Karabulut et al. (2002)	0	Nil	0	Abdominal mass	+	N/A	Chemotherapy	6
18	Sakamoto et al. (2006)	4	Lung	12	Abdominal pain, Haematuria	+	N/A	No treatment	0.5
19	Marec-Bérard et al. (2008)	3	Lung	5	Abdominal pain, Abdominal mass	+	N/A	Chemotherapy	2
20	Yu & Yao (2009)	0	Nil	0.17	Asymptomatic	–	N/A	Radiofrequency thermal ablation	13
21	Vyas et al. (2010)	1	Bone	11	Haematuria, Abdominal pain	+	N/A	No treatment	1
22	Akasbi et al. (2012)	0	Nil	6	Haematuria, Abdominal pain, Abdominal mass	N/A	N/A	Chemotherapy	N/A
23	Sigdel et al. (2021)	1	Lung	1.5	Abdominal pain	+	N/A	Nephrectomy	N/A

N/A: not available.

Authors' contributions

Reuben Yih Khai Ooi: Conceptualization, Writing - Original Draft. **Syaza Ab Rahman:** Writing – Review & Editing. **Yen Fa Toh:** Investigation, Writing – Review & Editing. **Ahmad Nazran Fadzli:** Writing – Review & Editing. **Teng Aik Ong:** Supervision, Writing – Review & Editing.

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