

Fig. 1. Axial contrast enhanced CT section obtained as a baseline study after completion of radiation therapy shows the expected appearance of the pre-epiglottic space following radiation therapy. No abnormality is seen in the pre-epiglottic space.

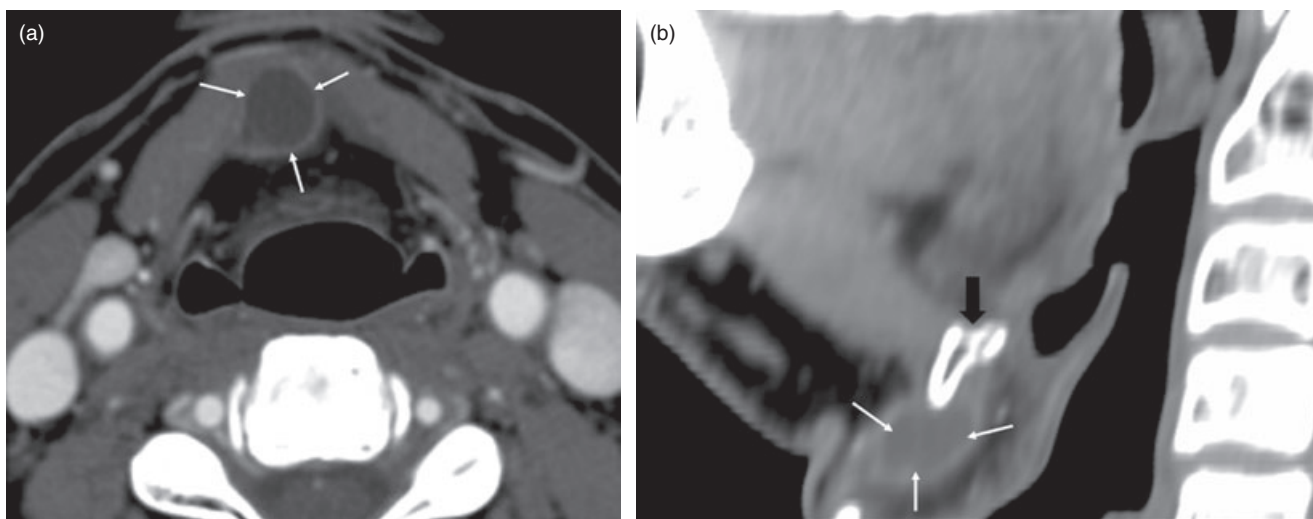


Fig. 2. (a) Axial contrast enhanced CT section performed about 18 months later reveals a low-attenuation structure (white arrows) in the pre-epiglottic space with a mildly enhancing rim. (b) Sagittal reformatted CT demonstrates the close proximity of the cystic structure (white arrows) to the hyoid bone (black arrow).

Rare presentation of thyroglossal duct cyst after radiation therapy to the neck

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SUMMARY

Thyroglossal duct cyst (TDC) is a congenital lesion which usually manifests as an enlarging painless neck mass in a child or young adult. We highlight an unusual presentation of TDC in an adult which developed after radiation therapy to the neck. We discuss the probable sequence of events and emphasize the need for radiologists, radiation oncologists and the surgeons to be aware of this entity when evaluating patients after radiation therapy.

Key words: congenital; radiation therapy; thyroglossal duct cyst.

INTRODUCTION

Thyroglossal duct cyst (TDC) results from a developmental anomaly of the thyroid gland and accounts for 70% of congenital neck masses.¹ The majority of patients present within the first two decades of life but nearly a third of cases may manifest in young adulthood beyond 20 years of age.² They usually manifest as an enlarging painless neck mass in a child or young adult and in most circumstances, the diagnosis can be made by history and physical examination.³ We wish to highlight an unusual presentation of TDC in a 47-year-old man patient which developed after external beam radiation to the neck for an unknown primary.

CASE REPORT

A 45-year-old man patient was referred from his primary care physician to the otolaryngology service of our hospital for an incidentally detected left neck mass that the patient had noted while shaving. The patient had undergone a contrast enhanced CT scan of the neck which demonstrated a 2 × 1 cm left level 2 lymph node and mild asymmetric fullness of the left nasopharynx. The remainder of the scan was normal. Ultrasound guided fine needle aspirate of the left level 2 node had shown a keratinizing squamous cell carcinoma. An extensive workup including multiple site biopsies for detection of primary tumour was negative except for carcinoma *in situ* in the left tonsil, which was not fluoro-deoxyglucose avid with Positron emission tomography. It was felt that the diagnosis of carcinoma *in situ* of the tonsil was not consistent with metastatic disease and hence it was decided to treat the patient as a Tx, N1, M0 squamous cell cancer of the neck with unknown primary. In total, 7000 cGray of external beam radiation was delivered over seven cycles with significant nodal regression. A contrast-enhanced CT was performed at this time which showed resolution of the neck mass and no evidence of a visceral space mass (Fig. 1).

After an asymptomatic period of about 18 months following radiotherapy, the patient presented to our institution with left-sided otalgia. He denied any dysphagia, odynophagia,

headache, cough, shortness of breath or hoarseness of voice. Contrast enhanced CT scan of the neck was performed. Compared with the baseline CT scan performed after completion of radiotherapy (Fig. 1), the current scan demonstrated development of a new cystic midline mass in the pre-epiglottic space. The lesion was located immediately inferior to the hyoid bone between the strap muscles. It measured about 1.3 × 1.1 cm in anteroposterior and transverse dimensions and demonstrated a peripheral rim of mild enhancement (Fig. 2a,b). The remainder of the scan was unremarkable with no evidence of other enlarging masses compared with the prior CT. The differential diagnosis included metastatic prelaryngeal node versus a TDC.

Ultrasound-guided fine needle aspiration biopsy of the lesion showed cystic contents with no neoplastic cells. Because of the lack of a definitive diagnosis, it was decided to resect the lesion. The cyst was excised using a modified Sistrunk procedure.

Pathology revealed a cystic cavity lined by pseudostratified ciliated epithelium and presence of thyroid follicles in the stroma adjacent to the cyst consistent with a diagnosis of TDC (Fig. 3a,b). No neoplastic features were observed. The midline location, proximity to the hyoid bone and pathological features of simple cyst with thyroid follicles in adjacent stroma helped establish the final diagnosis of benign TDC. The patient's otalgia was felt to be related to upper respiratory tract infection which resolved spontaneously.

DISCUSSION

The present case illustrates a rare presentation of TDC that developed after external beam radiation to the neck. The appearance of a new, low attenuation lesion in the pre-epiglottic space in a patient treated with radiotherapy for a Tx, N1, M0, unknown primary carcinoma suggested a high likelihood of a metastatic prelaryngeal lymph node rather than a benign entity. Histopathological examination proved otherwise and helped establish the final diagnosis of TDC.

Thyroglossal duct cysts develop from persistence of the mid portion of the thyroglossal duct, an embryonic structure that

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Submitted 1 June 2006; accepted 29 August 2006.

doi: 10.1111/j.1440-1673.2007.01842.x

traces the path of descent of the thyroid gland.⁴ The usual manifestation of a TDC is an enlarging, painless, midline neck mass in a child or young adult that moves upward with tongue protrusion.^{2,3,5} The cyst may rarely manifest as a floor of mouth mass.^{6,7} To the best of our knowledge, presentation of a TDC after radiation therapy to the neck has not been reported earlier. We hypothesize that repeated local inflammation because of radiation may have caused scarring or oedema of an 'occult' or subclinical thyroglossal duct and triggered secretions from the epithelial lining resulting in cyst formation. The fact that the cyst could not be appreciated, even in retrospect, on the CT scan performed after completion of radiation therapy (Fig. 1) further strengthens our hypothesis that radiation played a key role in development of the cyst.

As in our case, the differential diagnosis of anterior midline/paramidline cystic masses includes cystic/necrotic metastatic prelaryngeal lymph node, TDC, dermoid or epidermoid cysts and second branchial cleft cysts.^{8,9} Cystic metastatic nodes usually arise from papillary carcinoma of the thyroid or squamous cell carcinoma of the upper aerodigestive tract, and can show solid component with abnormal vascularity on ultrasound. Punctate calcifications may be observed in metastases arising from papillary carcinoma of thyroid. Though dermoid and epidermoid cysts can mimic TDCs in their location (midline) and cystic nature, presence of fat (hyperechoic on ultrasound or fat attenuation on CT) is a clue favouring a dermoid or epidermoid cyst.^{8,9}

The lesion in our patient lies in the vicinity of the expected location of the prelaryngeal group of lymph nodes, which belongs to the anterior deep cervical chain consisting of the prelaryngeal, prethyroid, pretracheal and laterotracheal groups. These nodes receive afferent lymphatics arising from the supraglottic larynx including the epiglottis, aryepiglottic folds and the piriform sinuses.¹⁰ Hence, our primary concern after the CT scan indicating an enlarging mass in the pre-epiglottic space was that of a metastatic prelaryngeal lymph node that drained the supraglottic larynx.

Lymph nodes and lymphoid tissue usually regress with radiation therapy and enlarging lymph nodes after definitive radiotherapy are considered an ominous sign.^{11,12} In our case, it was unclear to our multidisciplinary tumour board as to whether this midline cystic mass in the anterior neck, which developed in a patient with squamous cell carcinoma following radiation therapy, was a benign or a malignant process. As a result, the patient underwent surgery to identify the histology and determine if further treatment was necessary.

In conclusion, this case demonstrates an interesting result of radiation therapy which, to the best of our knowledge has not been reported. It also emphasizes the need for radiologists, radiation oncologist and the head and neck surgeon to be aware of this entity when evaluating patients after radiation therapy.

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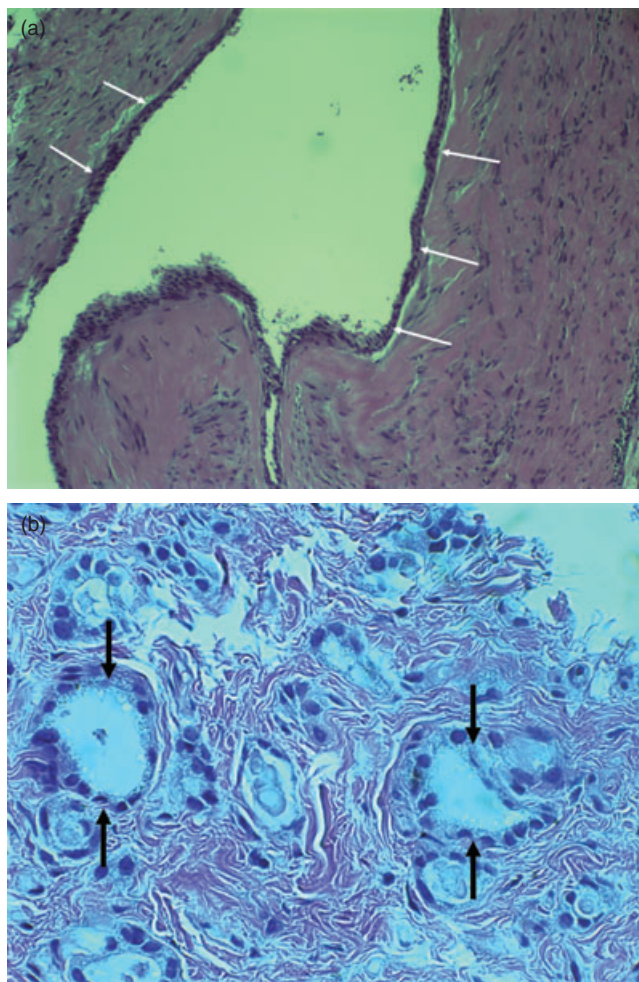


Fig. 3. (a) Histopathological examination of the cyst using haematoxylin and eosin stain, viewed at 10× magnification, reveals a cavity lined by pseudostratified columnar epithelium (white arrows). (b) Histopathological examination of the stroma adjacent to the cyst using haematoxylin and eosin stain, viewed at 40× magnification, demonstrates multiple thyroid follicles (black arrows).