10.5152/turkjnephrol.2023.2247320

Case Report

Turkish Journal of Nephrology

Confusing Tumoral Lesions in the Chest Wall of a Deceased-Donor Transplant Candidate

Ahmet Murt[1], Mehmet Eliçevik[2], Nil Çomunoglu[3], Salih Pekmezci[2], Nurhan Seyahi[1], Sinan Trabulus[1]

[1]Department of Internal Medicine, İstanbul University-Cerrahpaşa, Cerrahpaşa Medical Faculty, İstanbul, Türkiye
[2]Department of General Surgery, İstanbul University-Cerrahpaşa, Cerrahpaşa Medical Faculty, İstanbul, Türkiye
[3]Department of Pathology, İstanbul University-Cerrahpaşa, Cerrahpaşa Medical Faculty, İstanbul, Türkiye.

ABSTRACT

Patient selection and preparation for kidney transplantation may pose clinical challenges. A 35-year-old male patient was presented with chest wall tumoral lesions on the day when he was matched with a deceased-donor kidney. Apart from the tumoral lesions, his medical, surgical, and immunologic evaluations were quite favorable to proceed with the operation. Pre-operative thorax surgery and orthopedics consultations were asked for cytopathological evaluation to rule out a possible malignancy. However, considering the cold ischemia time of the donated kidney, there was not enough time to plan and wait for the pathological evaluation of the biopsy. On chest tomography imaging, the expansile tumoral lesions had a lytic–cystic character with well-defined borders. High parathyroid hormone and alkaline phosphatase levels of the patient suggested Brown tumor the most likely diagnosis and we chose to proceed with the operation. Post-operative biopsy findings supported our clinical diagnosis. We herein share the role of clinical evaluation and imaging studies for differential diagnosis of tumoral lesions in kidney transplantation candidates.

Keywords: Brown tumor, kidney transplantation, renal osteodystrophy, kidney transplantation, Brown tumor

Corresponding author: Sinan Trabulus ✉ sinantrabulus@gmail.com

Received: March 17, 2022 **Accepted:** February 25, 2023
Publication Date: July 3, 2023

Cite this article as: Murt A, Elicevik M, Çomunoglu N, Pekmezci S, Seyahi N, Trabulus S. Confusing tumoral lesions in the chest wall of a deceased-donor transplant candidate. *Turk J Nephrol.* 2023;32(3):260-263.

INTRODUCTION

While kidney transplantation is the best form of kidney replacement therapy, patient selection is of paramount importance for its success. A countrywide centralized system is used in Turkey for proper allocation of donated organs. Even with a scoring system that automatically matches donated kidneys to eligible patients, instant clinical evaluation before the operation can still be challenging. Here, we present a transplantation candidate, who had a chest wall tumoral lesion on the day when he was matched with a donated deceased kidney.

CASE REPORT

A deceased-donor kidney was matched to a 35-year-old male patient. He had nephrotic syndrome in childhood that eventually resulted in kidney failure. He was attending dialysis sessions in another center for 15 years and was registered in the transplantation waiting list of our center 4 years ago. He has not attended our regular outpatient visits since then. He had congenital syndactyly involving his third- and fourth-hand digits bilaterally. On the day of transplantation, his blood pressure was 140/90 mmHg and oxygen saturation on room air was 99%. He had bilateral clear lung sounds; he did not have S3 or any cardiac murmurs. His chest looked asymmetrical due to irregularities caused by unspecified painless swellings. He had never smoked. His surgical history included a series of operations for parathyroid adenomas and ectopic parathyroid tissues. Thyroidectomy and thymectomy were also performed as part of these operations. He was on levothyroxine,

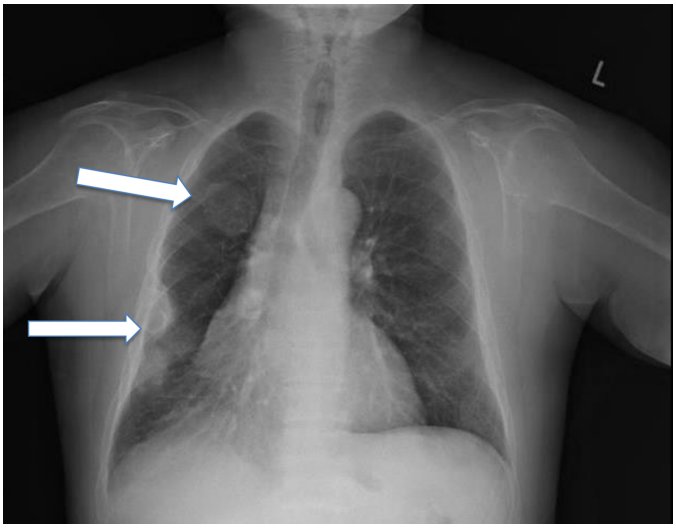

Figure 1. Multiple opacities on chest x-ray.

lansoprazole, sodium bicarbonate, calcium carbonate, and cinacalcet.

We have seen multiple opacities on pre-operative chest x-ray (Figure 1). Chest computed tomography (CT) showed heterogeneous lytic bone lesions in the anterolateral aspect of the right-sided second and fifth ribs (Figures 2-4). Among his laboratory values on the day of transplantation, parathyroid hormone (PTH) was 2411 pg/mL and alkaline phosphatase (ALP) was 1356 U/L. Serum calcium and phosphorus levels were 9 mg/dL and 5.34 mg/dL, respectively. Based on his past medical history and laboratory values, we thought that the lesions were related to mineral bone disease of chronic kidney disease (CKD). Brown tumor was the most likely clinical diagnosis. Pre-operative thorax surgery and orthopedics consultations recommended a biopsy to rule out malignancy. However, considering the cold ischemia time of the donated kidney, there was not enough time to plan and wait for the pathological evaluation of the biopsy.

MAIN POINTS

- Patients in the kidney transplantation waiting list may have different comorbidities.
- Instant clinical evaluation on the day of kidney transplantation may be challenging for patients who are matched with a deceased donor.
- To avoid facing complex clinical conditions on the day of transplantation, patients in the transplantation waiting lists should be evaluated at regular intervals.
- Lytic tumoral lesions with well-defined non-sclerotic borders are most probably Brown tumors if the patient also has high levels of parathyroid hormone and alkaline phosphatase levels.

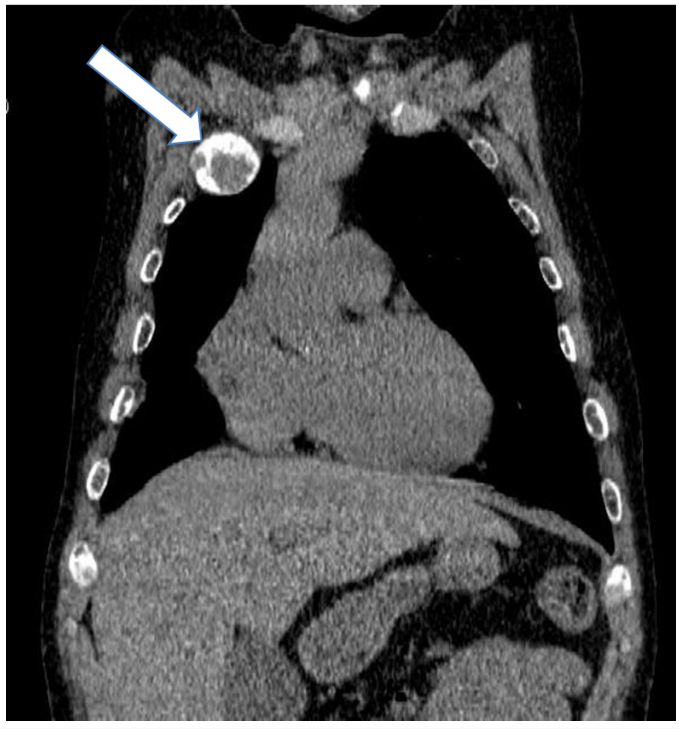

Figure 2. Expansile lytic lesion of the right-sided second rib.

The patient had successful kidney transplantation and is doing well with a functional graft after 1 year. The fine needle aspiration and tru-cut biopsies of the tumoral lesion were done post-operatively, and light microscopy revealed giant cells with interstitial hemorrhage (Figure 5). When evaluated with

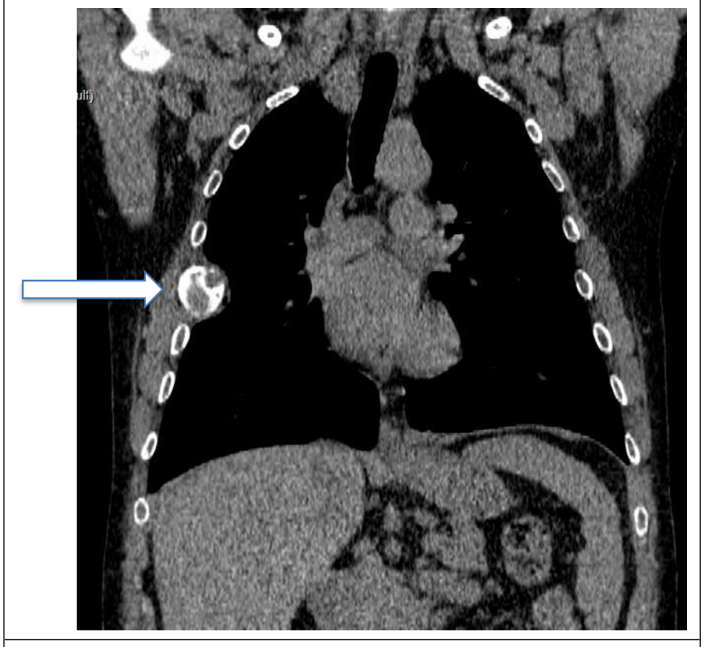

Figure 3. Lytic bone lesion with well-defined non-sclerotic border in the right fifth rib.

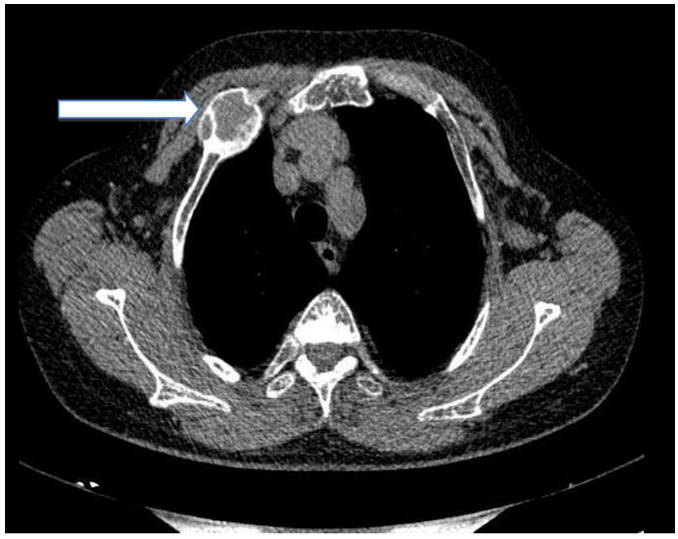

Figure 4. Lytic–cystic bone lesion with expansile character.

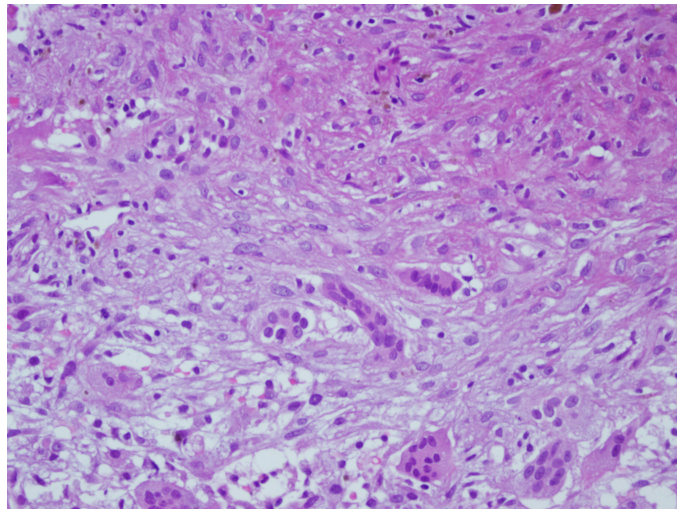

Figure 5. Numerous giant cells with interstitial hemorrhage, hemosiderin, vascularized fibrous tissue with fibroblasts (hematoxylin and eosin ×400).

the clinical findings, biopsy findings were consistent with the Brown tumor. The ALP level that was as high as 1356 U/L preoperatively gradually declined to 180 U/L. His PTH levels also declined from 2411 pg/mL to 981 pg/mL. The control chest CT after 9 months postoperatively showed persistence of lesions with similar size. A subsequent upward trend in PTH (1930 pg/mL) levels necessitated Tc99m sestamibi scan, and it showed ectopic parathyroid tissue near the arcus aorta.

DISCUSSION/CONCLUSION

Brown tumors are erosive bone lesions with increased osteoclastic activity. As a component of osteitis fibrosa cystica, they are consequences of high bone turnover of CKD-related hyperparathyroidism. They can occur in the jaw, skull, mandible, clavicles, pelvis, ribs, spine, and extremities.[1] A biopsy may shed light on the diagnosis, but histologic findings may not fully differentiate brown tumors from giant cell tumors, aneurysmal bone cysts, and osteosarcomas.[2] Thus, clinical evaluation and imaging studies should be utilized to diagnose tumoral bone lesions in CKD patients. Encountering the tumoral lesions on the day of transplantation was quite challenging. They might have been identified previously; however, the patient was receiving his maintenance hemodialysis in another center and was not attending our routine checks. This is another drawback for proper preparation of patients for transplantation and regular health checks of patients should be secured with an inter-institutional approach.

We might have decided to do a biopsy for the costal lesions and cancel the operation. In that circumstance, the transplantation coordination system might have matched this donated kidney to other suitable candidates. However, preparations for another candidate would mean additional time loss. Besides, this particular patient should not have missed the transplantation opportunity, which could also cure his Brown tumors. Post-operative cytopathology evaluation confirmed our preoperative clinical diagnosis. Contrary to our decision, KDIGO clinical practice guidelines on the evaluation and management of candidates for kidney transplantation recommend postponing transplantation until patients are treated adequately for hyperparathyroidism.[3] However, this patient was already parathyroidectomized and there was no further treatment he could receive. On the other hand, in line with our clinical decision, the European Renal Best Practice transplantation guideline recommends against refusing a deceased-donor kidney just because of hyperparathyroidism.[4]

If not a Brown tumor, the mass in the chest x-ray of this patient could be a giant cell tumor, fibrous dysplasia, aneurysmal bone cyst, or reparative granuloma as well as osteosarcomas, metastatic lesions, or myeloma.[5] The expansile lytic feature with a well-defined non-sclerotic border of the tumoral lesion pointed out to a Brown tumor as the most likely diagnosis.[6] High phosphorus, PTH, and ALP levels, which are findings of high bone turnover, supported the diagnosis. Additionally, other bone structures seen in the CT scan had extensive heterogeneous lytic lesions, and the consultant radiologist reported a rugger jersey spine appearance, which is also pathognomonic for renal osteodystrophy.[7] Subperiosteal resorption, another finding of high bone turnover, was apparent in hand radiographs (Figure 6). Congenital syndactyly, in this case, is most probably a sporadic finding; however, it may also be a component of a type of acro-renal syndrome as he had otherwise non-defined nephrotic syndrome.[8]

The tumoral lesion in the chest CT of our patient raised the suspicion of a malignancy. However, a malignancy located in the chest was not that expectable in a non-smoker young patient. Different studies on solid tumors of CKD found varying rates of distribution among organ systems; urologic malignancies,

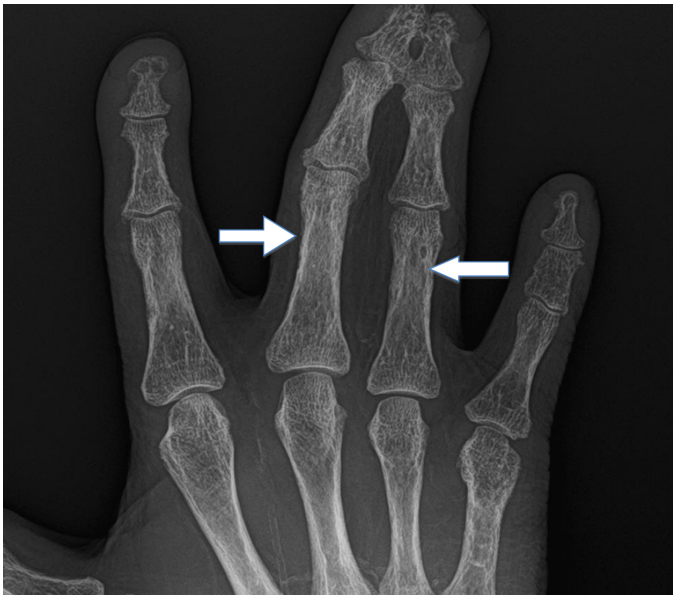

Figure 6. Subperiosteal resorption in the phalangeal bones.

parathyroid adenoma, and skin malignancies being the most common.[9] Lung or chest wall tumors are not among those, which have higher incidences. The average standard incidence ratio for lung cancer in CKD is between 1.1 and 1.4.[10]

Brown tumors may regress after kidney transplantation with a decline in PTH levels. We observed decreasing PTH levels in the early postoperative period. However, PTH levels showed an increasing pattern lately. As the patient had a prior parathyroidectomy, increasing PTH levels were evaluated by a Tc99m sestamibi scan and ectopic parathyroid tissue was detected. Increasing PTH levels in CKD patients with parathyroidectomies should raise the suspicion about ectopic parathyroid tissues.

CONCLUSION

In conclusion, Brown tumors should be the first-line differential diagnosis for tumoral lesions of the bone structure in kidney transplant candidates who have hyperparathyroidism.[11] If needed, biopsies should be planned in a timely manner for patients in the waiting list. Detailed clinical evaluation is indispensable for accurate diagnosis and patients in the waiting lists should be rigorously followed up at regular intervals.

Ethics Committee Approval: The case was presented in accordance with the World Medical Association Declaration of Helsinki. The patient has given informed consent to publish his case including publication of images. All information revealing patient's identity is avoided.

Informed Consent: Written informed consent was obtained from the patient who agreed to take part in the study.

Peer-review: Externally peer-reviewed.

Author Contributions: Consept – A.M., N.S., S.T.; Design – A.M., S.T., N.S.; Supervision- S.P., N.S., M.E.; Resources – A.M., N.Ç.; Materials – N.Ç.; Data Collection and/or Processing – A.M., N.Ç.; Analysis and/or Interpretation – A.M., S.T., N.S.; Literature Search – A.M., S.T.; Writing Manuscript – A.M., S.T.; Critical Review – N.S., S.P., N.Ç, M.E.

Declaration of Interests: The authors have no conflicts of interest to declare.

Funding: The authors declared that this study has received no financial support.

REFERENCES

1. Fatma LB, Barbouch S, Fethi BH, et al. Brown tumors in patients with chronic renal failure and secondary hyperparathyroidism: report of 12 cases. *Saudi J Kidney Dis Transpl*. 2010;21(4):772-777.
2. Bahrami E, Alireza T, Ebrahim H, Mohammadreza S. Maxillary and orbital brown tumor of primary hyperparathyroidism. *Am J Case Rep*. 2012;13:183-186. [CrossRef]
3. Chadban SJ, Ahn C, Axelrod DA, et al. KDIGO clinical practice guideline on the evaluation and management of candidates for kidney transplantation. *Transplantation*. 2020;104(4S1):S11-S103.
4. Abramowicz D, Cochat P, Claas, F, et al. The European Renal Best Practice (ERBP) transplantation guideline development group. *Nephrol Dial Transplant*. 2013;28(suppl 2):ii1-ii71.
5. Vaishya R, Agarwal AK, Singh H, Vijay V. Multiple Brown tumors masquerading as metastatic bone disease. *Cureus*. 2015;7(12):e431. [CrossRef]
6. Wiederkehr M. Brown tumor complicating end stage renal disease. *Clin Nephrol Case Stud*. 2020;8:72-79. [CrossRef]
7. Appelman-Dijkstra NM, Navas Cañete A, Soonawala D. The rugger jersey spine. *Kidney Int*. 2016;90(2):454. [CrossRef]
8. Natarajan G, Jeyachandran D, Subramaniyan B, Thanigachalam D, Rajagopalan A. Congenital anomalies of kidney and hand: a review. *Clin Kidney J*. 2013;6(2):144-149. [CrossRef]
9. Stengel B. Chronic kidney disease and cancer: a troubling connection. *J Nephrol*. 2010;23(3):253-262.
10. Lowrance WT, Ordoñez J, Udaltsova N, Russo P, Go AS. CKD and the risk of incident cancer. *J Am Soc Nephrol*. 2014;25(10):2327-2334. [CrossRef]
11. Diacinti D, Cipriani C, Biamonte F, et al. Imaging technologies in the differential diagnosis and follow-up of brown tumor in primary hyperparathyroidism: case report and review of the literature. *Bone Rep*. 2021;14:100745. [CrossRef]