

Dual ectopic thyroid: a report of two cases

PRODUL HAZARIKA, M.S., F.A.C.S., SHAHAB AHMED SIDDIQUI, KAILESH PUJARY, PARUL SHAH,
DEEPAK RANJAN NAYAK, M.S., R. BALAKRISHNAN, M.S.

Abstract

Ectopic thyroid is an uncommon embryological aberration characterized by the presence of thyroid tissue at a site other than in its usual pretracheal region. Usually it occurs along the path of descent of the developing thyroid primodium from the foramen caecum, commonest being lingual followed by sublingual and in the anterior midline of neck at, or below, the level of the hyoid bone. It is unusual for lingual thyroid to present simultaneously with another ectopic thyroid; reported here are two such cases.

Key words: Thyroid gland, ectopic tissue

Introduction

The thyroid primodium develops in a 3–4 mm embryo as an endodermal bud from the anterior floor of the pharynx and descends down on either side of the trachea to its pretracheal site where it fuses with the caudal prolongation of the fourth pharyngeal pouch to form the thyroid gland. Arrest in descent can occur just below the foramen caecum (lingual), between the geniohyoid and mylohyoid muscles (sublingual) or just above or below the level of the hyoid bone. Due to the close relationship between the developing thyroid primodium and the aortic trunk and septum transversum, ectopic thyroid can rarely present in the superior mediastinum, pericardial sac or even in the developing heart, commonly in the upper anterior mediastinum adjacent to the aortic arch and between the aortic and pulmonary trunks (Thompson, 1986). Rarely, it can also present at other sites such as intra-tracheal and intra-oesophageal (Hazarika *et al.*, 1985).

Hickman, 1869, has been credited with the first diagnosis of lingual thyroid in a 16-hour-old female infant who died of suffocation caused by a mass obstructing the oropharynx. It is very unusual for two ectopic foci of thyroid tissue to present simultaneously. Only four such cases have been reported (Quigley *et al.*, 1962; Kuehn *et al.*, 1966; Rosen and Walfish, 1967; Gabatin and Pretorius, 1985) being lingual and subhyoid with pretracheal thyroid.

Case history 1

A 32-year-old male patient presented with a history of swelling in the tongue for five years and swelling below the chin (submandibular and submental regions) for three years. The tongue swelling had been gradually increasing in size for six months, during which time the patient was experiencing a foreign body sensation in the throat, and a change in voice for one month, causing embarrassment to the patient. The submandibular swelling was not enlarging.

Fine needle aspiration cytology (FNAC) of the submandibular swelling showed features of thyroid tissue and follicles. A thyroid scan with Technetium (Tc99m) showed

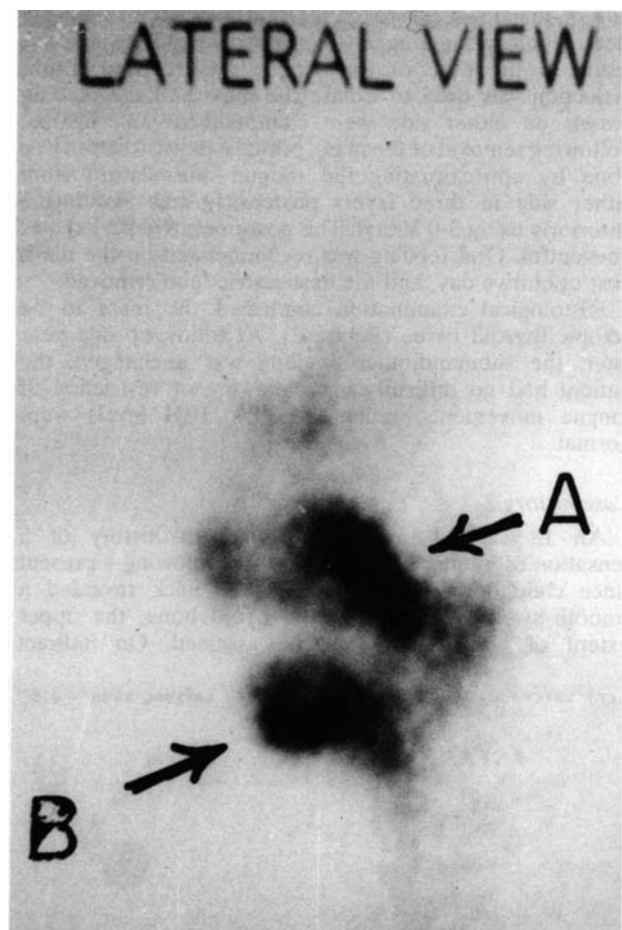


FIG. 1
Thyroid scan (Tc99m) Case 1: Uptake in tongue base (A) and submandibular region (B).

From the Department of Otolaryngology, Kasturba Medical College, Manipal 576119, India.
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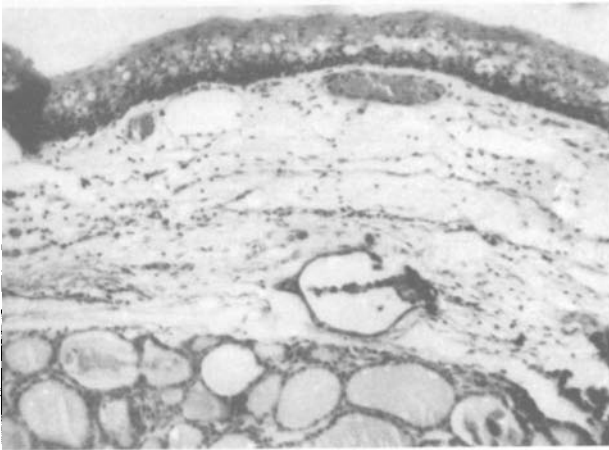


FIG. 2

Histopathology – lining squamous epithelium with underlying thyroid follicles filled with colloid.

uptake in the region of the tongue base and in the region of the submandibular swelling, most apparent on lateral view with no other hot or cold nodules (Figure 1). Serum T3, T4, TSH levels were normal.

A transoral tongue – splitting approach (without labiomandibulotomy) was used to excise the lingual thyroid: Two long No. 1 silk sutures were placed on either side of the tip of the tongue. Using a surgical knife and electrocautery, the tongue was split in the midline until the lesion at the base of the tongue was reached. Blunt dissection was used to isolate the mass and the feeding vessels on either side were clamped, cut and ligated. Following removal of the mass, primary reconstruction was done by approximating the tongue musculature from either side in three layers posteriorly and two layers anteriorly using 3-0 Vicryl. The post-operative period was uneventful. Oral feeding was recommended on the ninth post-operative day, and the nasogastric tube removed.

Histological examination confirmed the mass to be ectopic thyroid tissue (Figure 2). At follow-up one year later, the submandibular swelling was unchanged, the patient had no difficulty in swallowing or restriction of tongue movements; repeat T3, T4, TSH levels were normal.

Case history 2

An 18-year-old male presented with history of a sensation of a lump in the throat on swallowing – present since childhood. Examination of the neck revealed a smooth swelling just above the hyoid bone, the upper extent of which could not be assessed. On indirect



FIG. 3

Thyroid scan (Tc99m) Case 2: Uptake in tongue base and upper neck (suprahyoid).

laryngoscopy, a smooth hemispherical swelling was seen in the region of the tongue base extending to the vallecula. On palpation it was 4 × 3 cm firm, and non-tender. A Tc99m scan revealed two foci of uptake one in the region of the tongue base and the other in the region of the upper neck. No uptake was noted in the region of the normal cervical thyroid (Figure 3). Serum T3, T4, TSH levels were normal. The patient was counselled regarding the anomaly and has been advised to come for regular follow-up. When seen after two years, both the swellings were constant with no change in his symptoms. Repeat T3, T4, TSH were normal.

Discussion

Most ectopic thyroids are asymptomatic or may present with mild symptoms in which case the patient needs only counselling and regular follow up is advised. Common presenting symptoms are a feeling of foreign body or lump in the throat, change in voice, interference with deglutition, haemorrhage, dyspnoea or orthopnoea (Weider and Parker, 1977). Occasionally, a lingual thyroid may be an accidental finding complicating intubation especially since mild trauma can cause profuse bleeding (Harris, 1990).

This condition is best diagnosed by physical examination. Lingual thyroid presents as a firm, pinkish, hemispherical mass covered by a thin epithelium through which vascular markings can be plainly seen. Radioactive scan using Tc99m, or I121, or I123 allows estimation of size and degree of activity of the ectopic thyroid and also ascertains the presence or absence of normal cervical thyroid tissue. FNAC may reveal numerous loosely arranged honey-combed sheets of round, cuboidal epithelial cells resembling thyroid follicular cells with small or large deposits of colloid. Without colloid the smears can be mistaken for a minor salivary gland tumour or granular cell myoblastoma (Kumar *et al.*, 1996). Cervical ultrasonography, computerized tomography, and magnetic resonance imaging (MRI) are excellent methods of describing the size and position of the lesion within the tongue or in the neck (Guneri *et al.*, 1991).

Differential diagnosis should include thyroglossal duct cysts, angioma, fibroma, adenoma, lipoma and malignant conditions (Sethi and Chatterji, 1984) and minor salivary gland tumours.

Management of patients with ectopic thyroid tissue is dependent on several factors including the size of the lesion, the presence of local symptoms, age of the patient, status of the thyroid gland and presence of complicating factors such as ulceration, haemorrhage, cystic degeneration, or malignancy (Gabatin and Pretorius, 1985; Guneri *et al.*, 1991). Suppressive treatment with thyroid hormone prevents hypertrophy of the ectopic thyroid tissue followed by replacement with thyroxine for life. Radioactive iodine ablation is reserved for older patients who remain asymptomatic and in whom surgery is not thought appropriate but is contraindicated in children in whom the systemic doses required have potentially damaging effects on the gonads and other organs. There is also a possibility of malignant transformation in normal functioning thyroid tissue (Gabatin and Pretorius, 1985).

Autotransplantation has been performed with varying success since its first description by Buckman, in 1936; in these cases some degree of hypothyroidism should be expected for at least four months before the transplanted tissue will regain full function. It is best done in children when obstructive symptoms present without evidence of hypothyroidism and when the tissue shows no degenerative or cystic changes (Hazarika *et al.*, 1988). The ectopic thyroid can be transposed into the lateral cervical gutter in

the neck, under the sternocleidomastoid. This technique has the advantage of an attached vascular pedicle, based on the dorsalis linguae artery and can be done via a median or lateral pharyngotomy approach. The median pharyngotomy approach provides adequate visualization and easy control of bleeding vessels and is preferred to the lateral pharyngotomy or the transhyoid approach.

The posterior midline tongue splitting approach has been reported to have a good exposure for control of bleeding from the dorsal lingual arteries, and restricted dissection within the tongue. It avoids injury to vital structures of the neck (including the lingual nerves), fistula formation, salivary contamination of the neck tissues and an obvious scar (Atiyeh *et al.*, 1995). The technique used in our case (*Case 1*), the transoral midline tongue splitting approach, offers similar benefits with little more morbidity and can therefore be used for surgical ablation before autotransplantation in the rectus abdomini when indicated.

In our *Case 1*, after excision of the lingual thyroid no transplantation was needed as the other ectopic thyroid tissue functioned normally. In the second case, as the symptoms were mild and thyroid function tests were normal, only counselling of the patient was required and the patient has been followed up on a regular basis.

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Address for correspondence:
Dr Deepak Ranjan Nayak,
Department of ENT,
Kasturba Medical College,
Manipal 576119,
India.