



Fig 3.

to occur. Bile also provides a good medium for bacterial growth and so infective sequelae often occur in the setting of a cholethorax.

Biliary pleural fistulas and the formation of bilious pleural effusions are known complications of hepatic trauma<sup>1,2</sup>, parasitic liver disease<sup>3</sup> and development of a subphrenic abscess in the setting of biliary obstruction. Iatrogenic causes include biliary stent migration<sup>4</sup>, radio-frequency ablation<sup>5</sup> and following cholecystectomy<sup>6</sup> and liver biopsy<sup>7</sup> However, it is the increasing use of percutaneous biliary drainage which has led to the greatest number of cases.<sup>8-10</sup> For a Cholethorax to arise disruption of the pleural space needs to have occurred and this may not necessarily be obvious during the procedure. Rapid thoracentesis, correction of the cause of the fistula, adequate analgesia and the treatment of infective sequelae are essential in the management of this group of patients.

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#### Diffuse sclerosing variant of papillary thyroid carcinoma – a rare cause of goitre in a young patient

Editor,

Papillary thyroid carcinoma is the most common thyroid malignancy. We report a case of a rare variant - diffuse sclerosing papillary thyroid carcinoma (DSPC).

**Case History:** An 18 year old girl presented with a smooth symmetrical goitre. She was clinically euthyroid and had no palpable cervical lymph nodes. Thyroid function tests and anti-thyroid peroxidase level were normal. Ultrasound scan of thyroid showed marked nodular enlargement of the entire gland in keeping with a multinodular goitre. A hypoechoic 1cm nodule was identified at the right lobe which was found to be 'cold' on radio-isotope scanning. A fine needle aspiration of this 'cold' nodule was reported as papillary carcinoma.

She was booked for total thyroidectomy. At surgery she had an enlarged thyroid, with a gross appearance in keeping with a thyroiditis or lymphoma. Frozen section confirmed papillary carcinoma. The gland was hard and gritty. Several local lymph nodes were also excised. Post-operative recovery was uneventful.

Sectioning revealed a diffusely firm, white, gritty gland (fig 1). Histopathology showed this to be the rare diffuse sclerosing

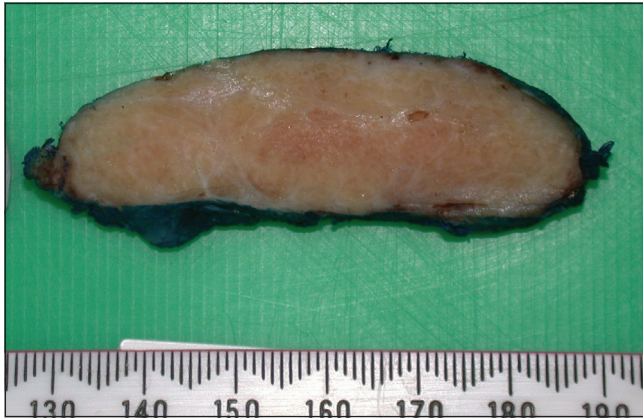


Fig 1. Sclerotic hemisection of thyroid lobe

variant of papillary thyroid carcinoma. Scattered islands of tumour tissue, squamous metaplasia, stromal sclerosis, heavy lymphoplasmic infiltrate and abundant psammoma bodies were found diffusely throughout both lobes and through the capsule (fig 2). All excised lymph nodes contained metastatic carcinoma.

She underwent radioablative therapy followed by replacement levothyroxine. There was no residual uptake on subsequent 123-Iodine isotope scanning.

**Discussion:** First described in 1985, diffuse sclerosing papillary carcinoma of the thyroid (DSPC) is a rare variant malignancy, recently reported to account for 0.8% of papillary thyroid carcinomas.<sup>1,2</sup> Patients present with a diffuse goitre and are mostly clinically euthyroid, but can also be hypothyroid or hyperthyroid. It occurs most frequently in young females and may be mistaken clinically for benign disease particularly thyroiditis.<sup>3-5</sup> Most patients have lymph node metastases at the time of diagnosis and lung metastases are common.<sup>3</sup> Cerebral metastases have also been reported.<sup>6</sup>

The presence of several pathological characteristics is diagnostic: diffuse firm enlargement of the thyroid gland, scattered islands of papillary carcinoma, extensive lymphatic permeation and lymphocytic infiltration, squamous

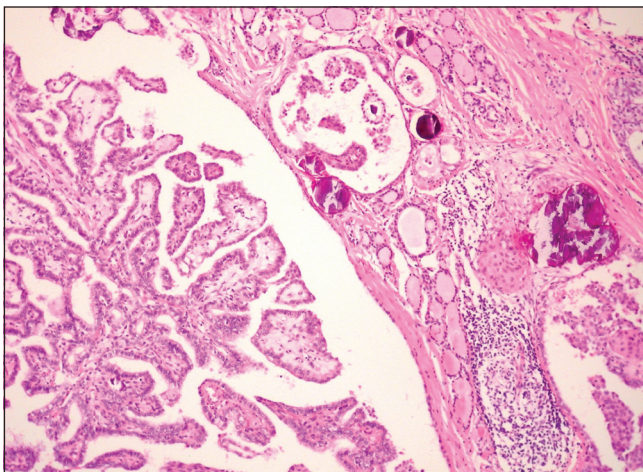


Fig 2. Papillary carcinoma, with psammoma bodies and squamous metaplasia. Sclerosis and chronic inflammation, to right of field. (x 200)

metaplasia, sclerosis and numerous psammoma bodies.<sup>7</sup> Detection of abundant psammoma bodies on ultrasonography may provide pre-operative evidence of DSPC.<sup>4</sup> This could facilitate improved surgical planning for a technically challenging thyroidectomy.

Early studies suggested that DSPC had a poorer prognosis than classical papillary carcinoma due to its aggressive nature with frequent lymph node and distant metastases at the time of presentation. It had also been reported that eradication required a more aggressive therapeutic approach.<sup>3</sup> However, more recent studies suggest that DSPC patients have a similar prognosis and that the treatment should be that for classical papillary thyroid carcinoma i.e. radical surgery, radio-iodine ablation and/or external radiotherapy.<sup>2,8</sup>

There are potential pitfalls which may delay the diagnosis of DSPC. In this case, the clinical presentation, biochemical, serological and initial radiological findings were all indicative of benign pathology. FNA indicated malignancy leading to surgery demonstrating its importance in the diagnosis of DSPC. As metastases are frequently present it is therefore important to consider this rare malignancy when investigating a goitre in a young patient.

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