Turkish Journal of Endocrinology and Metabolism, (2004) 2 : 71-73 **CASE REPORT**

Inflamed Multiple Keratinous Nodules in a Thyroglossal Cyst

Nihat Kaymakçıoğlu* **Mutlu Yakut*** **Mehmet Fatih Can**** **Salih Deveci**** **Turgut Tufan***

Gülhane Military Medical Academy, Ankara
** General Surgery Department*
*** Pathology Department*

The authors report a case of an unusual inflammatory reaction and multiple Keratinous nodules in a thyroglossal cyst. It consisted of broad papillary intraluminal projections and multiple keratinous nodules. This benign process should not be confused with a carcinoma, branchial cyst and dermoid cyst.

Key words: Thyroglossal cyst, inflammation, keratinous nodules

Introduction

A thyroglossal cyst is a congenital anomaly resulting from retention of an epithelial tract between the thyroid and its origin, the foramen caecum, in the floor of the pharynx (1,2).

It usually presents as a painless swelling in the midline of the neck, below the thyroid bone and may become painful increasing rapidly in size, in association with infection (2,3). It is often complicated by infection occasionally by fistula, and rarely by carcinoma (4).

Histologically, it is lined by pseudo stratified ciliated columnar epithelium and/or squamous epithelium. Accumulations of chronic inflammatory cells, mucous glands and thyroid follicles are commonly seen in the subjacent stroma (1,5,6).

Case Report

A 20 year old man presented with a three month history of slowly enlarging mass in the anterior midline aspect of the neck. There was no history of previous irradiation, trauma, infection drainage or erythema. According to the patient the mass had been present for more than 15 years and apart from several episodes of minor pain and redness of the overlying skin, was asymptomatic. On examination, a firm, cystic nodule 5x4x4 cm, was present in the anterior aspect of the neck, in the midline at the level of the incisura jugularis. It was freely mobile and moved upward with either swallowing or protrusion of the tongue. The remainder of his head and neck examination was unremarkable. Ultrasonography showed multiple nodularities in the cyst (Figure 1).

The cystic nodule was surgically excised. It was cystic containing about 10 ml of cloudy fluid. In microbiologic examination coagulase negative staphylococcus was founded in this fluid. There were multiple keratinous structured in a view of the possibility of pyogenic inflammation (Figure 2). Histologically, the cyst was partially lined by ciliated columnar epithelium and partially by squamous epithelium. The wall of the cyst was thickened and consisted of fibrous and granulation tissue, with focal mononuclear cell infiltration and multifocal thyroid tissues. In a part of the cyst, the epithelium was replaced by a branching papillary proliferation projecting into the lumen (Figure 3). Keratinous structures typically showed in the cyst anucleate kreatinising and squamous epithelial cells of variable maturity in background of debris (Figure 4).

Correspondence address:

Dr. Nihat Kaymakçıoğlu
Gülhane Military Medical Academy, Department of General Surgery 06018 Etlik/ANKARA
e-mail : mfcan@superonline.com
Tel : (0 312) 304 51 12-304 51 10

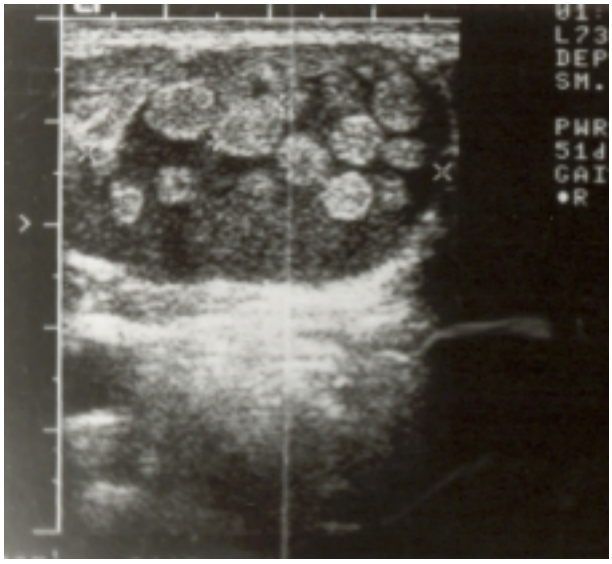

Figure 1. Ultrasonography showed multiple nodularities in the cyst.

Figure 2. Macroscopic Histological exam showed multiple anuclear keratinous nodularities into the cyst.

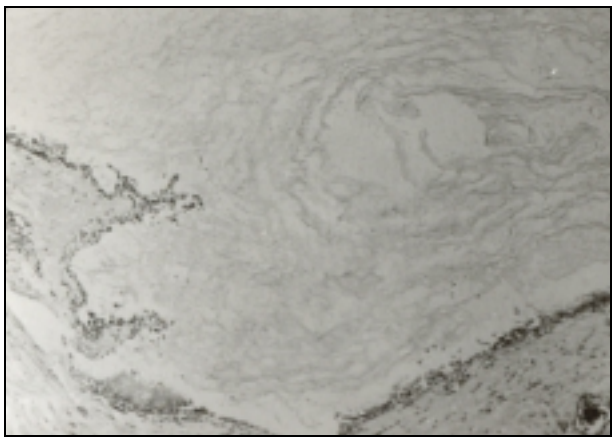

Figure 3. The epithelium was replaced by a branching papillary proliferation projecting into the cyst and keratinous material in the luminal part (Hematoxylin and eosin x100).

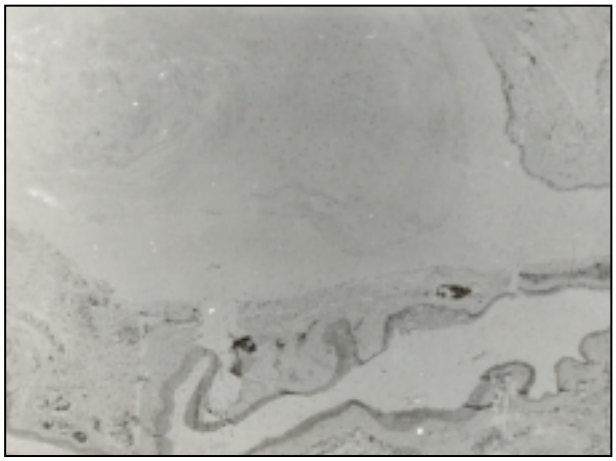

Figure 4. The keratinous structures typically showed anucleate keratinizing, squamous epithelial cells and debris (Hematoxylin and eosin x 50).

Discussion

Among benign cervical masses, thyroglossal duct cyst is one of the most frequently encountered. In childhood, only benign cervical lymphadenopathy is seen more frequently (7). The thyroid gland originates at the foramen caecum, located on the anterior floor of the pharyngeal gut. The thyroid gland is first seen about the 17th day of development as a proliferation of epithelium that quickly penetrates the underlying mesoderm, enlarges, and descends in front of the pharynx as a bilobed diverticulum remains patent and is known as the thyroglossal duct. By the seventh week the thyroid has taken its final position in front of the trachea and the eighth to tenth week the thyroglossal duct disappears (8).

A thyroglossal cyst does not usually present a diagnostic problem to the pathologist. The removed specimen usually consists of a single primary ductal structure connected to a cyst and passing up to the foramen caecum in intimate association with the hyoid bone. The cyst contains a colorless viscous secretion. The ducts are lined by stratified squamous epithelium or ciliated pseudo stratified columnar epithelium. Mucus-secreting glands also may be seen in association with them. It is of interest that the epithelial lining of single specimens has been described as both squamous and columnar in adjacent parts (9). Detailed histological examination is essential not only to establish the diagnosis of a thyroglossal cyst, but also to exclude one serious complication, namely carcinoma (1,3,10).

The high recurrence rate (%20) in some reported series (11) is due primarily to failure of the surgeon to perform the sistrunk operation removing cyst, hyoid bone and proximal duct as a core through the base of the tongue to the foramen caecum (12). The operation is performed under endotracheal anesthesia with the patients neck extended. A transverse skin incision is made directly over the cyst in the infrahyoid region. The tract is then dissected up to the hyoid bone. The muscular attachments to the superior and inferior aspects of the body of the hyoid bone are divided. The duct is ligated and divided at the base of the tongue. The hyoid bone need not to be reapproximated. A small penrose drain may be inserted for 24 hours, particularly if there is a history of past infection obviously indicated is excision of the lesion by a sistrunk procedure and removal of involved lymph nodes. Most of the reported cases have not included thyroidectomy (9).

This benign process should not be confused with a true papillary neoplasm which is a rare but well documented complication of a thyroglossal cyst (10).

The differential diagnosis in the present case would include colloid nodule with cystic degenerating, branchial and dermoid cyst. Their differentiation is based mainly on the anatomic location of the lesion; branchial cyst is laterally located, while thyroglossal cyst is situated in the midline as in our case, moves upward with either swallowing or protrusion of the tongue (13). Though dermoid cyst in the neck is also located in the midline, a fine needle aspiration sample of it usually consist of thick, greasy material, distinctly different from that of thyroglossal cyst (14). We report a case of an unusual inflammatory reaction and multiple keratinous nodules in a thyroglossal cyst.

A thyroglossal cyst usually contains any or little cholesterol (15). The major factor is tissue breakdown due to chronic or recurrent infection and inflammation in our case, the nodular lesions in the cyst typically showed anucleate keratinization and squamous cells, debris and polymorphs. In our patient the episodes of minor pain associated with erythema of overlying skin, most probably represented recurrent attacks of infection and inflamemation which might lead to the formation and accumulation of such a large amount of keratinized debris in this long-standing thyroglossal cyst.

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