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Thyroglossal cyst carcinoma

by

S. KRISTENSEN,* A. JUUL,* and J. MOESNER† (Esbjerg, Denmark)

Introduction

A thyroglossal duct cyst is a congenital anomaly often manifesting itself as a swelling in the front of the neck. The development of a carcinoma in a cyst of this type is rare (Batsakis, 1979). Malignancy is generally not established until the cyst has been removed (Villet and Kemp, 1981). The first case of carcinoma in a thyroglossal duct cyst was reported by Uchermann in 1915 (Trail *et al.*, 1977) and since then less than 100 cases have been described in the literature (Magsalin *et al.*, 1982). The most frequent tumour type (approximately 80 per cent) is the papillary thyroid carcinoma arising in ectopic thyroid tissue, which is seen in up to 75 per cent of these cysts (Batsakis, 1979). More infrequent is the follicular thyroid carcinoma. Primary planocellular carcinoma and non-thyroid adenocarcinoma originating from the epithelium lining the inside of the cyst has been described in only a few cases (White and Talbert, 1982; Vlaeminck, 1977). Papillary thyroid carcinoma arising in a thyroglossal duct cyst is seen most frequently in young women, with a sex ratio of 1:1.5 (Batsakis, 1979).

A case of papillary thyroid carcinoma arising in a thyroglossal duct cyst is described and, owing to their malignancy risk, the importance of extirpating these neck anomalies is stressed.

Case Report

An 18-year-old euthyroid and previously healthy girl was admitted with a slowly progressing infrahyoid cystic swelling which had been present for four years. There was no anamnestic basis for infection but an increasing globular feeling. Palpation of the neck demonstrated a normally located thyroid gland and no cervical lymph nodes. Thyroid scanning with Tc-99 confirmed a normal position and the thyroid parameters (T3, TT4, FT4 and TSH) were all within the standard area. Sistrunk's (1928) surgical technique was used and a cyst, about 3 cm in diameter, was excised together with the central portion of the hyoid bone, to which it was attached.

Histologic examination (Figs. 1 and 2) showed a cyst with a thick, fibrous wall. The cyst lumen was filled with papillary tumour tissue lined with a single layer of columnar epithelium with uniform, oval nuclei of empty or ground-glass structure. Mitoses were very rare. The fibrovascular stroma contained a few groups of follicles delimited by a similar epithelium. A few calcospherites were seen. The tissue on the outside of the capsule contained a few small foci of normal thyroid tissue. The characteristics of a papilliferous thyroid carcinoma arising in a thyroglossal duct cyst were thus present. There was no tumour invasion of the cyst wall nor to blood or lymph vessels in the surrounding tissue.

The patient's post-operative course was uneventful. Thyroid scanning three weeks later showed uptake only in the glandular area. Five weeks post-operatively CT-scanning of the neck with contrast medium was made and no signs of lymph node or local tumour recurrence were demonstrated.

Three months after surgery the patient was in good health but still under medical control.

Discussion

The thyroid gland develops at the end of the third fetal week as a proliferation of endodermal cells in the tuberculum impar in the floor of the

*From the ENT Department, St Joseph's Hospital
†Department of Pathology, Central Hospital, DK-6700 Esbjerg, Denmark.

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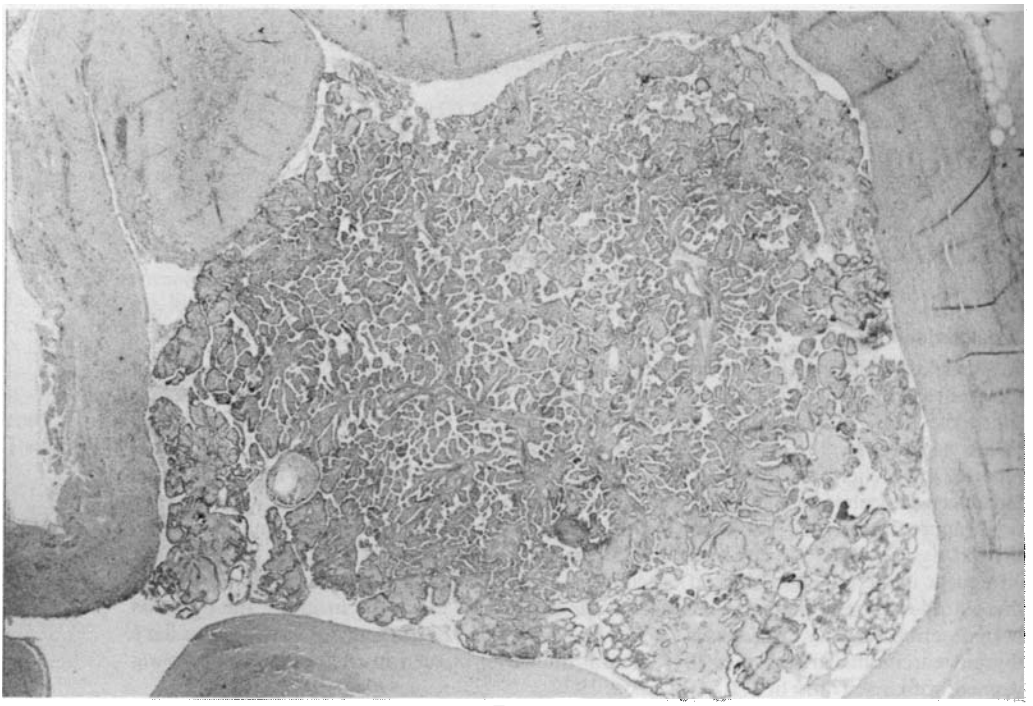


FIG. 1

Thyroglossal duct cyst with papillary tumour tissue. Original magnification ×3 (photo ×15).

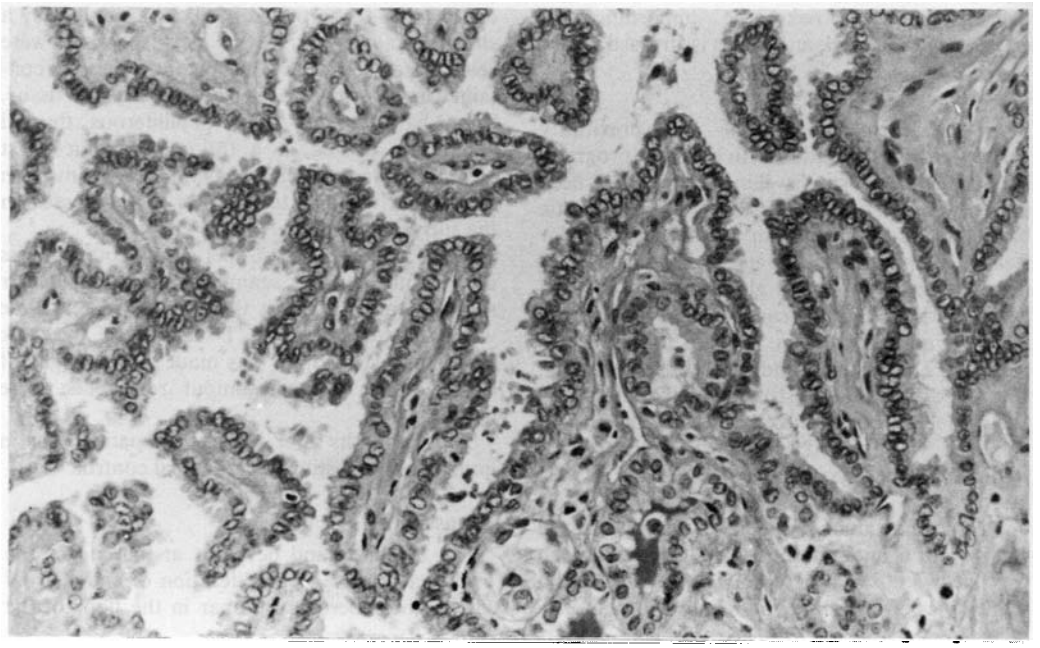


FIG. 2

Close-up of tumour lined with columnar epithelium with ground-glass nuclei characteristic of a papillary thyroid carcinoma. Original magnification ×75 (photo ×375).

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pharynx. During fetal development the central portion of the gland descends from the foramen cecum of the tongue, posterior to the first and anterior to the second branchial arch. From this point the hyoid bone develops to its final position in the lower portion of the neck anterior to the trachea. During this migration the thyroid gland remains connected with the foramen cecum through the thyroglossal duct, which starts degenerating and obliterating at the end of the sixth fetal week (Ellis and van Nostrand, 1977).

However, if the thyroglossal duct does not become obliterated totally, e.g. due to localized dilatation, part of the ductal epithelium may remain, and a cyst develops. According to its location, this cyst may be classified as suprahyoid (20 per cent), hyoid (15 per cent), or infrahyoid (65 per cent). Most cysts are subcutaneous, generally with no fistulation to the skin unless infection has been present (Batsakis, 1979).

Owing to its embryonic development, ectopic thyroid tissue may also be located anywhere along the thyroglossal duct.

There seems to be a hereditary accumulation of thyroglossal duct cysts and the lesion tends to recur after surgery (Magsalin *et al.*, 1982).

The literature reports various views of how malignant tumours arise in cysts of this type (Magsalin *et al.*, 1982). Some think that a thyroglossal duct cyst carcinoma represents a spreading of an occult thyroid carcinoma. Consequently, subtotal thyroidectomy is recommended, if necessary supplemented by neck dissection and radioactive iodization (Judd, 1963). Others think that the primary carcinoma arises in ectopic thyroid tissue in thyroglossal duct cysts. They recommend only the use of Sistrunk's surgical technique by which the cyst, the central portion of the hyoid bone and the foramen cecum are removed (Hans *et al.*, 1976; Page *et al.*, 1974).

However, the long-term results of the various treatments have not been mentioned in the literature and a final assessment of the prognosis cannot be made at the present time. It seems reasonable to accept that ectopic thyroid carcinoma arising in a thyroglossal duct cyst represents a malignant potential to be compared with identical forms in the thyroid gland itself. The prognosis is best in young people with well differentiated carcinomas (Vlaeminck, 1977).

As stated, the diagnosis of thyroglossal duct cyst carcinoma is most often made postoperatively. Excision of the entire duct in accordance with Sistrunk's surgical technique must be regarded as sufficient if the following criteria have been fulfilled: (1) that histologically normal ectopic thyroid follicles can be found in the cyst; (2) that the tumour has not yet extended through the cyst wall; (3) that the thyroid gland is fully normal; and (4) that no cervical lymph nodes are present. Prolonged control is indicated. If these criteria are not forthcoming, thyroidectomy with neck dissection should be made, if necessary followed by radioactive iodization in order to destroy any active endocrine foci which may remain (Vlaeminck, 1977).

Summary

A case of papillary thyroid carcinoma arising in a thyroglossal duct cyst is presented. The tumour is rare and is generally not expected preoperatively. The clinical and pathological characteristics are emphasized, the treatment is discussed, and the importance of excision of thyroglossal duct cysts is stressed.

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Address for correspondence:
Søren Kristensen, M.D.
ENT Department
St. Josephs Hospital
DK-6700 Esbjerg
Denmark

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