Rare disease

Papillary carcinoma of thyroid arising from ectopic thyroid tissue inside branchial cleft cyst: a rare case

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Ectopic thyroid glands generally occur in the midline as a result of abnormal median migration. The presence of these ectopic glands, lateral to the midline is rare. Here, the authors present one case of papillary carcinoma of thyroid arising from an ectopic thyroid tissue in branchial cleft cyst presented as a swelling in lateral neck diagnosed after cystectomy. Total thyroidectomy and neck dissection were done to rule out occult primary carcinoma of thyroid. Histopathology report showed thyroid and lymph nodes were normal.

BACKGROUND

Human thyroid gland derives mainly from one median anlage, which develops from invagination in the floor of the primitive pharynx at the base of the tongue. This anlage during its maturation, migrates downward along the transient thyroglossal duct which undergoes atrophy before the definitive thyroid formation. At the same time, lateral anlagen of two fourth branchial pouches share the development of the gland. From the last two, superior parathyroid glands and the lateral thyroid are derived.¹ A number of anomalies may develop either from the gland or from parts of it during this process. These ectopic tissues may develop the same diseases as the thyroid gland.¹

CASE PRESENTATION

A 34-years-old female patient presented with 2-year history of a painless, non-tender cystic swelling at right side of upper neck at anterior border of right sternocleidomastoid muscle. No associated cervical lymph nodes or other masses were palpable.

INVESTIGATIONS

The high resolution ultrasonography evaluation showed a well-defined cystic lesion with internal echoes in muscle plane below right ear, measuring 3.35×1.38×2.69 cm, volume=6.5 ml (figure 1) and thyroid was normal. Fine needle aspiration cytology (FNAC) examination was suggestive of benign cystic lesion.

DIFFERENTIAL DIAGNOSIS

Branchial cleft cyst, metastatic cystic lesion of cervical lymph node, parasitic cyst.

TREATMENT

Cystectomy was done. The histopathology of resected cystic lesion demonstrated a thyroid papillary carcinoma arising within the neck branchial cleft cyst (figures 2 and

3). Immunohistochemistry staining by thyroglobulin was positive. Total thyroidectomy with right-sided modified radical neck dissection was done to rule out occult primary carcinoma of thyroid. Biopsy report of that specimen was normal. There were no evidence of malignancy in thyroid and lymph nodes even after immunohistochemistry staining.

OUTCOME AND FOLLOW-UP

The patient is in follow-up since last 1 year, there is no recurrence clinically and radiologically.

Figure 1 High resolution ultrasonography neck showing cyst deep to sternocleidomastoid muscle.

Figure 2 Scanner view (H&E 4X) showing lymphoid tissue of branchial cyst (Left arrow) and the papillary carcinoma thyroid on the right upper corner.

Figure 3 Magnified view of figure 1 of right upper showing papillary carcinoma thyroid (H&E X400).

DISCUSSION

Thyroid gland derives from one median anlage at the base of the tongue, and from the two fourth branchial pouches. A number of anomalies may occur during their migration. These can be in form of ectopic tissues, which are frequently found along the course of thyroglossal duct and

rarely in other sites. Many of these may develop same diseases as the thyroid gland.¹ Ectopic thyroid tissue is reported in 7% of adults² and is frequently found along the course of thyroglossal duct or around the two lobes of the gland. Other possible sites of ectopic localisation are anterior tongue, larynx, trachea, oesophagus, mediastinum,

pericardium, diaphragm and rarely neck branchial cyst. Previous reports showed that ectopic thyroid tissue may present metastasis from thyroid carcinoma, and very rarely it may harbour a primary thyroid carcinoma. Of the latter, about 100 cases have been so far described in literature.³ Most of them have been shown to occur in the thyroglossal duct, 1% out of all thyroglossal cysts carcinomas are papillary carcinomas.³ In ectopic thyroid tissue most common type of malignancy is papillary carcinoma of thyroid. It has been reported that primary thyroid papillary carcinoma is found in 64% of cases of occult papillary carcinoma after total thyroidectomy.⁴ Papillary carcinoma of thyroid arising in branchial cleft cyst is very uncommon. Only 10 cases of primary thyroid carcinomas arising in neck branchial cyst have been reported so far.⁵ The present case is the 11th documented case. Papillary carcinoma arising from branchial remnants has the ability to metastasise to regional lymph nodes. Neck node metastases are found in 20%,⁶ but distant metastases are not reported.⁵ In appearance of a lateral neck cyst, the possibility of malignancy of the lesion, the occurrence of a branchial cyst including thyroid ectopic tissue and the presence of a primary thyroid carcinoma arising in this lesion have to be taken into account.⁷ Ultrasonography (USG), FNAC were not able to give diagnostic information in the present case. FNAC may not be diagnostic because of the cystic structure of the lesion. Measurement of thyroglobulin protein and mRNA in needle wash out, although are able to recognise the presence of ectopic thyroid tissue, is not able to discriminate between benign and malignant lesion.⁸ Ultrasound seems to be the most useful diagnostic tool for the study of the thyroid gland and a cystic mass in the neck. Tumours as small as 1 to 2 mm in diameter can be detected with the use of high resolution transducers (7.5 MHz). USG of a malignant cervical mass is useful in confirming the cystic nature of the lesion as well as demonstrating its complex pattern by the presence of solid elements in the cyst wall.⁹ Sidhu's criteria for the papillary carcinoma in branchial cleft cyst⁹ was suggested as (1) an epithelial lining layer, subepithelial lymphoid tissue collection, (2) normal thyroid tissue adjacent to the focus of papillary carcinoma within the wall (3) and no

evidence of papillary carcinoma in the thyroid or other area. In our case, cyst was lined by squamous epithelium and subepithelial lymphoid tissue was present but no normal thyroid tissues were present. It may be justified as ectopic thyroid tissue was limited to part of cyst and whole of remaining tissue was transformed by malignancy. Histopathological examination of total thyroidectomy and lymph nodes specimen showed no evidence of carcinoma even after immunohistochemistry staining.

Learning points

- In a lateral cystic neck mass, although rare, occurrence of ectopic thyroid tissue and presence of a papillary thyroid carcinoma should be kept in mind.

Competing interests None.

Patient consent Obtained.

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