

Squamous cell carcinoma in thyroglossal duct cyst

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Abstract

Although a thyroglossal duct cyst (TDC) is a common cyst occurring in the neck, carcinomas arising in the TDC are a rare event. To date, approximately 100 cases have been reported, the majority of them being papillary carcinomas. Squamous cell carcinomas (SCC) are very rare, and only nine cases have been reported so far in the literature.

We present a 55-year-old female patient with a SCC in a TDC to highlight the clinical and pathological features of this condition.

Key words: Carcinoma, squamous cell; Thyroglossal cyst

Introduction

Carcinomas arising in a thyroglossal duct cyst (TDC) have been reported previously, most of them being papillary carcinomas and anaplastic carcinomas. Squamous cell carcinoma (SCC) arising in a TDC is very rare, only nine cases (Table I) being reported in the literature so far (Ashurst and White, 1925; Clute and Smith, 1929; Daalgard and Wetteland, 1956; Jacques *et al.*, 1970). A carcinoma arising from a TDC may rise from the lining of the cyst or from normal thyroid tissue present in the wall of the cyst. This occurs in only about one per cent of cases. Since the pre-operative diagnosis is usually benign in most cases, a high degree of suspicion is necessary for this diagnosis. We present a 10th case of squamous cell carcinoma arising from a TDC which was removed and treated with radiotherapy post-operatively.

Case report

A 55-year-old female patient was admitted with a swelling in front of her neck and hoarseness of voice which had been present three months. She was a non-smoker and there was no history of diabetes, hypertension or other systemic disease. Her general and systemic examination, including a PR and PV examination, showed no abnormality. On local examination, there was a round 2 × 2 cm swelling in the neck, situated near the midline which moved with deglutition and with protrusion of the tongue. The swelling was firm to hard to the touch. The cervical lymph nodes were not palpable, the carotids were felt and there were no systemic signs of thyrotoxicosis.

Nasopharyngoscopy, laryngoscopy and bronchoscopy were unremarkable. A chest X-ray was normal. An X-ray of the neck revealed a soft tissue mass in the lower part of neck. No calcification or vertebral destruction was seen.

TABLE I
REPORTED CASES OF SQUAMOUS CELL CARCINOMA IN TDC

Author(s)/Year	Age/Sex	Pathology	Comments
Clute and Smith (1929)	56/M		Post-operative radiotherapy. Tumour progression after first treatment
Nachlas (1950)	2	SCC	
Daalgard and Wetteland (1956)	44/F	SCC in TDC	Suprahyoid mass. Repeat surgery after 13 years
Ruppman and Georgsson (1966)	51/F		Presented with persistent and recurrent drainage: five resections
Shepard and Rosenfield (1968)	28/F	Cystic nodule, in the wall of which was SCC	Post-operative radiotherapy, SCC recurrence in larynx
Mobini <i>et al.</i> (1974)	50/F	Transition zone noted between SCC and normal duct lining	Post-operative radiotherapy. Patient did well
White and Talbert (1982)	61/F	SCC	Asymptomatic submental and submandibular mass
Lustmann <i>et al.</i> (1989)	80/F	SCC in lingual TDC	Post-operative radiotherapy
Yanagisawa <i>et al.</i> (1992)	65/M	SCC in TDC	Chronically draining sinus

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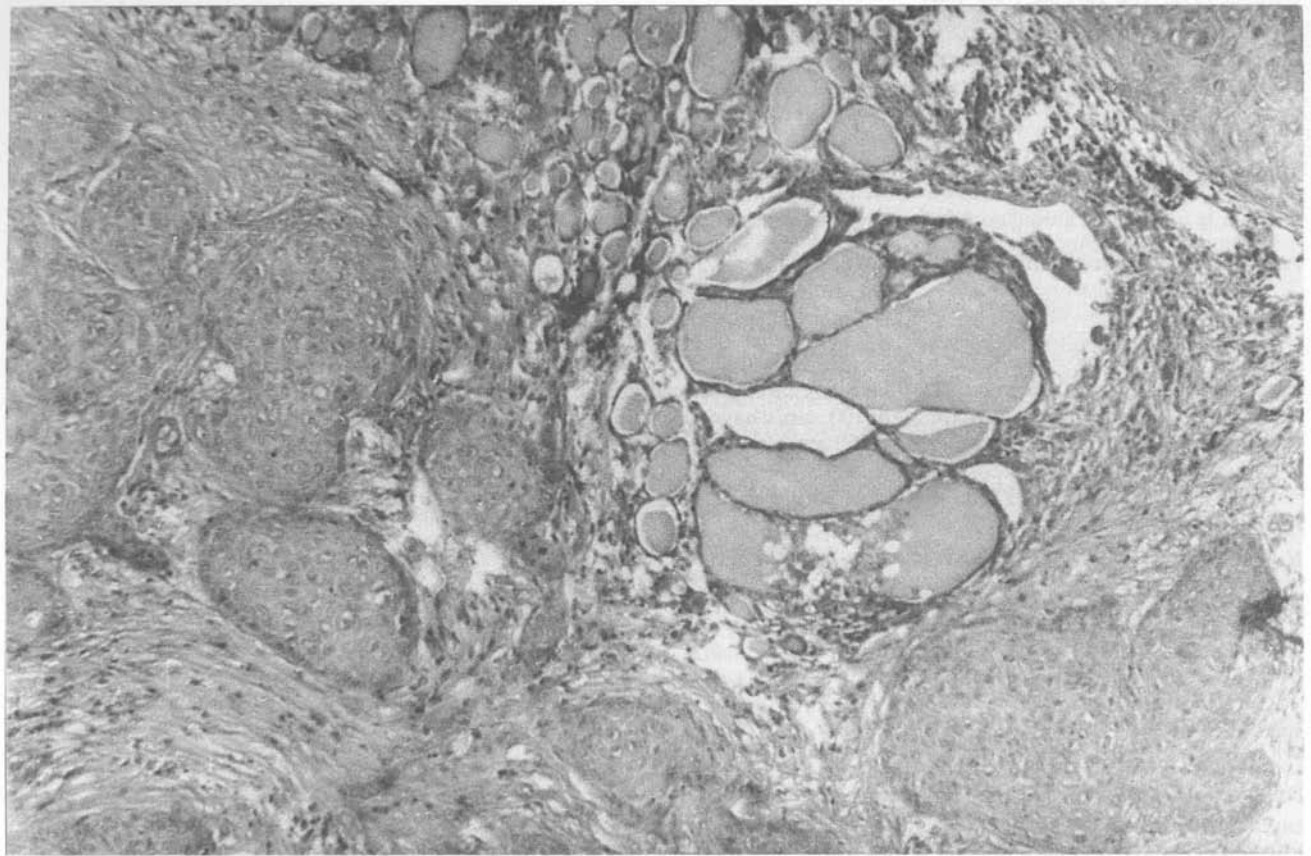


FIG. 1

Photomicrograph showing deposits of squamous cell carcinoma in the thyroid (H&E; $\times 10$).

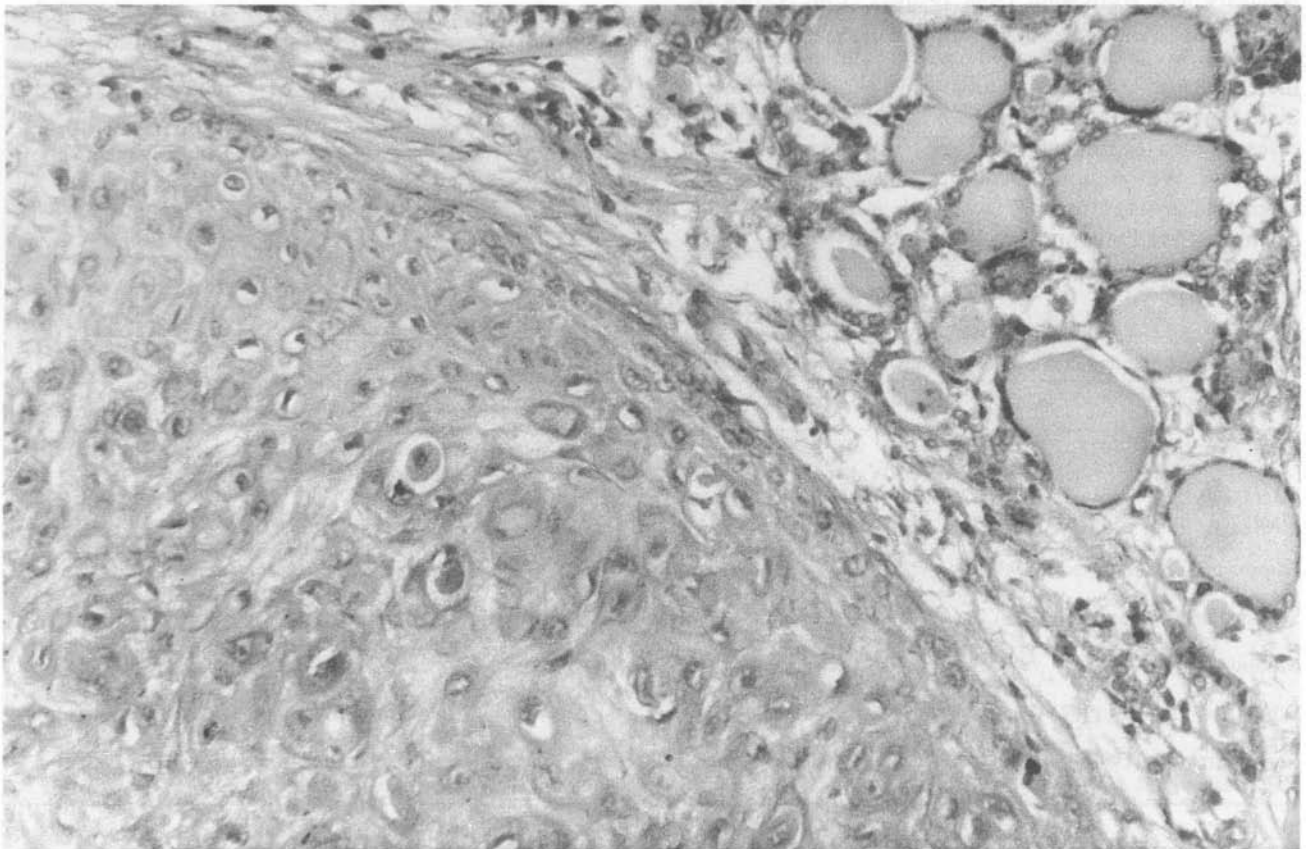


FIG. 2

Photomicrograph showing details of squamous deposits. (H&E; $\times 45$).

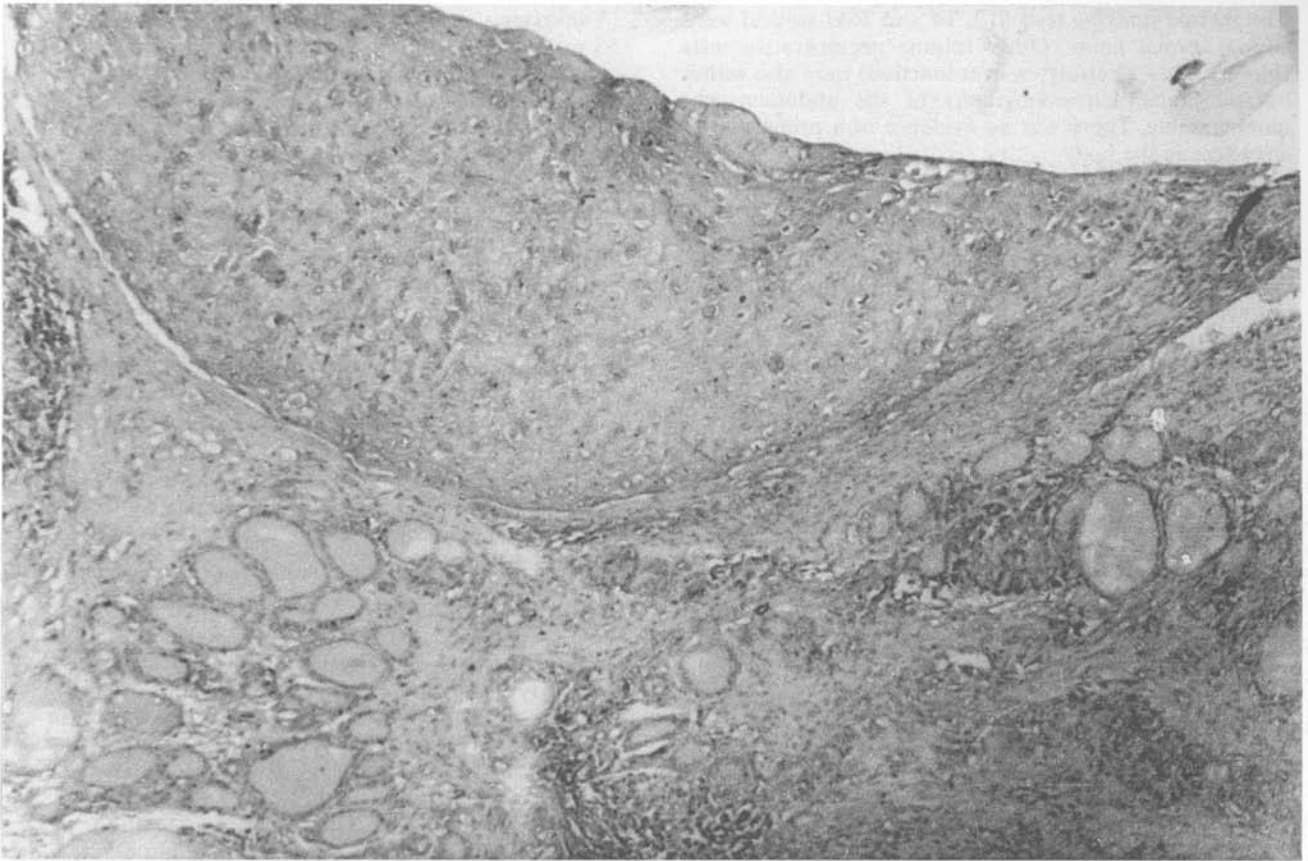


FIG. 3
Photomicrograph of a TDC lined by malignant squamous cells. (H&E; $\times 10$).

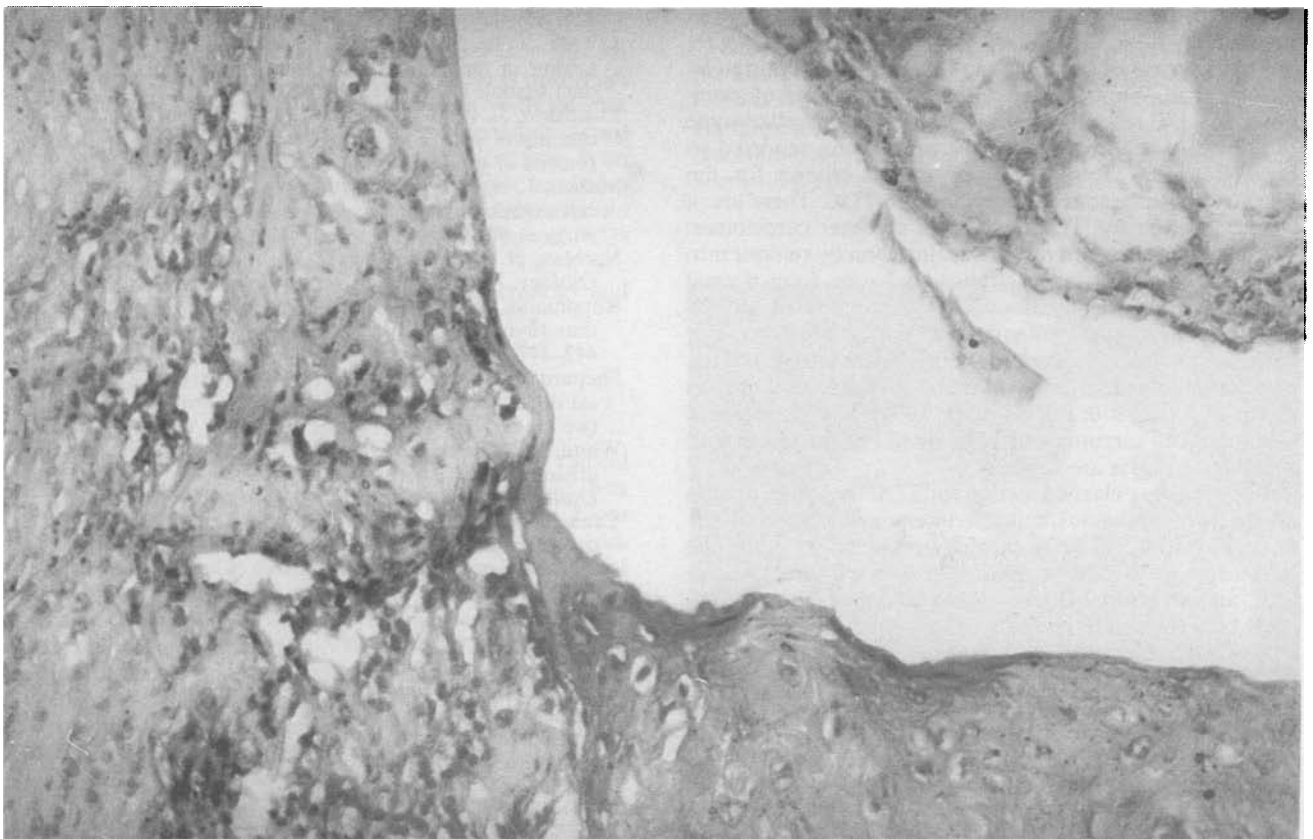


FIG. 4
Photomicrograph showing transition zone between malignant squamous cells (lower right) and flattened lining of TDC (left). (H&E; $\times 45$).

The thyroid function tests (T3, T4 and TSH values) were within normal limits. Other routine pre-operative tests (blood counts, electrolytes, liver function) were also within normal limits. Ultrasonography of the abdomen was unremarkable. There was no evidence of a primary SCC anywhere in the body.

Intra-operatively, a hard nodule was found in the midline extending into right lobe of thyroid. It was fixed to the strap muscles from which it had to be separated. We observed the hemithyroidectomy specimen with the excised nodule, measuring 2.5×3 cm. The thyroid looked normal macroscopically with a well demarcated rounded nodule with a greyish white cut surface showing a few haemorrhagic areas. Histological examination showed a cystic space containing a transition zone from normal TDC into invasive squamous cell carcinoma. The cyst was lined by flattened epithelium (Figure 1). Nests of well differentiated malignant squamous cells were also seen in the adjacent thyroid gland (Figure 2). The rest of the thyroid gland was normal and there was no evidence of papillary, follicular or anaplastic carcinoma. There was evidence of invasion of surrounding skeletal muscles, proving the malignant nature of the tumour. Figure 3 shows a thyroglossal duct cyst lined by malignant squamous cells and Figure 4 the transition zone between malignant squamous cells and the flattened lining of the TDC.

Discussion

Thyroglossal duct remnants are situated anywhere along the course of the thyroglossal duct, from the base of the tongue to the thyroid. The lining may be respiratory ciliated or metaplastic squamous. Mucous glands, thyroid tissue and lymphoid follicles are commonly seen in the wall of the cyst. Carcinoma of the TDC may arise from the cyst lining or from normal thyroid tissue present in the wall of the cyst.

The first case of carcinoma in the TDC was reported by Ashurst and White (1925). Subsequent reports of carcinoma in TDC revealed that most are of the papillary type (86 per cent) approximately 100 cases being reported so far. White and Talbert (1982) offered criteria for the diagnosis of squamous cell carcinoma in TDC. These are: a classic location for TDC; exclusion of other carcinomas; presence of normal thyroid tissue; invasion by tumour into surrounding tissues; and a transitional zone from normal TDC into invasive squamous cell carcinoma. It is well known however that inflammation may destroy part, or whole of the lining epithelium of thyroglossal cyst, and the cyst wall may contain only granulation tissue and fibrosis (Jacques *et al.*, 1970; LiVolsi *et al.*, 1974). The diagnosis of squamous cell carcinoma in TDC should be made when all the above criteria are fulfilled.

In our case, a classic location for TDC was present, and all primary squamous cell carcinomas especially of the head and neck had been carefully excluded by doing the necessary investigations. Histology also showed invasive SCC amidst normal thyroid tissue and a clear transition zone between normal TDC and invasive SCC. The lining of the TDC was made up of granulation tissue and fibrosis.

The average age of any patient presenting with benign TDC disease is 20 years (Daalgard and Wetteland, 1956), those presenting with any type of carcinoma in TDC is 39.2 years (Allard, 1982), whereas the average age of patients with squamous cell carcinoma in TDC is 54.3 years

(Yanagisawa *et al.*, 1992). The age in the present case is 55 years.

Some patients with a midline neck mass of long duration that may develop into a recurrent draining fistula (Ruppman and Georgsson, 1966; Yanagisawa *et al.*, 1992) or may present with a midline neck mass with a duration of weeks to months (LiVolsi *et al.*, 1974). At other times the surgeon may remove the cyst and find a warty growth attached to the inside or outside the wall (Jacques *et al.*, 1970). Squamous cell carcinoma may arise from the squamous lining of a TDC whereas papillary carcinoma may arise from normal thyroid tissue present in the cyst wall (Ruppman and Georgsson, 1966).

To the best of our knowledge this is the 10th reported case of squamous cell carcinoma in a TDC. Our patient was treated with radiotherapy post-operatively and the patient's course was uneventful.

Since the clinical diagnosis is usually of benign TDC disease, a high degree of suspicion and the knowledge of this entity is useful for diagnosis.

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