INTRODUCTION

Thyroglossal cysts are the most common congenital cystic lesions of the neck related to embryological development of the thyroid gland. Sistrunk (7) in 1920, first described a method of excising these cysts including the removal of central core of the hyoid bone and tongue up to foramen caecum. Uchermann (8) first described in 1910, a malignant neoplasm arising in this structure. Since then, less than 100 cases have been described in literature. There has not been a case previously of this condition from Turkey.

CASE REPORT

A 32-year-old woman was admitted to the hospital with a history of nausea, tremor and approximately 5 kg of weight loss within the last 6 months.

Physical examination was not remarkable except for a mid-line mass with a diameter of 1.5x1.5 cm in the neck, 1 cm above the thyroid cartilage. The mass was mobile and painless.

Routine laboratory tests were within normal limits except for a mild iron deficiency anemia. Thyroid hormone levels were normal. Thyroid scanning showed minimal hyperplasia in the left lobe with a hyperactive nodule formation in the distal half whereas the right lobe was not visualized.

The mass was excised by the ENT Department with the diagnosis of thyroglossal cyst and Sistrunk type procedure was done. Pathological examination revealed the mass to be a papillary thyroid carcinoma with hyoid bone invasion, without any evidence of thyroglossal cyst tissue. The patient was referred to the General Surgery Department.

Physical examination, on admission to our department showed nothing remarkable except for a 6 cm cervical incision scar of the previous Sistrunk operation. But, about in a weeks time, a left submental, painless and partially mobile lymph node, 0.5 cm in diameter occurred.

Routine laboratory tests were normal.
Thyroid functions were diminished.

\[
\begin{align*}
T_3 & 0.21 \text{ ng/ml (N: 0.52-1.75)} \\
T_4 & 0.81 \mu \text{g/ml (N: 4.80-12.80)} \\
TSH & 54.40 \mu \text{U/ml (N: up to 5.0)}
\end{align*}
\]

Ultrasonographic examination of the neck showed a remarkable decrease in the volume of the thyroid gland and the dimensions were measured 19x8x7 mm on the left and 5x6x1 mm on the right lobe.

Whole body scanning with 2 mCi $^{131}$I showed no other pathological accumulation except for a 1x1.5 cm area just above the thyroid region, identical to submental lymph node.

According to physical and laboratory findings, a cervi-
cal exploration was planned for papillary thyroid carcinoma arising in an ectopic thyroid tissue. On the exploration, neither a normally localized thyroid tissue nor any other lymph node was found except from the submental one. Therefore only a suprahoid lymphatic dissection was performed. Pathological examination showed no malignancy and the lymph node was told to be reactionary.

The patient was given 100 mg L-thyroxine two times a day. Thyroid function re-examined on the twentieth day postoperatively were in normal limits. Whole body scanning with 2 mCi $^{131}$I on the first and the sixth months revealed nothing pathological.

COMMENTS

The anlage of thyroid gland migrates from the region of foramen caecum of the tongue and descends to its normal anatomic position. It passes anterior to the developing hyoid bone, however it has been found to be in close proximity to the posterior aspect of this bone as well (2). Generally in the sixth to eighth week embryo, the tract is obliterated (1). However accessory thyroid tissue and/or cysts can arise anywhere along this tract and on rare occasions, such as in our case, the patients only functioning thyroid tissue can be found along this pathway (4). In our case, after the removal of the remainder of functioning thyroid tissue following the Sistrunk procedure, despite the US and the scanning, there was no source of thyroid hormones. Postoperative thyroid function tests also confirm this fact.

Papillary adenocarcinoma has been presented in 87% of the malignancies arising from the thyroglossal cysts. The incidence of thyroid tissue within thyroglossal duct remnants has varied anywhere between 5 to 75% depending on the pathological details. However in our case there was no evidence of thyroglossal cyst remnant in the specimen. Such cases are extremely rare in the literature.

Postoperatively, the patient received suppressive doses of thyroid medication as recommended. Since there was no pathological finding in whole body scans, no other therapeutic modality was planned and she is being followed for recurrence or metastases and up to date no evidence is found.

REFERENCES


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