Case Report

PAPILLARY CARCINOMA ARISING IN A THYROGLOSSAL CYST WITH THYROID MICROCARCINOMA NOT SEEN BY SONOGRAPHY

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ABSTRACT
The thyroglossal duct cyst is the most commonly seen one among congenital neck masses. It is resulted from persistence of thyroglossal tract in some parts and to continue secretion, which should have to close during embryological development. Rarely, a thyroglossal duct cyst may exhibit malign transformation. It is an uncommon event to see a concurrent carcinoma in thyroid gland. Papillary thyroid carcinoma is the most commonly seen malignancy. The diagnosis is usually made by histopathological evaluation of the mass excised. In this study, we aimed to present a 60 years old man in whom synchronous papillary carcinoma arising in a thyroglossal cyst and papillary thyroid microcarcinoma were detected. In our study thyroid scan was normal but we detected micro papillary thyroid carcinoma in histopathological evaluation.

INTRODUCTION
A thyroglossal duct cyst (TDC) occurs due to regression failure of the tract which attaches thyroid gland to the root of tongue during descent of thyroid gland to normal localization. It normally regresses at sixth week of life (Ellis, 1977). TDC, the most common congenital neck anomaly, is seen in the 7% of the adults. Malign transformation is observed in 1% of these benign cysts at the fourth decade (Motamed, 2004). It is very rare to see a synchronous papillary thyroid microcarcinoma (PTC). The diagnosis is usually made by histopathologic examination of the excised mass (Kwon, 2012). Papillary thyroid carcinoma is the most common tumor in terms of differentiated thyroid cancer. In the current study, we aimed to present a patient who had a papillary carcinoma arising from TDC with PTC not seen by sonography. To the best of our knowledge, this is the third report in aspect of this association.

CASES
A 60 years old man was admitted to our clinic with neck mass which occasionally appeared within last 3 years and then became permanent within the past year.

Figure 1: Image of the mass on the CT scan
In the physical examination, there was a cystic mass at mid-cervical line which moves with gulping. There was no other finding on the neck examination regarding lymphadenopathy and thyroid nodule. On the neck sonography, a cystic mass (3.0x3.5x3.0 cm in size) consistent with thyroglossal cyst was shown. On the computerized tomography (CT), a lesion (3.0x3.0 cm in size) with irregular contours, septated cystic areas and microcalcifications were detected, and it was consistent with thyroglossal cyst (Figure 1). The patient had surgical operation. In the microscopic examination of surgical material, papillary configurations were detected composed of atypic cells which had strong positivity in thyroglobulin and Cytokeratin 19 (CK 19) staining, whereas positivity in HMBE 1 staining (Figure 2).

According to these findings, the patient was diagnosed as TPC arising from TDC. No nodule was detected in thyroid sonography Therefore, total thyroidectomy and central neck dissections were performed in the patient. Focal areas of papillary microcarcinoma were determined at left lobe of the surgical specimen not seen by sonography, (Figure 3).

**DISCUSSION**

TDC is the mostly seen mass at midcervical region. It has a frequency of 75% in children and 7% in adults (Banerjee, 2007). Embryological development of thyroid gland begins at the fifth week of gestation, and it is completed at the seventh week (Banerjee, 2007). It use thyroglossal duct during migration from pharynx to neck. This duct closes over time by losing its characteristics. Otherwise, TDC develops.

It is usually presented at the third or fourth decades of life, and more common in women than men. It is rarely seen under the age of 14 years. Etiology of malign transformation is unclear but radiation exposure may have a role in this transformation. The tumor usually exhibits a non-aggressive behavior, and it may rarely show a lymphatic involvement (Dedivitis, 2000). TDC accounts for 1% of all thyroid cancers. Malign transformation risk is 0.7-1.0% in TDCs (Banerjee, 2007). The most common malign tumor is papillary carcinoma (80%) among TDCs followed by mixed papillary-follicular carcinoma (8%), squamous cell carcinoma (6%), follicular carcinoma (3%), adenocarcinomas and unidentified tumors (3%) (Weiss, 1991). Our case was a male patient at sixth decade, which had a papillary carcinoma arising from TDC without lymphatic involvement.
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Various theories have been proposed for the development of papillary carcinoma arising in TDC. According to metastatic theory, papillary carcinomas may be multifocal and multicentric and malignancy in the cyst develops through metastasis from the malignancy in thyroid gland (Tew 1995, Forest, 2011). However, some authors advocate that there are simultaneous cancer focuses in both TDC and thyroid gland (Pribitkin, 2002). In the present case, cystic mass had an epithelial wall with papillary configuration in histopathological evaluation. Papillary configurations expressed strong positivity in thyroglobulin and CK19 staining, whereas positivite in HBME staining. Focal areas of micro-papillary carcinoma were detected at left lobe of total thyroidectomy specimen in the histopathological evaluation. Clinical presentation of TDC is similar to papillary carcinoma arising from TDC. However, sudden increase in size, pain, dysphagia and regional lymphadenopathy should be considered as a malignancy (Dedivitis, 2000). Diagnosis of papillary carcinoma is made by histopathologic examination (Banerjee, 2007, Kusunoki, 2007, Heshmati, 1997). Although sonography and CT scan are adequate to make the diagnosis of cyst, they are generally inadequate to diagnose the malignancy (Martín-Pérez, 1997). In the present case, there was a painless lump which exhibited occasional enlargement within last 3 years and the enlargement became progressive within the past year. On the neck sonography, a mass (3.0x3.5x3.0 cm in size) was detected at the mid-cervical line. No nodule in thyroid gland and no lymphadenopathy at neck were detected. Mass was excised by Sistrunk procedure in our case, as there was no clinical finding suggesting malignancy. In the histopathological evaluation of the mass, a papillary carcinoma arising from TDC was detected.

There is no consensus on the surgical treatment of papillary carcinoma arising from TDC due to insufficient number of cases and lack of information regarding long-term follow-up. Some authors suggested that Sistrunk procedure is sufficient in well-differentiated thyroid cancers with well-defined margins by cyst wall and normal findings in thyroid sonography which have no cervical metastasis or local involvement (Tradati, 2000). On the other hand, total thyroidectomy should be added in case of thyroglobulin follow up. This suggestion is based on the assumption that papillary carcinoma arising from TDC is resulted from a metastasis from thyroid gland or high likelihood of presence of a synchronous thyroid microcarcinoma (LaRouere, 1987). TPC has been found in 11-27% of total thyroidectomy specimens excised from the cases with papillary carcinoma (Mesolella, 2010). In our case, no nodule was determined on thyroid sonography. However, focal areas of micropapillary carcinoma were seen at the total thyroidectomy specimen.

CONCLUSION

Although TDC is a benign disorder, malign transformation may be seen in 1% of TDCs. Thyroid microcarcinoma very rarely occurs in association with a concurrent papillary carcinoma arising from TDC. As in our case, we suggest that it is appropriate to perform a total thyroidectomy due to difficulties in the detection by sonography. Therefore, we think that an individualized treatment plan should be considered in each patient. Total thyroidectomy, radioactive iodine and hormone replacement therapies should be added, when needed. Here, we present a patient who had a papillary carcinoma arising from TDC with PTC not seen by sonography. To the best our knowledge, this the third report in case of this concurrence.

REFERENCES


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