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Sawab Al Hosni *

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Case Report

Parathyroid Carcinoma in a Young Patient with End-Stage Renal Disease: A Case Report from Oman

Sawab Al Hosni [1], Yahya Al Badaai [2], Abdelhadi Shebl [3]

[1] Oman Medical Specialty Board, Oman

[2] Sultan Qaboos University Hospital, Oman

[3] Sultan Qaboos University Hospital, Oman

***Corresponding author:** Sawab Al Hosni, Oman Medical Specialty Board, Oman

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Abstract

Parathyroid carcinoma is an extremely rare malignancy, accounting for less <0.005% of all cancers and <1% of parathyroid disorders. Pre-operatively, it can be suspected from the tumor size, which usually exceeds 4cm, as well as a high parathyroid hormone (PTH). However, histopathological features confirm the diagnosis. The main objective of treatment is en block resection of the tumor, with excision of the ipsilateral thyroid. We describe a young female with end-stage renal disease (ESRD) who got diagnosed with parathyroid carcinoma. Ultrasound (US) and Sestamibi scans were both suggestive of parathyroid adenoma. Post-operatively, histopathological diagnosis of the specimen revealed parathyroid carcinoma. We introduce this case that was initially suspected to be parathyroid adenoma until histopathological diagnosis revealed parathyroid carcinoma. Patients with ESRD and hypercalcemia with sky-high PTH may have the suspicion for parathyroid carcinoma masked as those lab results can commonly get attributed to the ESRD.

Key Words: parathyroid carcinoma; thyroid carcinoma; endocrine surgery; parathyroid adenoma; parathyroid surgery

Introduction

The first case of parathyroid carcinoma to appear in the literature was in 1909 by De Quervain, in which the diagnosis was attributed to the macroscopic characteristics of the lesion [19]. Today, it is known that parathyroid carcinoma is an extremely rare malignancy, accounting for less <0.005% of all cancers and <1% of parathyroid disorders [13] , [8]. It is most commonly found to be sporadic, but cases have also been reported associating parathyroid carcinoma with familial primary hyperparathyroidism as hyperparathyroidism-jaw syndrome and in multiple endocrine neoplasia type one [3]. Parathyroid carcinoma can be suspected preoperatively from the size of the tumor, which usually exceeds 4 cm, as well as a high level of PTH. However histopathological features confirm the diagnosis [9]. The main objective of treatment is en block resection of the tumor with negative margins. Excising the ipsilateral thyroid lobe may be necessary to perform this, however, it has not been proven to improve survival in patients with parathyroid carcinoma [23]. The aim of this report is to present the management of case that is rarely diagnosed and which might help guide future studies on the same topic.

Case report

A 27-year-old lady was referred from her Nephrologist to the Head and Neck clinic for parathyroidectomy in view of hypercalcemia and sky-high PTH level of around 366 pmol/L. The only clinical complaint she had was diffuse bone pain. Her background medical history also includes ESRD secondary to adult polycystic kidney disease (ADPKD) in which she has been on hemodialysis (HD) for 7 years prior to presentation.

Her physical examination revealed a 2cm lesion in the left thyroid region with no cervical lymphadenopathy.

Laboratories' investigations reveled a sky-high PTH level of 366 pmol/L (normal range 1.6-6.9 pmol/L), a high albumin-adjusted calcium level of 2.64 mmol/L (normal range 2.15-2.55 mmol/L), high alkaline phosphatase of 497 U/L (normal range 35-104 U/L), normal vitamin D of 116 mmol/L (normal range 50-125 nmol/L) and high phosphate of 2.07 mmol/l (normal range 0.81-1.45 mmol/L).

US of the neck showed a well-defined fairly rounded lesion in relation to the lower pole of the left thyroid gland measuring 2cm in diameter and with obvious increased vascularity (Figure 1a). In addition, a well-defined ovoid-shaped hypoechoic nodule, with mild increased vascularity, was seen in the right thyroid lobe measuring about 0.8 x 0.46 cm. There were no suspicious lymph nodes (Figure 1b).

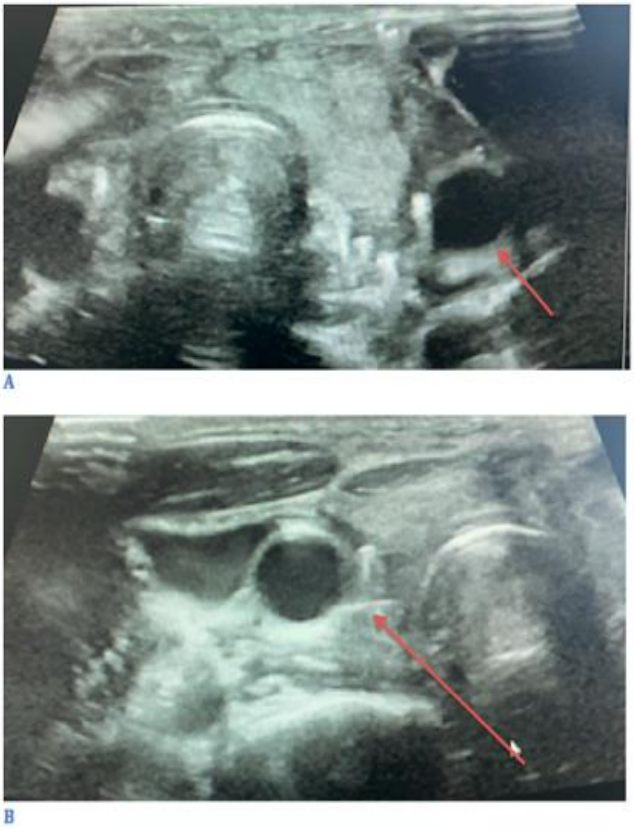

Figure 1

A *technetium 99m Sestamibi scan* was done which showed homogenous uptake by the thyroid glans, with an intense focal uptake in the left lower lobe. The findings were suggestive of a parathyroid adenoma (Figure 2).

Figure 2

In view of the US findings, fine-needle aspiration cytology (FNAC) was performed for the thyroid nodule. Diagnosis was a benign follicular nodule. Bone densitometry scan was also done and revealed the patient's bone mineral density to be below the expected range for her and gender. Intra-operative findings revealed a large left superior parathyroid lesion and prominent remaining parathyroid glands.

She underwent a total parathyroidectomy and a right hemithyroidectomy for the right thyroid hypoechoic nodule. Intraoperative frozen section confirmed the parathyroid tissue from all four glands.

Histopathological diagnosis of the specimens reveled the left superior parathyroid carcinoma (3 x 1.6 x 1.3 cm). Sections prepared revealed encapsulated tumor tissue with a thick capsule. The tumor is composed of

chief cells with some oncocytic and transitional cells. The cells are arranged predominantly in a diffuse pattern. A rim of thyroid tissue is focally seen compressed. There are fibrous bands within the neoplasm with foci of hemosiderin-laden macrophages. Capsular invasion and lymphovascular emboli are identified. There is focal pleomorphism, infrequent mitotic activity, and no tumor necrosis. The neoplastic cells are positive for GATA-3, Cam-5.2, PGP-9.5, and parathyroid H. BCL-2 is weakly expressed. The cells are negative for CDC73. About 5% of tumor cells are Ki67-positive. Resection margins were negative. The rest of the parathyroid glands showed parathyroid glandular tissue with chief cell hyperplasia. The right hemi-thyroid was negative for malignancy (Figure 3).

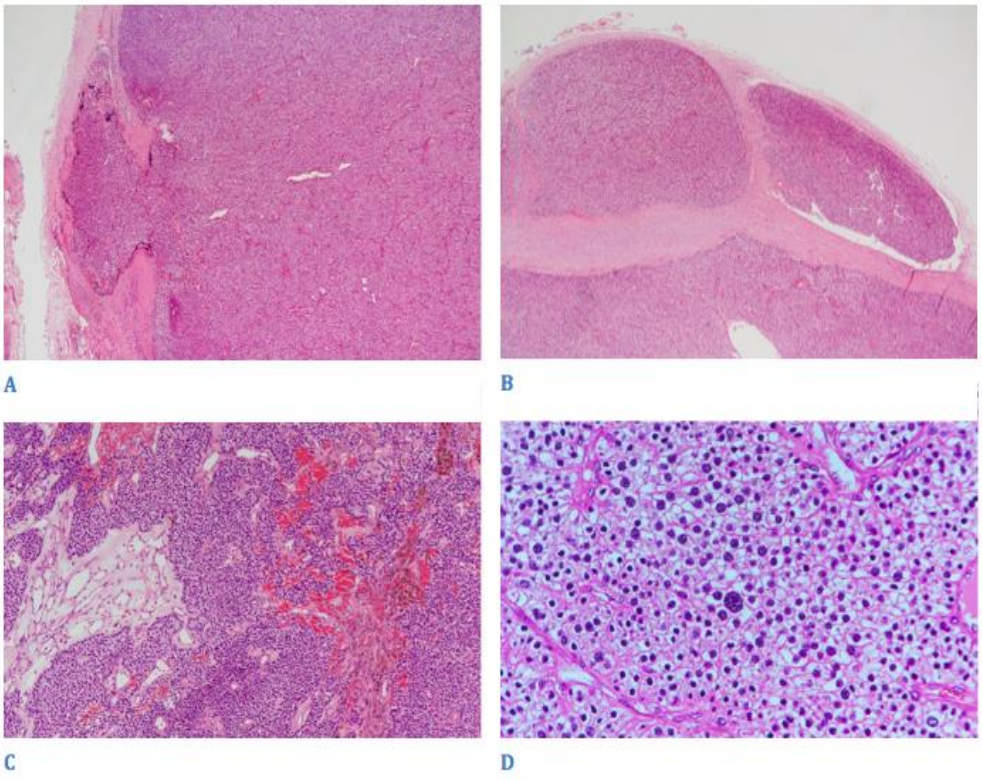

Figure 3

Discussion

Clinical characteristics of patients on hemodialysis who happen to develop parathyroid carcinoma vary widely and have still not been understood completely (Zivalijevic et al. 2021). In the literature, there are 36 cases published to date of patients on hemodialysis who have been diagnosed with parathyroid carcinoma (24; 10, 15); The mean age of those patients at diagnosis is 49.4 years. The age of the patient in our study is 27 years. Up to our knowledge, there are only two cases in the literature of patients in their 20s with ESRD who were diagnosed with parathyroid carcinoma (Tseng et al., 1999 and Srouji et al., 2004). Furthermore, of the 36 cases reported, the majority of them were females (22 cases) (24 and our report is presenting another female case to the literature. This may propose that parathyroid carcinoma in ESRD has a female preponderance. However, further studies are required to claim this finding true.

If parathyroid carcinoma is suspected, dissection of the central neck nodal compartment should be done [17]. Frozen section can be utilized to identify metastasis to lymph nodes, but because it has been associated with a high false-negative rate, it should not overtake clinical judgment. Usually, the recurrent laryngeal nerve can be preserved to maintain its function, however, if the tumor abuts or invades the nerve, it may require resection [5].

Chemotherapy has not been shown to have a role in the treatment of parathyroid carcinoma [7,2]. Some reported studies have shown effectiveness of monotherapy using dacarbazine or combination therapy, while other studies showed no response [16; 1]. The decision to go for adjuvant chemotherapy should be tailored to the individual, with appropriate counseling about the lack of supporting evidence. Likewise, adjuvant radiotherapy has not been shown to be effective in the treatment of parathyroid carcinoma [12]. Similar to adjuvant chemotherapy, the decision to treat with adjuvant radiotherapy should be individualized and discussed in a multidisciplinary team [5].

In the index case, CT staging has shown no local or distant metastasis; therefore, the decision was made to not treat with adjuvant chemotherapy or radiotherapy and keep the patient on close follow up and surveillance.

Conclusion

Parathyroid carcinoma is a rare entity and is challenging to diagnose. Patients with ESRD and hypercalcemia with sky-high PTH may have the suspicion for parathyroid carcinoma masked as those lab results can commonly get attributed to the ESRD itself. Moreover, it is important to note that the young age of the patient should not dismiss the possibility of parathyroid carcinoma.

References

1. Bukowski, R.M., Sheeler, L., Cunningham, J. et al. (1984). Successful combination chemotherapy for metastatic parathyroid carcinoma. *Arch Intern Med.* **144**(2), pp. 399-400.
2. Busaidy, N.L., Jimenez, C., Habra, M.A., et al. (2004). Parathyroid carcinoma: A 22-year experience. *Head Neck.* **26**(8), pp. 716-726.

3. Cetani, F., Frustaci, G., Torregrossa, L., Magno, S., Basolo, F., Campomori, A., Miccoli, P. and Marcocci, C. (2015). A nonfunctioning parathyroid carcinoma misdiagnosed as a follicular thyroid nodule. *World J Surg Oncol.* **13**, pp. 270.
4. Dudney, W.C., Bodenner, D. and Stack, B.C. (2010). Parathyroid carcinoma. *Otolaryngol Clin North Am.* **43**(2), pp. 441-453.
5. Fingeret, A. (2021). Contemporary Evaluation and Management of Parathyroid Carcinoma. *JCO Oncol Pract.* **17**(1), pp.17-21.
6. Fuster, D., Torregrosa, J.V., Esteve, V., et al. (). Parathyroid carcinoma associated to secondary hyperparathyroidism in patients on dialysis: two cases reports. *Nefrologia*. **27**(2), pp. 209–213.
7. Givi, B., Shah, J.P. (2010). Parathyroid carcinoma. *Clin Oncol (R Coll Radiol).* **22**(6), pp. 498-507.
8. Hakaim, A.G. and Esselstyn, C.B. Jr. (1993). Parathyroid carcinoma: 50-year experience at The Cleveland Clinic Foundation. *Cleve Clin J Med*. **60**(4), pp. 331-335.
9. Harari, A., Waring, A., Fernandez-Ranvier, G., Hwang, J. et al. (2011). Parathyroid carcinoma: A 43-year outcome and survival analysis. *J Clin Endocrinol Metab.* **96**(12), pp. 3679-3686.
10. Iwamoto, N., Yamazaki, S., Fukuda, T., et al. 1990. Two cases of parathyroid carcinoma in patients on long-term hemodialysis. *Nephron*. **55**(4), pp. 429–431.
11. Kebebew, E. Parathyroid carcinoma. (2001). *Curr Treat Options Oncol.* **2**, pp. 347-354.
12. Koea, J.B. and Shaw, J.H. (1999). Parathyroid cancer: Biology and management. *Surg Oncol.* **8**(3), pp. 155-165.
13. Lee P.K., Jarosek, S.L., Virnig, B.A., Evasovich, M and Tuttle, T.T. (2007). Trends in the incidence and treatment of parathyroid cancer in the United States. *Cancer.* **109**(9), pp. 1736-1741.
14. Levin, K.E., Galante, M., Clark, O.H. (1987). Parathyroid carcinoma versus parathyroid adenoma in patients with profound hypercalcemia. *Surgery.* **101**(6), pp. 649-660.
15. Rademaker, P., Meijer, S., Oosterhuis, J.W. et al. (1990). Successful surgical treatment of parathyroid carcinoma in two haemodialysis patients. *Nephrol Dial Transplant*. **5**(7), pp. 545–548.
16. Sandelin, K., Auer, G., Bondeson, L., et al. (1992). Prognostic factors in parathyroid cancer: A review of 95 cases. *World J Surg.* **16**(4), pp.724-731.
17. Sandelin, K., Auer, G., Bondeson, L., Grimelius, L. and Farnebo, L.O. (1992). Prognostic factors in parathyroid cancer: A review of 95 cases. *World J Surg.* **16**(4), pp. 724-731.
18. Srouji, I.A., Resouly, A. and Cree, I.A. (2004). Case of thymic parathyroid carcinoma in a haemodialysis patient: application of tumour chemosensitivity testing. *J Laryngol Otol*. **118**(2), pp. 162–164.
19. Sturniolo, G. Gagliano, E. Tonante, A. Taranto, F. Papalia, E. Cascio, R. Damiano, C. and Vermiglio, F. (2013). Parathyroid carcinoma: case report. *G Chir*. **34**(5-6), pp. 170-172.
20. Tominaga, Y., Tsuzuki, T., Matsuoka, S., et al. (2008). Expression of parafibromin in distant metastatic parathyroid tumors in patients with advanced secondary hyperparathyroidism due to chronic kidney disease. *World J Surg*. **32**(5), pp. 815–821.
21. Tseng, C.C., Huang, J.J., Wang, M.C., et al. (1999). Parathyroid carcinoma with multiple lung metastases. *Nephrol Dial Transplant*. **14**(2), pp. 449–451.
22. Wilhelm, S.M., Wang, T.S., Ruan, D.T., Lee, J.A., Asa, S.L. et al. (2016). The American Association of Endocrine Surgeons guidelines for definitive management of primary hyperparathyroidism. *JAMA Surg.* **151**(10), pp. 959-968.
23. Wynne, A. G., van Heerden, J., Carney, J.A., et al. 1992. Parathyroid carcinoma: Clinical and pathologic features in 43 patients. *Medicine (Baltimore).* **71**(4), pp. 197-205.
24. Zivalijevic, V. Zivic, R., Slijepcevic, N. et al. (2021). Parathyroid carcinoma in chronic renal disease - a case series of three patients and review of literature. *Acta Chir Belg*. pp. 1-8.

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