

Thyroid papillary carcinoma arising in ectopic thyroid tissue within a second branchial cleft cyst

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İkinci bir brankiyal yarık kisti içindeki ektopik tiroid dokusundan gelişen tiroid papiller karsinomu

Brankiyal kist, boyun lateralinde en sık gelişen kistik oluşumdur ve sıklıkla yaşamın ikinci ya da üçüncü on yılında görülür. Bununla beraber, bu kist içerisinde ektopik tiroid dokusu nadir bir anomalidir ve bu ektopik dokudan gelişen papiller karsinom oldukça nadirdir. Bilgimize göre, İngilizce literatürde daha önce rapor edilmiş bu hastalığa ait sadece üç olgu mevcuttur. Bu çalışmada, brankiyal kist içerisindeki ektopik tiroid dokusundan gelişen papiller tiroid karsinomlu 34 yaşında bir bayan hasta sunulmuştur.

Anahtar Sözcükler: Ektopik tiroid dokusu, papiller karsinom, brankiyal yarık kisti.

Abstract

Branchial cyst is the most common cyst to arise in the lateral neck, and frequently appears in the second or third decades. However, ectopic thyroid tissue within this cyst is a rare abnormality, and papillary carcinoma arising in this ectopic tissue is extremely rare. According to our knowledge, only three cases of this disease have been reported in English literature previously. A 34-year-old woman with papillary carcinoma arising in ectopic thyroid tissue within a branchial cyst was presented in this paper.

Key Words: Ectopic thyroid tissue, papillary carcinoma, branchial cleft cyst.

Türk Otolarengoloji Arşivi, 2006; 44(2): 103-106

Turk Arch Otolaryngol, 2006; 44(2): 103-106

Introduction

Branchial cyst is the most common cyst to arise in the lateral neck.¹ However, ectopic thyroid tissue within this cyst is a rare abnormality, and papillary carcinoma arising in this tissue is extremely rare.^{2,3} Only three cases of this disease have been reported in English literature previously.⁴

Case Report

A 34-year-old woman presented with a left lateral neck mass that had been slowly growing in size for the last 10 years. Physical examination on initial presentation revealed a 70x20 mm, non-tender, smooth, painless mass deep in the upper two thirds of the left ster-

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nocleidomastoid muscle. The remainder of the physical examination was unremarkable. A magnetic resonance imaging (MRI) scan revealed a 73x18x12 mm cystic mass in the left neck consistent with a branchial cyst (Figure 1).

The preoperative diagnosis was a secondary branchial cleft cyst, and the patient underwent surgery. A multiloculated cystic mass which contained chocolate-brown colored serous fluid was found at the anterior border of sternocleidomastoid muscle, lateral to the great vessels and it was carefully dissected from the surrounding tissues. Total extirpation of the cystic mass was achieved (Figure 2). The post-operative course was uneventful. Histopathologic diagnosis was made as thyroid papillary carcinoma within a branchial cyst wall. It was also reported that, normal thyroid tissue had been identified within the cyst wall adjacent to the tumor on histologic examination (Figure 3). Thyroid ultrasonography and scintigraphy examinations revealed no abnormality. Serial thyroid gland fine-needle aspiration biopsies (FNAB) were also performed, and revealed papillary thyroid carcinoma cells. Because of that

result, total thyroidectomy and left lateral neck dissection was performed. Specimens were pathologically examined. Papillary thyroid carcinoma was detected in thyroid gland, but no lymph node metastasis was revealed on neck dissection material.

Body scintigraphy scanning was performed by 5 mCi I-131 post-operatively, and revealed within the normal limits. 0.1 mgr/day levothyroxin sodium was given to the patient. At the most recent follow-up examination, 6 months after the operation, the patient was in good health and there was no sign of the disease.

Discussion

Ectopic thyroid tissue is commonly found along the thyroglossal duct tract and around the bilateral main lobes of thyroid glands.^{5,6} The most frequent site is the tongue base. Other occurrence reported locations include, anterior tongue, larynx, trachea, esophagus, mediastinum, pericardium, diaphragm, and teratomas of the ovary.^{4,6}

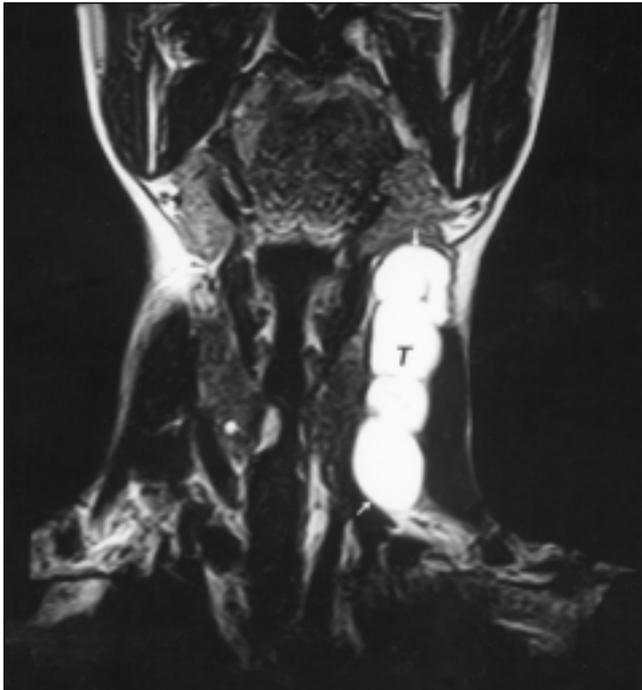


Figure 1a. Coronal MRI scan shows a cystic mass in the left neck.

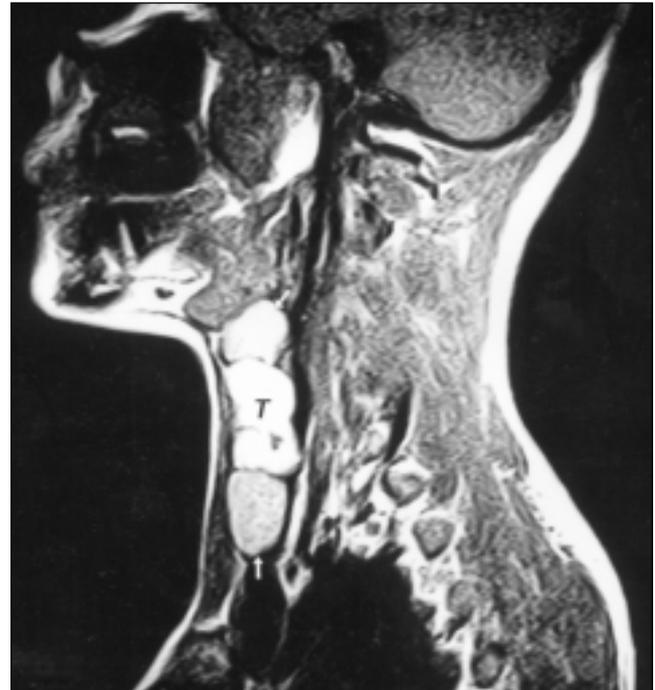


Figure 1b. Sagittal MRI scan shows a cystic mass in the left neck.



Figure 2. Resected cystic mass.

Carcinoma in ectopic thyroid tissue is a rare condition, however over a hundred thyroid carcinoma cases arising in the median ectopic thyroid have been reported.^{7,8} Except medullary carcinoma of parafollicular cell origin, all histological types of carcinoma arising from ectopic thyroid tissue have been reported previously. The most common histological type is papillary thyroid carcinoma, accounting for approximately 85% of cases.^{9,10}

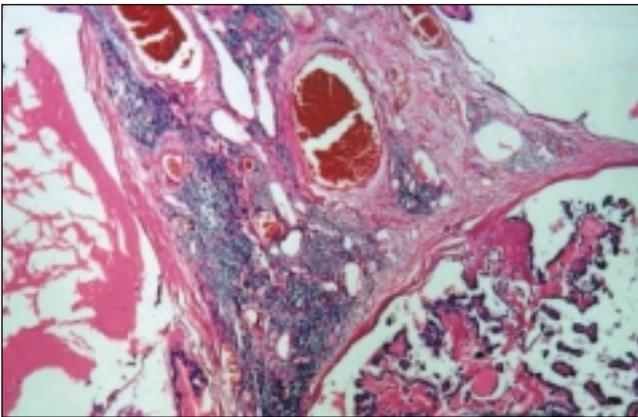


Figure 3. Ectopic thyroid papillary carcinoma within a branchial cyst wall is detected by histopathological examination (HE; Bar=0.5 mm).

Branchial cyst is the most common cyst to arise in the lateral neck, and frequently appears in the second or third decades of life.^{1,11} However, ectopic thyroid tissue within this cyst is a rare abnormality, and papillary carcinoma arising in this tissue is extremely rare.^{2,3} This

is the fourth report of papillary carcinoma case arising in ectopic thyroid tissue within a branchial cyst.⁴ Other three cases were described by Balasubramaniam et. al. in 1992, Jadusingh et. al. in 1996, and Matsumoto et. al. in 1999. There were no lymph node metastases in first two cases reported. However, it was accompanied by lymph node metastasis in the third case.²⁻⁴

Balasubramaniam et. al. reported the first papillary carcinoma case in ectopic thyroid tissue within a branchial cyst. Authors pointed out that normal thyroid tissue had been identified within the cyst wall adjacent to the tumor in histopathological examination. These features strongly supported that this tumor had arisen in ectopic thyroid tissue within a branchial cyst and in addition did not represent a metastasis from a primary thyroid gland carcinoma. Radioisotope scan and ultrasound examination of the thyroid gland had revealed no abnormality.² Matsumoto et. al. reported another case. Different from the previous report, authors pointed out that as it was difficult to morphologically distinguish between the original cancer and metastatic cancer, a hemi-thyroidectomy and modified neck dissection had been performed. It was also reported in that paper that lymph node metastasis was detected in histopathological examination.⁴

In our case, because the mass examined had the typical feature of a branchial cyst, surgery was performed as for routine excision of a branchial cyst. A multiloculated cystic mass which contained chocolate-brown colored serous fluid was found, and total extirpation of the mass was achieved. Histopathologic diagnosis was made as thyroid papillary carcinoma within a branchial cyst. However, normal thyroid tissue was also identified within the cyst wall adjacent to the tumor on histological examination. As in the case reported by Balasubramaniam et. al., this diagnosis strongly led us to conclude that this tumor had developed in ectopic thyroid tissue within a branchial cyst.² Nevertheless, we thought that it could be possible that papillary carcinoma might arise in the thyroid gland at the same time. Thus, thyroid ultrasonography and scintigraphy examinations were carried out and revealed no abnormality.

FNAB is a simple, inexpensive, and rapid technique with minimal complication risk. Lesions in head and neck region are especially appropriate for FNAB, and it

may be repeated until the appropriate specimen is obtained. A suspicious or definitive diagnosis of cancer by FNAB allows the surgeon the opportunity to plan the optimum surgical procedure.¹³ Because of these features of FNAB, serial blind fine-needle aspiration biopsies were performed to exclude the possible occult thyroid tumor. Cytological examination of these revealed papillary thyroid carcinoma. So, total thyroidectomy and left lateral neck dissection were performed.¹² Histopathologically, papillary thyroid carcinoma was detected in thyroid gland, but no lymph node metastasis was revealed on neck dissection material.

The main emphasis of this report is that, if a cancer is found to develop from an ectopic thyroid tissue in a patient, all other possible ectopic tissues, and especially the thyroid gland should be examined in detail. In our case, although thyroid ultrasonography and scintigraphy examinations revealed no abnormality, on cytological examination of fine needle aspiration biopsies occult cancer cells were detected and the type of further surgery to be performed was chosen accordingly.

References

1. **Howie AJ, Proops DW.** The definition of branchial cysts, sinuses and fistulae. *Clin Otolaryngol* 1982; 7: 51-7.
2. **Balasubramaniam GS, Stillwell RG, Kennedy JT.** Papillary carcinoma arising in ectopic thyroid tissue within a branchial cyst. *Pathology* 1992; 24: 214-6.
3. **Jadusingh W, Shah DJ, Shaw H, Lyn C.** Thyroid papillary carcinoma arising in a branchial cleft cyst. *West Indian Med J* 1996; 45: 122-4.
4. **Matsumoto K, Watanabe Y, Asano G.** Thyroid papillary carcinoma arising in ectopic thyroid tissue within a branchial cleft cyst. *Pathology Int* 1999; 49: 444-6.
5. **Batsakis JG, El-Naggar AK, Luna MA.** Pathology consultation: thyroid gland ectopias. *Ann Otol Rhinol Laryngol* 1996; 105: 996-1000.
6. **Rosai J, Carcangiu ML, DeLellis RA.** Tumor of the thyroid gland. In: Atlas of tumor pathology. Washington DC: Armed Forces Institute of Pathology; 1990. p. 317-26.
7. **Grabowska H.** Papillary carcinoma arising in ectopic thyroid gland in the wall of a thyroglossal duct cyst. *Path Res Pract* 1993; 189: 1228-9.
8. **Kum CK, Goh P, Teh M.** Papillary carcinoma arising in a thyroglossal duct cyst. *Aust NZJ Surg* 1993; 63: 738-40.
9. **LiVolsi VA, Perzin KH, Savetsky L.** Carcinoma arising in median ectopic thyroid (including thyroglossal duct tissue). *Cancer* 1974; 34: 1303-15.
10. **Joseph TJ, Komorowski RA.** Thyroglossal duct carcinoma. *Hum Pathol* 1975; 6: 717-29.
11. **McDermott ID, Watters WR.** Metastatic papillary thyroid carcinoma presenting as a typical branchial cyst. *J Laryngol Otol* 1996; 110: 490-2.
12. **Gardner RE, Tuttle RM, Burman KD, et al.** Prognostic importance of vascular invasion in papillary thyroid carcinoma. *Arch Otolaryngol Head Neck Surg* 2000; 126: 309-12.
13. **Yang YJ, Haghiri S, Wanamaker JR, Powers CN.** Diagnosis of papillary carcinoma in a thyroglossal duct cyst by fine needle aspiration biopsy. *Arch Pathol Lab Med* 2000; 124: 139-42.

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