Case Report

SAGE Open Medical Case Reports

Papillary thyroid carcinoma arising from ectopic thyroid tissue in a neck branchial cyst

SAGE Open Medical Case Reports
Volume 11: 1–4

DOI: 10.1177/2050313X231168555
journals.sagepub.com/home/sco
SAGE

Alex Mremi[1,2], Gilbert Nkya[1], Patrick Amsi[1,2], Adnan Sadiq[2,3], Jay Lodhia[2,4] and Angela Pallangyo[1,2]

Abstract

A 35-year-old man presented with a right lateral neck mass for 6 years. Thyroid function test was within normal limits. Computed tomography scan of the neck was suggestive of branchial cyst, tuberculous lymphadenopathy and normal thyroid gland. Fine needle aspiration cytology of cervical lymph node was suggestive of metastatic carcinoma. Branchial cystectomy spearing the thyroid gland was undertaken. Histopathology analysis of the resected specimen confirmed it to be papillary thyroid carcinoma originating from ectopic thyroid tissue in a branchial cyst. The patient was scheduled for total thyroidectomy and neck dissection. Unfortunately, he was lost to follow-up. A brief review of the literature regarding this unusual presentation of thyroid cancer has been provided.

Keywords

Branchial cyst, ectopic thyroid, papillary thyroid carcinoma, Tanzania

Date received: 19 January 2023; accepted: 22 March 2023

Introduction

The vast majority of thyroid cancers arise eutopically within the thyroid gland.[1] Uncommon ectopic sites of origin of thyroid cancer include struma ovarii and other sites that are mostly confined to the neck. These reflect an aberrant embryological origin and location of thyroid tissue.[2,3] We report unusual case of thyroid papillary carcinoma arising in a branchial cyst. A brief review of the literature regarding epidemiology, diagnosis and treatment of this unusual presentation of thyroid cancer has been provided.

A case presentation

A 35-year-old man reported to our facility with a complaint of right neck swelling for 6 years. The swelling was gradually increasing in size but not painful or causing difficulty in swallowing or pressure symptoms. There was no history of prior head and neck irradiation or illnesses. On examination, there was a mass on the right side of the neck measuring 2 cm × 3 cm, which was mobile, fluctuating and non-tender at level 2 inferior. Review of other systems was essentially unremarkable.

Computed tomography (CT) scan of the neck showed two enlarged necrotic cervical lymph nodes in level 2 and 4a (Figure 1).

The largest measured 2 cm × 1 cm in size in level 4a. Other neck structures including both lobes of the thyroid gland were normal. There was no bone destruction seen. The impression tuberculous adenitis was entertained.

Biochemistry reported an elevated serum creatinine level of 118 µmol/L (normal range, 62–106). Thyroid function test was within normal limits. The results from fine needle aspiration cytology (FNAC) of an enlarged cervical lymph node were pointing to a metastatic epithelial neoplasm and thus recommended an excisional biopsy. Patient was planned for surgery, and intraoperatively, there was a mass on the deep cervical fascia, soft, cystic-like containing blood inside and

[1]Department of Pathology, Kilimanjaro Christian Medical Centre, Moshi, Tanzania
[2]Faculty of Medicine, Kilimanjaro Christian Medical University College, Moshi, Tanzania
[3]Department of Radiology, Kilimanjaro Christian Medical Centre, Moshi, Tanzania
[4]Department of General Surgery, Kilimanjaro Christian Medical Centre, Moshi, Tanzania

Corresponding Author:
Alex Mremi, Department of Pathology, Kilimanjaro Christian Medical Centre, 3010 Moshi, Tanzania.
Email: alex.mremi@kcmuco.ac.tz

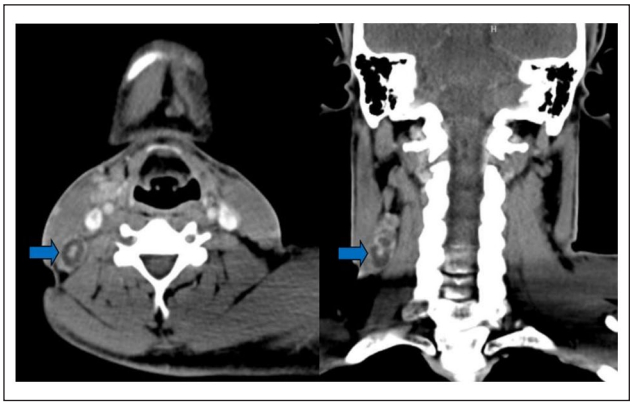

Figure 1. Axial and coronal contrasted CT of the neck shows a well-defined hypodense cystic mass with peripheral and central enhancement arising from the right posterior cervical space measuring 2.0 cm (AP) × 1.4 cm (T) × 3.1 cm (CC) in size. CT findings are consistent with brachial cleft cyst.

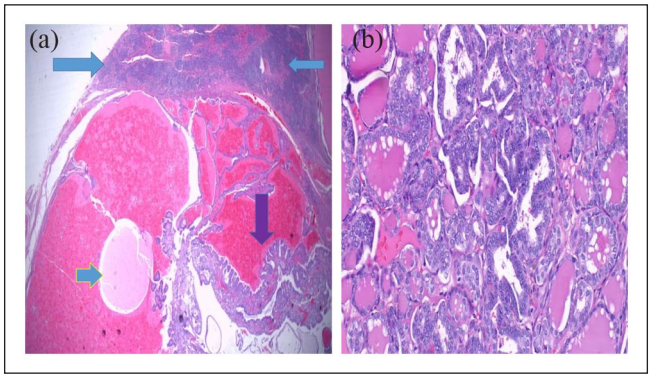

Figure 2. Histopathology of branchial cyst highlighting reactive lymphoid follicles resembling lymph node (upper arrows), mucinous glands (lower left arrow) as well as an area with papillary thyroid carcinoma (lower right arrow); H&E staining 40× original magnification (a); photomicroscopy of thyroid papillary carcinoma displaying characteristic cellular nuclear features characterized by atypia, enlargement, and overlapping with pseudo inclusion and ground glassy appearance; H&E staining 200× original magnification (b).

not vascularized. Excision of the mass was done with the differential diagnosis of branchial cyst.

The specimen was sent for pathological analysis. Grossly, it was an irregular tissue fragment measuring 3 cm × 2 cm × 2 cm. On cut section, it was capsulated and brownish. Microscopically, the sections displayed fibrotic cystic lesion whose wall was lined with stratified squamous epithelium. The presence of reactive lymphoid follicles resembled lymph node and mucinous glands (Figure 2(a)). Additionally, an area with papillary thyroid carcinoma characterized by atypical cells that were enlarged, elongated and overlapping with pseudo inclusion and ground glassy appearance was evident (Figure 2(b)). There was no capsular, vascular or perineural invasion seen. The diagnosis of encapsulated variant of papillary thyroid carcinoma originating from and ectopic thyroid tissue was established. The patient was scheduled for total thyroidectomy and lymph node dissection surgery to rule out occult primary carcinoma of the thyroid. Unfortunately, for unknown reasons, the patient was lost to follow-up.

Discussion

Ectopic thyroid may be defined as functional thyroid tissue outside the normal anatomical location. This condition arises as a result of abnormal migration from the median thyroid anlage by the foramen caecum along the thyroglossoid duct in the fetal stage. All disease conditions of thyroid potentially can arise in ectopic tissue.[3] Carcinoma in an ectopic thyroid tissue is a rare complication.[1] A number of case studies of carcinoma of thyroid arising from an ectopic thyroid tissue have been documented. But thyroid carcinoma originating from ectopically placed thyroid tissue in the branchial cyst is very uncommon.[2] Ectopic thyroid tissue is documented in about 7% of healthy individuals. Mostly it is located along the course of thyroglossal duct or around the two lobes of the gland. Similarly, other unusual anatomical locations such as mediastinum, esophagus, diaphragm, tongue, trachea, larynx and pericardium have been reported.[2]

Previous reports have shown that ectopic thyroid tissue may present as metastasis from thyroid carcinoma, and very rarely it may harbor a primary thyroid carcinoma.[1,2] Of the reported cases, most of them have shown to occur in the thyroglossal duct.[4] To date, only a couple of cases of primary thyroid carcinomas arising in neck branchial cyst have been described.[4,5] Papillary thyroid carcinoma usually spreads to regional lymph nodes, but branchial cyst and cervical lymph node metastasis as the only manifestation of occult papillary thyroid carcinoma is very uncommon.[6,7] It is essential to emphasize that lateral cervical cystic lesions in adults are regularly associated with primary or secondary cancerous disease entities even if the masses have existed for some years. Therefore, a thorough history-taking, head and neck examination, FNA as well as a thyroid imaging could be of great help despite frequently being negative. Thyroidectomy specimen biopsies and excessive sampling are important for diagnosis and surgeons should be prepared for a more radical procedure if a malignant neoplasm is revealed.

Approximately 1%–3% of all ectopic thyroid tissue is located in the lateral neck. The disease may represent the only functional thyroid tissue in the body. Malignant transformation of ectopic thyroid is very uncommon.[8–10] In our patient, we did not perform total thyroidectomy upfront due to the fact that the clinical picture was that of typical branchial cyst with normal thyroid gland. However, after histopathological results, the patient was counseled for

thyroidectomy, but unfortunately he did not turn up. The presence of necrosis found in the cervical node on CT made us think of extra-pulmonary tuberculosis which is endemic in our setting. However, FNAC was suggestive of metastatic disease.

It has been suggested that diagnosis of branchial cysts may be suggested on FNAC and CT or magnetic resonance imaging (MRI). Imaging may reveal certain characteristics associated with malignancy. Findings suggestive of malignancy include dense or enhancing mural nodules, calcification, an irregular margin or a thickened cyst wall.[10] The recommended management approaches and strategies include performance of total thyroidectomy, neck dissection and treatment with radioiodine. However, these should be based on individualized risk assessment.[11] Differential diagnoses for nodal metastasis on cervical region are broad. Typically, this includes malignant tumors that originate at primary sites in the head and neck. These include squamous cell carcinomas of the upper aerodigestive tract as well as metastases from salivary gland, thyroid and cutaneous malignancies. Similarly, primary lymphoma in neck nodes has to be considered as the potential differential diagnosis. In addition, neoplasms from primary sites outside of the head and neck region may unexpectedly metastasize to the neck occasionally.[12,13]

Potential caveat for our case is that it is difficult to confidently establish the diagnosis of malignant thyroid as ectopic since the patient absconded thyroidectomy surgery and thus denied us the opportunity to review thyroid specimen histologically. It is well known that the papillary thyroid carcinoma can last for long time in some less aggressive type without complication, and thus, the index case until now is carrying the risk of primary papillary thyroid carcinoma.[14]

Conclusion

The probability of an ectopic thyroid carcinoma should be taken into account in the differential diagnoses of a pathological mass in the neck including branchial cyst. The uniqueness of the index case strives in the rarity that the thyroid gland was essentially normal on clinical examination, despite cervical lymph node and the ectopic tissue being positive for thyroid carcinoma. Unfortunately, to date there are no robust clinical, biochemical or imaging parameters that can adequately determine the nature of these lesions. Thus, histopathological evaluation always is the mainstay for definitive diagnosis.

Acknowledgements

The authors would like to thank Ms Ruth Moshi, Ummil-Khairat Jaabir Koosa and Daniel Mbwambo for supporting this study as well as the patient for allowing us to use his medical information for academic purpose in this publication.

Author contributions

A.M. and A.P. conceived the study. A.M., A.P., G.N., P.A., A.S. and J.L. participated in data acquisition and were involved in the patient management. J.L. was the lead surgeon. A.S. reviewed radiological images. A.P. and A.M. reviewed histopathology specimen and made histopathology images. A.P. made initial manuscript draft. A.M. critically reviewed and made the final manuscript version. All authors read and approved the final manuscript.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship and/or publication of this article.

Informed consent

Written informed consent was obtained from the patient for his anonymized information to be published in this article. A copy of the consent is available on record.

ORCID iDs

Alex Mremi https://orcid.org/0000-0001-7226-0168
Jay Lodhia https://orcid.org/0000-0002-3373-5762

References

1. Fumarola A, Trimboli P, Cavaliere R, et al. Thyroid papillary carcinoma arising in ectopic thyroid tissue within a neck branchial cyst. *World J Surg Oncol* 2006; 4: 24.
2. Matsumoto K, Watanabe Y and Asano G. Thyroid papillary carcinoma arising in ectopic thyroid tissue within a branchial cleft cyst. *Pathol Int* 1999; 49(5): 444–446.
3. Ibrahim NA and Fadeyibi IO. Ectopic thyroid: etiology, pathology and management. *Hormones* 2011; 10(4): 261–269.
4. Chi HS, Wang LF, Chiang FY, et al. Branchial cleft cyst as the initial impression of a metastatic thyroid papillary carcinoma: two case reports. *Kaohsiung J Med Sci* 2007; 23(12): 634–638.
5. Karras S, Anagnostis P, Noussios G, et al. Thyroid papillary carcinoma arising in ectopic thyroid tissue within a branchial cleft cyst. *BMJ Case Rep* 2013; 2013: bcr2013009312.
6. Gollahalli PK, Chikkannaiah P and Annam V. Primary papillary carcinoma of the thyroid arising in a branchial cyst: case report and review of the literature. *Ear Nose Throat J* 2013; 92(2): E3–E5.
7. Sidhu S, Lioe TF and Clements B. Thyroid papillary carcinoma in lateral neck cyst: missed primary tumour or ectopic thyroid carcinoma within a branchial cyst? *J Laryngol Otol* 2000; 114(9): 716–718.
8. Cappellani A, Di Vita M, Zanghì A, et al. A case of branchial cyst with an ectopic thyroid papillary carcinoma. *Ann Ital Chir* 2004; 75(3): 349–351. Discussion 352.

9. Mehmood RK, Basha SI and Ghareeb E. A case of papillary carcinoma arising in ectopic thyroid tissue within a branchial cyst with neck node metastasis. *Ear Nose Throat J* 2006; 85(10): 675–676.
10. Monchik JM, De Petris G and De Crea C. Occult papillary carcinoma of the thyroid presenting as a cervical cyst. *Surgery* 2001; 129(4): 429–432.
11. Balasubramaniam GS, Stillwell RG and Kennedy JT. Papillary carcinoma arising in ectopic thyroid tissue within a branchial cyst. *Pathology* 1992; 24(3): 214–216.
12. López F, Rodrigo JP, Silver CE, et al. Cervical lymph node metastases from remote primary tumor sites. *Head Neck* 2016; 38(Suppl. 1): E2374–E2385.
13. Gordini L, Podda F, Medas F, et al. Tall cell carcinoma arising in a thyroglossal duct cyst: a case report. *Ann Med Surg* 2015; 4(2): 129–132.
14. Hwang CF, Wu CM, Su CY, et al. A long-standing cystic lymph node metastasis from occult thyroid carcinoma—report of a case. *J Laryngol Otol* 1992; 106(10): 932–934.