

Lingual thyroid: tongue-splitting incision for transoral excision

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Abstract

Two patients presenting to the Central Military Hospital of Beirut with symptomatic lingual thyroid are reported. I¹³¹ thyroid scanning revealed the lingual thyroid to be the only functional thyroid tissue present in each patient. Subsequent CT scanning demonstrated the large size of these ectopic thyroids causing significant mechanical obstruction. These were excised transorally using a posterior midline tongue-splitting incision and reimplanted in the rectus abdominis muscles.

Details of this modified tongue-splitting surgical approach are described. A brief review of the literature concerning lingual thyroid and its surgical treatment is also presented as well as three patients operated on for lingual thyroid at the American University of Beirut Medical Centre between 1975 and 1994 using an external neck incision.

Key words: Lingual thyroid; Transplantation, autologous; Reimplantation

Introduction

Embryologically, the thyroid is derived from three primitive sources (Volpe, 1973). A diverticulum in the area of the foramen caecum at the base of the tongue appears about the third week of gestation, then it migrates down the neck to its final destination (Gilchrist, 1968) where it is joined at about the seventh week by two lateral masses of cells derived from the fourth pharyngeal arches. These lateral anlagae are however inconsistently present (Ward *et al.*, 1954). Remnants of thyroid tissue along the course of migration of the major median anlagae result in the formation of thyroglossal duct cysts that may be present anywhere from the foramen caecum to the pretracheal area passing by the hyoid bone. Total failure of embryological migration produces clinically the lingual thyroid without any functional thyroid tissue in the normal cervical position in the majority of cases. Cystic changes within lingual thyroid tissue have been reported (Cancrini *et al.*, 1987).

Postmortem studies have shown an incidence of 10 per cent for lingual thyroids. The clinical incidence however, varies between 1:3000 and 1:10 000 (Williams *et al.*, 1989) the lingual thyroid being the only functioning thyroid tissue in 70 per cent of these cases (Al-Samarrai *et al.*, 1988; Kaplan, 1989). It is estimated that only one clinical case is seen at the Mayo Clinic every three to five years (Weider and Parker, 1977). The incidence of malignancy is

extremely low (Fish and Moore, 1963; Hazarika *et al.*, 1988). Roman *et al.* (1991) described the first known case of squamous cell carcinoma associated with ectopic lingual thyroid tissue.

Ectopic thyroid tissue is present three to four times more frequently in females than in males (Hillness and Black, 1976; Kansal *et al.*, 1987). Alderson and Lannigan (1994), based on an old reference, reported the female to male ratio to be 7:1. The age of patients at diagnosis ranges between six and 74 years (Roman *et al.*, 1991). Common presenting symptoms are dysphagia, dysphonia, foreign body sensation in the throat and bleeding from the oral cavity (Elprana *et al.*, 1984; Gallo and Ellis, 1985). Lingual thyroids however, are often asymptomatic (Kansal *et al.*, 1987). Diagnosis is confirmed by either technetium scintigraphy or by I¹³¹ scanning. Ultrasound and CT scanning are also helpful (Harris, 1990). When the lingual thyroid is associated with other normally located functioning thyroid tissue, total excision is curative, otherwise, reimplantation of the excised ectopic thyroid tissue in the abdomen or the neck (Al-Samarrai *et al.*, 1988) is the established way in which to avoid hypertrophy of lingual remnants and the development of a hypothyroid state. Surgical extirpation of the lingual thyroid can be performed transorally or through an external neck incision (transhyoid or lateral pharyngotomy). Lateral pharyngotomy seems to be the most widely accepted surgical

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Accepted for publication: 4 December 1994.

approach for this condition (Alderson and Lannigan, 1994).

Materials and methods

Review of the files at the American University of Beirut Medical Center showed that between 1975 and 1994 only three patients, one male and two female with an age range of 35 to 55 years, were operated on for symptomatic lingual thyroids. Surgery on all three patients was performed through a transverse suprahyoid skin incision. The duration of the surgery was between three and four hours. One patient developed post-operative drainage from the neck incision and had severe pharyngeal oedema necessitating prolonged nasotracheal intubation. No attempt was made in any of these patients to autotransplant the excised thyroid tissue and they were maintained on thyroid replacement therapy.

During 1988, two patients presented to the Central Military Hospital of Beirut with the same condition (Figure 1). In both these patients the lingual thyroid was excised transorally using a posterior tongue-splitting incision and the excised thyroid tissue was reimplanted under the rectus sheath in the para-umbilical area through two separate small incisions. The entire surgical procedure lasted about one and a quarter hours.

One patient developed an abscess in one of the abdominal incisions requiring drainage. Nevertheless there were no complications in the head and neck area attributable to the surgical procedure. None of these patients was given thyroid replacement therapy post-operatively. Thyroid scan performed three months after surgery demonstrated uptake of the transplanted thyroid tissues, with minimal residual uptake in the tongue area without recurrence of the mass. One patient remained slightly hypothyroid and was due to receive thyroid replacement when she was lost to follow-up. The second patient remained in a clinically adequate condition six years after his surgery though he persistently failed to report for control blood tests six months post-operatively.

Surgical procedure

In the supine position, with the head elevated, general anaesthesia is administered through a nasotracheal tube which is best inserted guided by a fibre optic endoscope. This eliminates the need for a tracheostomy. The mouth is held open and a traction suture is applied to the tip of the tongue. Several traction sutures are also applied along the lateral borders of the tongue progressing posteriorly as far as the palatoglossal arch. These sutures are deliberately made haemostatic, particularly at the base of the tongue, by including as much tongue tissue as possible within the needle bite. With steady traction on these sutures, the tongue is pulled anteriorly exposing the lingual thyroid under the palatal arch. An incision is then made in the midline over the posterior two-thirds of the tongue stopping

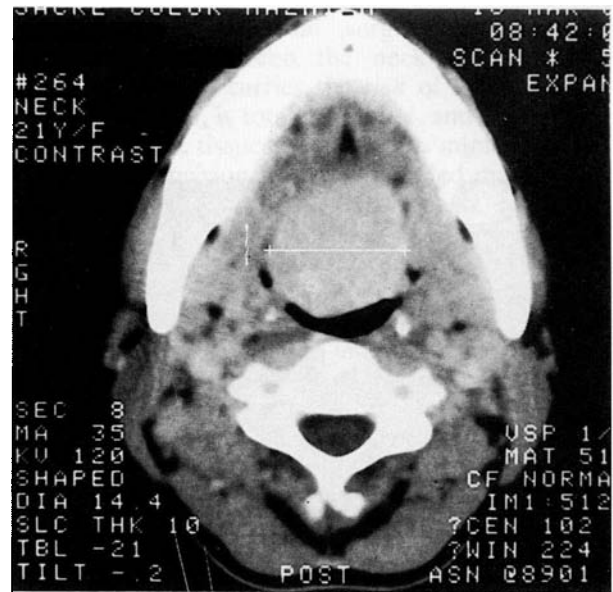


Fig. 1

CT scan of a patient with a lingual thyroid.

just short of the protruding mass. The incision is deepened, slicing the tongue muscle in half. Posteriorly this incision may be carried through the tongue substance down to the hyoid bone. At this stage of the procedure, brisk bleeding from the dorsal lingual arteries at the base of the tongue is easily controlled by heavy absorbable suture ligatures. Using blunt and sharp dissection, the lingual thyroid is excised, with the overlying mucosal lining, under direct vision. Finally the tongue is sutured in one layer starting from the posterior end of the surgical wound.

The excised thyroid tissue is immediately placed in cold saline. The overlying mucosa as well as the surrounding tongue muscle included in the resection are removed. The thyroid tissue is then cut into slices 4–5 mm thick using a dermatome blade. Through two separate transverse small abdominal incisions the slices of thyroid tissue are grafted into the rectus abdominis muscles under the fascia (Figure 2).

Post-operatively the patient is intubated and kept under observation in the intensive care unit for 36 to 48 hours. Appropriate antibiotics are also administered for seven days.

Discussion

There is no real consensus in the literature about the proper management of lingual thyroid due to the rarity of this condition and the small series reported. It seems that even in the best of hands, total excision of the ectopic thyroid is difficult irrespective of the surgical approach.

The goals of any comprehensive line of management should be to relieve the obstructive symptoms produced by the protruding mass at the base of the tongue without endangering the life of the patient during surgery, and also without producing mutilating scars. If possible lifelong hypothyroidism should

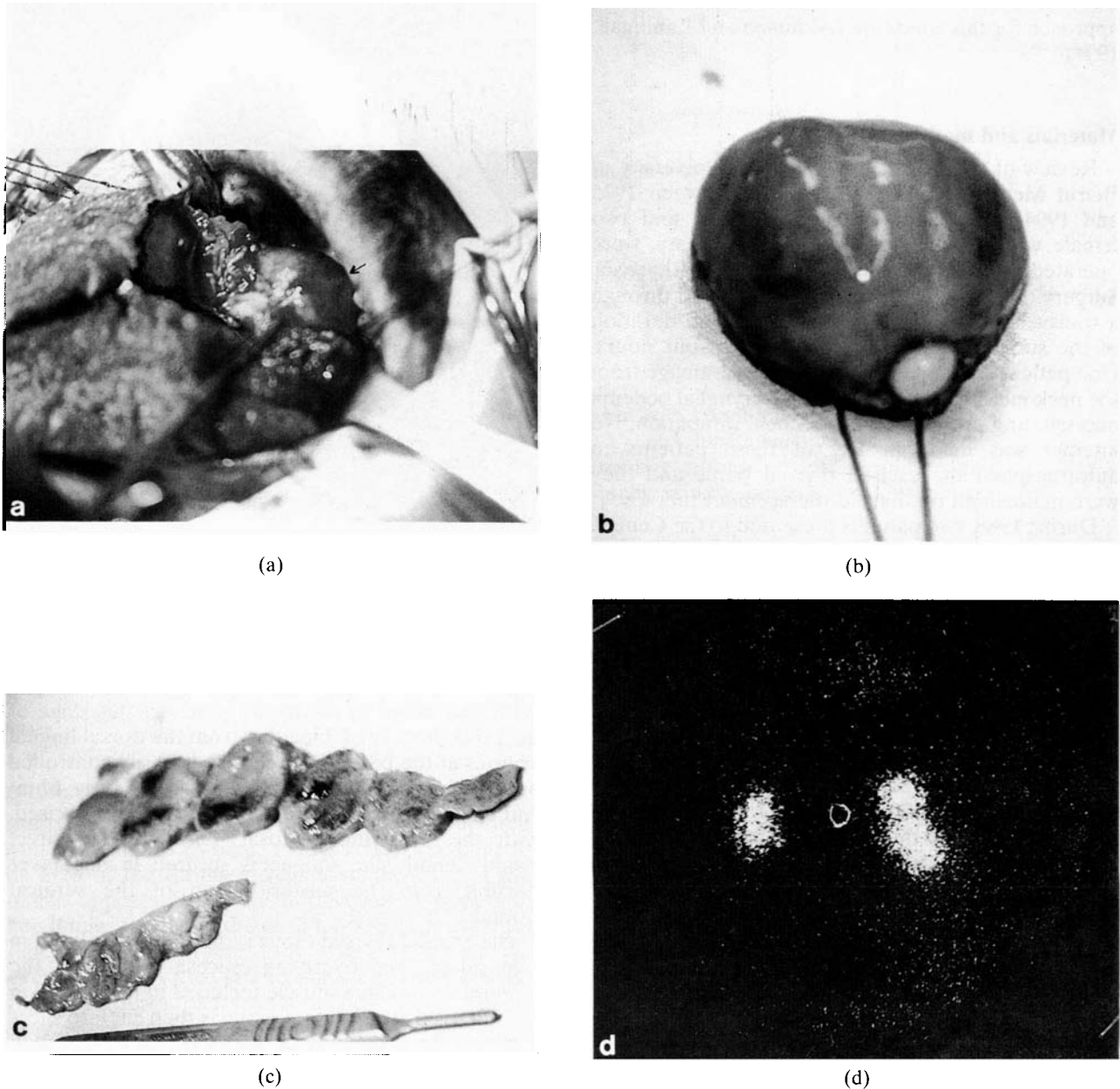


FIG. 2

(a) Posterior two-thirds midline tongue-splitting incision exposing the lingual thyroid (arrowed). (b) Excised lingual thyroid. (c) Thyroid tissue sliced with a dermatome blade. (d) I^{131} scan of the abdominal wall three months after autotransplantation.

be prevented particularly in a child or a young female of child-bearing age.

Treatment of a lingual thyroid depends on the sex and age of the patient as well as on the severity of the symptoms and the associated ulceration and haemorrhage. Patients with mild symptoms can be treated successfully by medical suppression (Fish and Moore, 1963). Reduction in size of the ectopic gland is however very slow with thyroxin treatment and no dramatic results should be expected. Patients with severe obstructive symptoms, ulceration and haemorrhage do not usually respond to hormonal treatment and should therefore undergo surgery (Kamat *et al.*, 1979; Kansal *et al.*, 1987). The ectopic thyroid tissue can also be ablated by a therapeutic dose of I^{131} after which the patient is placed on thyroid hormone replacement for life (Roman *et al.*,

1991). Radioactive iodine therapy is however particularly contraindicated in females of childbearing age (Hillness and Black, 1976). The best alternative in most symptomatic patients remains surgical excision (Ward *et al.*, 1954) with autotransplantation (Weider and Parker, 1977).

Total excision of lingual thyroid tissue is difficult to confirm at the time of surgery. At best thyroid remnants will be left at the base of the tongue, and these might hypertrophy again unless they are checked by replacement therapy or functioning transplanted thyroid tissue. Whether the transplanted thyroid tissue may produce enough hormones to meet the physiological needs of the patient is difficult to postulate, but when it does, it eliminates the necessity of lifelong thyroid hormone replacement therapy.

Surgical excision of the lingual thyroid can be performed transorally, or through a neck incision by a midline or lateral pharyngotomy. Irrespective of the surgical technique, intentional partial excision leaving thyroid tissue at the base of the tongue should be avoided as it may hypertrophy again causing myxoedema in most cases (Ward *et al.*, 1954). Remnants of thyroid tissue, after an attempt at total excision (as illustrated by our two cases) are usually located deep in the tongue substance and may be less likely to reproduce the obstructive symptoms should they hypertrophy again.

The majority of cases reported in the literature have been excised transorally. Several modifications have been introduced in order to improve the exposure such as splitting the cheek, lower lip, mandible or the tongue (Kaplan, 1989). This aggressive surgery seems to be extravagant for the resection of a benign ectopic thyroid gland that may not be complete. The major problem encountered with the oral approach, besides poor exposure, is inadequate control of bleeding which at times may be life threatening. Kamat *et al.* (1979), described bilateral ligation of the lingual arteries at the level of the cornu of the hyoid bone through separate skin incisions before completely splitting the tongue in the midline. This has been associated, in some cases, with devastating necrosis of the tongue.

The transhyoid or lateral pharyngotomy approaches are more major surgical procedures carrying the risk of serious complications without facilitating surgical exposure.

Steinwald *et al.* (1970), described the retention of portions of the lingual thyroid as vascularized flaps rotated down towards the neck. This is achieved by a transverse suprahyoid skin incision with an H-shaped tongue opening. There is however no evidence that the retained thyroid tissue in this technique is sufficient to avoid myxoedema, or that the technique is superior to autotransplantation of thyroid tissue. Needless to say extensive dissection and surgical manipulation in the oropharyngeal region produce severe post-operative oedema making tracheostomy mandatory in certain cases.

The modifications of the standard splitting of the tongue described in this paper offer many advantages by applying haemostatic traction sutures, slicing only the posterior two-thirds of the tongue in the midline down to the hyoid bone, exposing the anterior and superior aspect of the lingual thyroid for direct visual inspection, and widening the surgical field. Opening the tongue like a book exposes the dorsal lingual arteries at the base of the tongue, making the control of brisk bleeding they invariably produce, very easy to achieve under direct vision by the application of suture ligatures.

Contrary to the pharyngotomy or transhyoid approach, injury to vital structures like major blood vessels or nerves in the neck, is unlikely since the dissection is restricted to the midline within the tongue muscle substance. The lingual nerves which run in close association with the deep lingual vessels are unlikely to be injured since they are located

laterally away from the surgical field. Direct communication between the neck skin and the oropharynx which carries the risk of fistula formation, though rare, is totally avoided, and contamination of the neck tissues with saliva is minimized. An obvious skin incision is totally avoided making this approach highly attractive particularly to young females.

The use of neck incision for excision and grafting of the lingual thyroid tissues (Al-Samarrai *et al.*, 1988), is very hazardous because of the risk of contamination and infection leading to the loss of the grafted material.

When our method is compared to other surgical approaches, particularly to the suprahyoid external approach utilized on the three patients in our retrospective study, surgical time is sharply reduced which is a considerable factor in these times of increasing medical costs.

Conclusions

A modification of transoral excision of lingual thyroid through a midline posterior tongue splitting incision has been described. The advantages of such a procedure are its relative safety, technical ease, good cosmetic outcome and short surgical time. When combined with fiberoptic nasotracheal intubation and intentional delayed post-operative extubation, a tracheostomy can be avoided in most cases.

References

- Alderson, D. J., Lannigan, F. J. (1994) Lingual thyroid presenting after thyroglossal cyst excision. *Journal of Laryngology and Otolaryngology* **108**: 341–343.
- Al-Samarrai, A. Y., Crankson, S. J., Al-Jobori, A. (1988) Autotransplantation of lingual thyroid into the neck. *British Journal of Surgery* **75**: 287.
- Cancrini, F., Mezzetti, M., Frattarol, F. M., Berardi, A., Bonomolo, F., Reggio, D., Ospedale, S., Carlo Nancy, R. (1987) Lingual thyroid: a case report. *Italian Journal of Surgical Sciences* **17**: 265–268.
- Elprana, D., Mann, J. J., Smalls, A. G. (1984) Lingual thyroid. Case report and review of the literature. *Journal of Otolaryngology and Related Specialties* **46**: 147–152.
- Fish, J., Moore, R. M. (1963) Ectopic thyroid tissue and ectopic thyroid carcinoma. *Annals of Surgery* **157**: 212–222.
- Gallo, W. J., Ellis, E., IIIrd. (1985) Management of a large lingual thyroid in the orthognathic surgery patient. *Oral Surgery, Oral Medicine, Oral Pathology* **59**: 344–348.
- Gilchrist, G. F. (1968) *A Survey of Embryology*, McGraw-Hill Co., New York, p 313.
- Harris, W. E. (1990) Lingual thyroid cyst complicating a foreign body in the oesophagus. *Journal of Laryngology and Otolaryngology* **104**: 508–539.
- Hazarika, P., Murty, P. S., Nooruddine, M. S., Zachariah, J., Rao, N. R. (1988) Lingual thyroid. *Ear, Nose and Throat* **67**: 161–165.
- Hillness, A. D., Black, W. B. (1976) Lingual ectopia of the thyroid gland and autotransplantation. *British Journal of Surgery* **63**: 924–926.
- Kamat, M. R., Kulkarni, J. N., Desai, P. B., Jussawalla, D. J. (1979) Lingual thyroid: a review of 12 cases. *British Journal of Surgery* **66**: 537–539.
- Kansal, P., Sakati, N., Rifai, A., Woodhouse, N. (1987) Lingual thyroid: diagnosis and treatment. *Archives of Internal Medicine* **147**: 2046–2048.
- Kaplan, E. L. (1989) Thyroid and parathyroid. In *Principles of Surgery*. Vol. 2. 5th Edition. McGraw-Hill Co., New York, pp 1614–1616.

- Roman, P., Bukachevsky, J. D., Casler, J. O., Conley, J. (1991) Squamous cell carcinoma and thyroid gland. *Ear, Nose and Throat* **70**: 505–507.
- Steinwald, O. P., Muehcke, R. C., Economou, S. G. (1970) Surgical correction of complete lingual ectopia of the thyroid gland. *Surgical Clinics of North America* **50**: 1177–1186.
- Volpe, R. (1973) The thyroid. In *Systemic Endocrinology*. (Ezzin, C., Gooden, J. O., Volpe, R., Widson, R., eds.), Harper and Row, Maryland, pp 55–56.
- Ward, G. E., Canterell, J. R., Allan, W. B. (1954) The surgical treatment of lingual thyroid. *Annals of Surgery* **139**: 536–544.
- Weider, D., Parker, W. (1977) Lingual thyroid. *Annals of Otolaryngology* **86**: 841–848.
- Williams, E. D., Toyn, C. E., Harach, H. R. (1989) The ultimobranchial gland and congenital thyroid abnormalities in man. *Journal of Pathology* **159**: 135–141.

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