LINGUAL THYROID*

By H. W. SCHWARTZ (Halifax, Nova Scotia, Canada)

A WELL-DEVELOPED female aged 42, a school teacher by profession, consulted me April 6th, 1946, for hoarseness and discomfort in breathing which she had been aware of for about six months. No disease of the nose, throat or larynx could be recognized but an elongated bluish-red swelling in the midline of the tongue and about one-third its width seemed to extend from the epiglottis to the circumvallate papillae, that is the posterior third of the tongue.

The possibility of it being a cyst of some kind led me to explore with a fine needle. No fluid was secured and the puncture bled out of proportion to its size. I concluded it must be an angioma of some sort probably a hæmangioma. Several of my associates, surgeons both general and special, as well as a roentgenologist, to whom the condition was demonstrated—appeared to entertain a like opinion and accordingly suggested that a series of X-ray treatments be given. This was carried out in March, 1947. Re-examination after two months revealed no change.

It was felt that its removal might prove to be a bloody and dangerous matter and that further opinion was desirable. During my absence at the Canadian Medical Association, Dr. R. H. Stoddard made arrangements for her to go to the Lahey Clinic. On my return their report, dated August 6th, was passed to me and reads as follows:

> "Our examination of the nose and throat was not remarkable other than the growth in the base of the tongue. This was in the midline and approximately two centimeters in diameter. It was smooth and firm and very characteristic of the lingual thyroid. She was seen also by Dr. Lahey who could not feel any thyroid in her neck and we felt that the entire thyroid was in the tongue and that if this thyroid were removed it would be necessary for her to take thyroid the rest of her life in order to avoid myxœdema. We felt that this lingual thyroid was not large enough to cause any serious trouble or inconvenience and that it was not adenomatous and was, therefore, unlikely to become malignant. We felt that it would be less inconvenient for her to continue with the thyroid as is than to be troubled with the taking of thyroid extract the rest of her life if it was removed. We suggested the use of ammonium chloride and potassium iodide to increase secretions and be sooth-

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ing to her voice. We feel that further X-ray treatment is contra-indicated as it does dry up the mucous glands in the pharynx and causes more trouble in speaking because of the dryness.

After the summer holidays the patient was again examined and after a number of attempts a Kodachrome record was made.

The B.M.R. on October 25th was −12, the normal range is between −10 and +10. Blood cholesterol 198—normal range between 160 and 200 (Bloor's Method Modified).

Accepting the diagnosis as correct, then we have a condition of considerable rarity. It is difficult to be sure of the actual incidence of lingual thyroid because it is only those who are inconvenienced that seek relief.

Lahey (1923) from his goitre clinic found two cases in 7,600. When B. M. Chute discussed Dr. Miller's (1933) paper in 1932 the number of lingual thyroids still remained only two when the total had risen to 8,500, while Ulrich noted one in 4,000. More recently Schilling (1950), Karr and Hursh reported in 1950 the first case among 244,000 admissions to the Strong Memorial and Rochester (N.Y.) Municipal Hospitals, which included 11,900 autopsies and 4,200 cases of thyroid disease. Chute and Cattell (1930) consider that this anomaly, associated with the failure of the thyroid to decend, is one of the rarest. If these statistics reflect the true frequency then very few will see very many. They are, however, more often seen in a throat clinic and cases were shown at the Royal Society of Medicine in London from time to time. Bertein and Gelle have an exhaustive review of the subject and Smyth reported three cases of his own and 45 from the literature.

Hartley (1922) writes that: "The thyroid gland is present in all vertebrates and even in the most primitive is composed of closed vesicles containing so-called colloid material. Moreover, throughout the vertebrates, the thyroid takes origin, for the most part if not entirely, from a median anlage which grows down from the ventral wall of the pharynx. In the human embryo, the median anlage occurs at a very early stage (2·5 mm.) as an evagination from the mesial portion of the ventral wall of the primitive pharynx. The evagination quickly develops into a sac, the orifice of which opens on the summit of the tuberculum impar (first rudiment of the tongue) and then migrates backward to its posterior boundary furrow; the pouch becomes bilobed and travels caudally. The thyroglossal duct, which for a time has a double lumen, atrophies, and soon the thyroid breaks its connection with the pharynx (7 to 8 mm.). The organ then becomes a broad structure composed of irregular cords of cells disposed for the most part transversely. In 50 mm. vertex breech embryo, some of the thyroid cords develop lumina, become beaded, and soon thereafter break up into rudimentary follicles. Probably for some time after birth, the formation of thyroid continues. Iodine, which is so characteristic of the adult thyroid,

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is not present until some months after birth." Hartley in this article reviews the phylogenetic (evolutionary) and ontogenetic (development of the individual organism) histories and concludes that "the prevailing opinion, however, would appear to be that no proof exists of functionating thyroid tissue being derived from any other source than the median pharyngeal rudiment."

Opinion on this would appear to differ. Sir James Walton (1943) writes—"the lateral lobes are formed independently from down growths on each side from the fourth visceral cleft." And Krafka (1942) writes—"the thyroid is generally conceded to have its origin in a single median primordium arising as the thyro-glossal duct at the tubeculum impar (unpaired rudiment of the tongue) in the 1·4 mm. stage and from a pair of lateral primordia (at 4 mm. stage) from the epithelium of the ventral wall of the 4th branchial pouch. Contact is lost with the surface epithelium by the median anlage at 13 mm. and caudal migration separates the lateral primordia from the pouch of epithelium at the 16 mm. stage." The observation of Norris referred to by Cattell and Hoover (1929) supports the contention of Hartley—"That no proof exists of functionating thyroid tissue being derived from any other source than the median pharyngeal rudiment" and at the same time makes clear as to what happens the thyroid-like tissue that takes origin from 4th branchial pouch. Permit me to quote: "lateral aberrant thyroid tissue has been shown by Norris to develop from the posterior portions of the third and fourth branchial pouches in contrast to the thyroid gland which comes from the median portion of the floor of the pharynx. He has demonstrated the fusion of these lateral bodies with the main body of the gland in the 15 mm. human embryo. Following this these lateral bodies atrophy and take no part normally in the formation of the adult gland. The cessation of this process in any stage accounts for thyroid tissue in the lateral position." Seven such cases were observed in 7,600 goitre operations, an incidence of approximately one in a thousand.

Dr. Frank H. Lahey, when discussing Dr. Miller's paper, said when referring to these lateral aberrant thyroid masses: "These are peculiar tumours. You will be surprised to find that they look like melanotic sarcomas, bluish black in colour, very soft, running up each side of the internal jugular vein, papilliferous in character, very sensitive to radiation, but a definite number of them become malignant. If you encounter these bluish black tumours . . . they should be taken out radically."

Judging from the case reports, the majority of those operated on were performed in ignorance of the true nature of this lingual tumour. Either the pathologist or the subsequent development of myxœdema established the diagnosis. All do not develop hypothyroidism after operation, but how is one to tell? If symptoms become very troublesome operative treatment may become imperative. C. H. Mayo (1911) stated that only one in

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seven subsequently developed hypothyroidism. If such a statement still holds it in itself suggests that (in the majority of cases) some functioning tissue must exist in addition to the visible tumour. (This might support the triple origin contention for this type of tissue or that the mass was not totally removed in the majority of cases or the presence of sufficient aberrant thyroid tissue).

Regarding treatment I will quote Ulrich (1932): "When lingual thyroid attains such a size as to produce urgent dyspnoea and dysphagia at any age, active treatment is indicated. In the young, tracheotomy should be the first step. In the mature patient it may or may not be necessary before thyroidectomy depending upon the dexterity of the surgeon and the degree of dyspnoea. When doing tracheotomy in the young patient, a large enough incision should be made to allow for an exploration of the neck to determine whether or not there is any normally situated thyroid. If any amount is found, one may with impunity proceed with lingual thyroidectomy and assume on the basis of experimental evidence that the remaining portions of thyroid gland will undergo hyperplasia and care for the normal metabolic activity of the patient. If normally situated gland is absent, it is imperative to adopt a conservative programme. Such medical measures as are of use in the treatment of other physiological enlargements of the thyroid should be tried. In the presence of a low basal metabolic rate, mixed treatment with iodine and thyroid extract may be effective in reducing the size of the lingual goitre to the point where symptoms are relieved. If the basal metabolic rate is normal or high, iodine alone should be given. In event that medical treatment fails to reduce the size of the lingual goitre, thereby not relieving the obstructive symptoms, partial or complete thyroidectomy must be done. The electrosurgical knife may greatly facilitate this operation from the standpoint of its greatest technical hazard, i.e. hæmorrhage. In the absence of other thyroid tissue, as determined by exploration, the excision of a lingual thyroid should be followed immediately by the administration of thyroid extract, regardless of the age of the patient."

"I am particularly interested," continues Ulrich, "in that group of patients with lingual thyroids who have only mild or moderate symptoms referable to it. These patients should all be submitted to an exploration of the neck. The examination of the neck by palpation so frequently leaves one in doubt about the thyroid gland that I believe it is to the best interest of the patient, the family doctor and the consultant to know whether or not there is any thyroid tissue in or about its normal location. Exploratory operation entails so little risk, while the findings at operation are so valuable for the proper management of a case of lingual thyroid that one need not hesitate to recommend it. The high incidence of myxœdema after excision of a lingual thyroid from a patient of any age and the potential danger of arrested development, both physical and mental, in the young, justify this

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means of learning as much as possible about the patient from a thyroid standpoint.

In this group of patients, no active treatment should be directed toward the aberrant thyroid if no normally situated thyroid tissue is found which in my opinion will be the rule rather than the exception. The programme here should be one of watchful waiting with careful follow-up, personally or through the family doctor, and the judicious use of iodine during the times of excessive physiological demands upon the thyroid gland."

Excision in whole or in part would appear to be the usual procedure when surgical treatment is indicated. Whalen (1934) reports having treated a case with apparent success by using a bipolar electrocoagulating current under a local anæsthetic. He destroyed a sufficient amount of the mass to relieve the patient's distress in swallowing and breathing and left enough tissue to prevent the development of untoward symptoms. The report was made six months after the treatment.

Within relatively recent times radioactive iodine has been used in tracer doses in the diagnosis of this condition. Nachman, Crawford and Bigger report such an investigation followed by operation and the subsequent radioautograph revealed that most of the iodine was localized in the acinar walls, but that little was incorporated in the colloid.

Others have deliberately used radioactive iodine as a form of treatment. Impressed by the difficulties encountered in the surgical treatment Schilling, Karr and Hursh treated their patient, a 30-year old white woman, with radioactive iodine (I 131). The loss of vascularity, the onset of atrophy, and the development of fibrosis was slow anatomically and extended over a period of $2\frac{1}{2}$ years before it was considered complete. Functional change was prompt as shown by the early onset of symptoms of hypothyroidism and ·032 gm. of thyroid extract three times a day was given orally and has to be continued. Apart from this medical nuisance the patient was free from discomfort and in excellent health 3 years later. As the authors write—"In this instance the effective reduction in size of a lingual thyroid has been simply accomplished with no risk to the patient. This entity is so rare that few observers have the privilege of seeing or treating more than one such case in a life time. It is hoped that this report will be useful to others who might have an opportunity to treat such a patient."

The thyroglossal duct disappears by the beginning of the second month of fœtal life. Portions may persist and give rise to cysts. Thyroglossal cysts most frequently make their appearance during the first two decades and although they may remain stationary for years, they tend as a rule to grow larger and are prone to become infected. Not infrequently the swelling is incised in the belief that it is an abscess arising from an infected gland. The tract may pass in front of the hyoid bone, but more commonly behind or

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may be gripped between the two rudiments of the body of the hyoid. It is, therefore, advisable when a cyst is anchored to the thyroid to remove the middle portion of that bone and to search for a tract leading up to the foramen cæcum.

The lingual thyroid is much more common in women than in men. The ratio is about 8 to 1. The sizes observed have ranged from that of a pea to that of an orange. Several instances have been reported occurring in the newborn and one died of asphyxiation within a matter of hours. The average age of recognition would appear to be 23 (Grace and Weeks, 1932).

The case that is the text for this presentation has what is believed to be on clinical examination a lingual thyroid that has made no attempt to travel. Radio-active iodine was not available and a biopsy was not done for fear of an embarrassing amount of bleeding.

There are ultra curiosities the intra-lingual, the naso-pharyngeal, the intra-œsophageal, and its total absence in the myxœdematous still-born. That minute amounts of thyroid tissue may be found unexpectedly is well illustrated in a case that had been operated on by Sir James Hodsdon for epithelioma of the lip, an islet of thyroid tissue being accidentally found in a lymph node immediately behind the symphysis menti; the node was about the size of a pea. So far as could be ascertained, the thyroid was normally situated in the neck.

Recently at the Victoria General Hospital my associate Dr. D. M. MacRae, when performing a laryngectomy, removed what he believed to be a pre-thyroid cartilage lymph node. This solitary encapsulated pea-sized structure proved to be thyroid tissue. The thyroid gland was in its normal position.

I am greatly indebted to Dr. Richard L. Saunders, Professor of Anatomy at Dalhousie University, for combining his artistic talents and imagination with his knowledge of embryology and anatomy in helping to make clear to us clinicians what would otherwise be vague and obscure, and to Dalhousie University for a grant in aid of publication.

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LANTERN SLIDES

Lantern Slides 1 and 2

The thyroid gland first appears in early embryo as a thickening of the endoderm of the floor of the pharynx, at about 24 days. Here in Fig. 1 we see

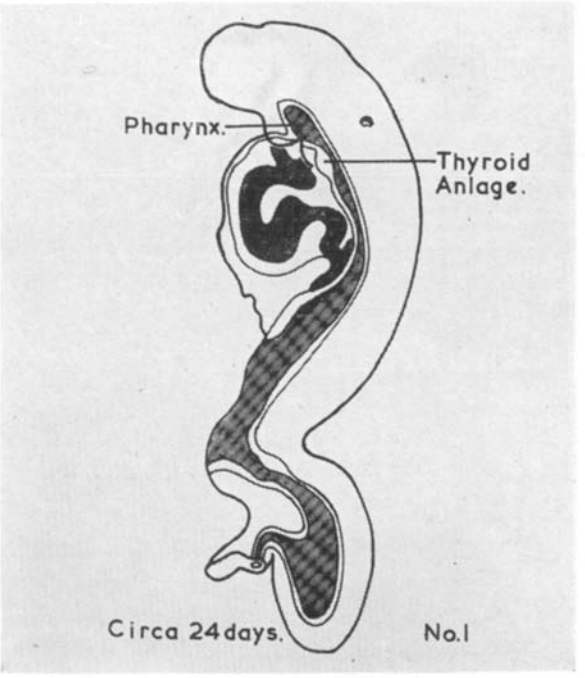


FIG. 1.

the thyroid anlage or precursor represented in white and lying above the primitive heart.

Here in Fig. 2 we see a median thyroid diverticulum arises from the floor of the pharynx, toward the close of the fourth week. In passing we note the relation of the thyroid diverticulum to the heart and aortic arches; the lung bud is also visible.

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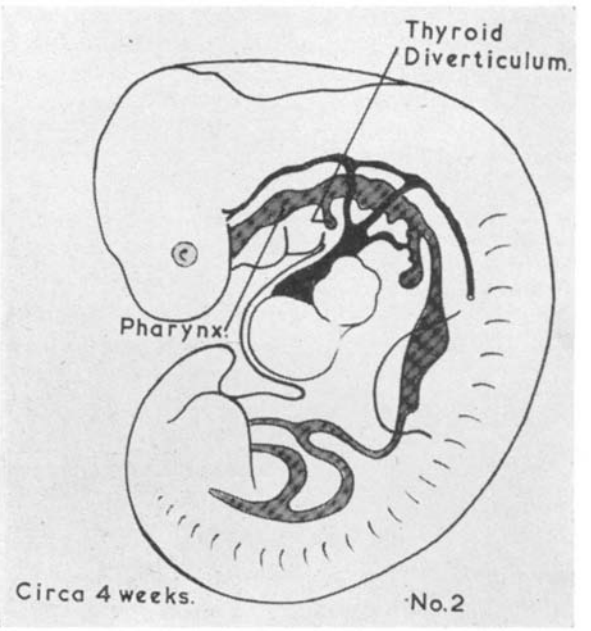


FIG. 2.

Lantern Slides 3 and 4

In Fig. 3 the thyroid diverticulum is seen to have migrated downward (caudally) below the pharynx and become bilobed. The stalk-like thyroglossal duct ruptures about this stage, namely at six weeks. Note the relation of the thyroid to the developing great vessels and the fact that there is as yet no neck. The lung bud is visible in this figure.

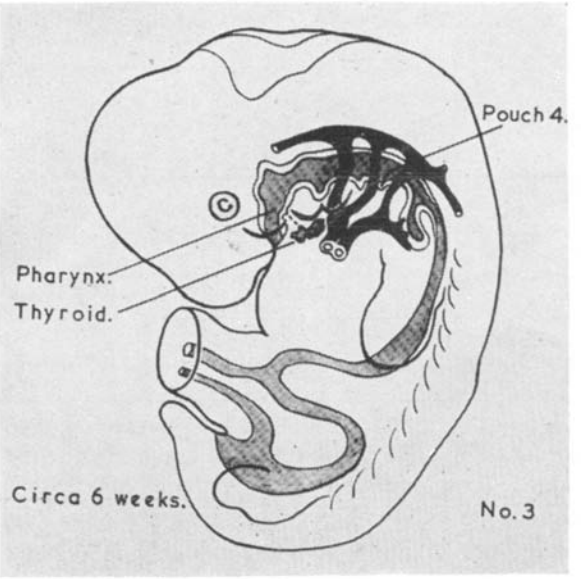


FIG. 3.

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Lingual Thyroid

Fig. 4 shows that with the development of the neck, and consequent elevation of the head, the thyroid separates from the heart and great vessels. The thyroid thus becomes draped across the trachea. The thyroglossal duct and hyoid bone are clearly seen; the great vessels depicted in Fig. 4 are derived from the aortic arches shown in Fig. 3.

Some believe that the thyroid also receives a contribution from the fourth pharyngeal pouch which ultimately unites with the median thyroid diverticulum. This is shown in Figs. 3 and 4, where we see the lateral thyroid primordium, indicated by pouch 4 in Fig. 3, being approximated to the main thyroid mass by virtue of the fact that the latter has been carried upward and backward

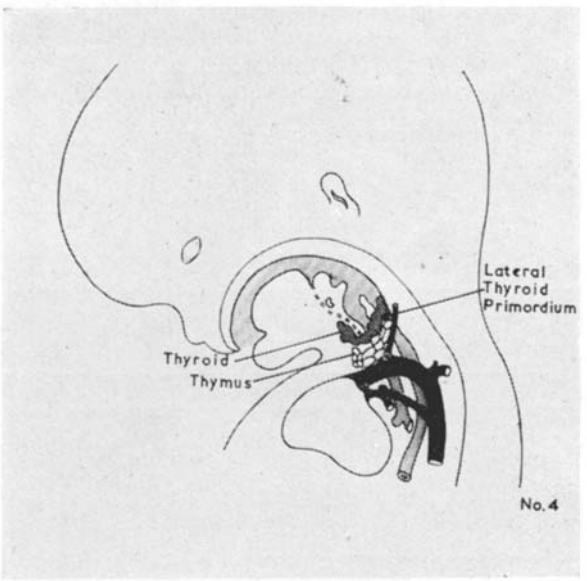


Fig. 4.

by the growth of the neck as seen in Fig. 4. In brief the median thyroid primordium is carried toward the lateral thyroid primordium by the growth processes attending the formation of the neck. Some controversy exists among embryologists as to the contribution afforded by the lateral thyroid primordium, but from the clinical viewpoint there appears to be little doubt that the lateral primordium may give rise to malignant change.

Lantern Slide 5

This diagram shows the possible sites that may be occupied by an aberrant thyroid; it also shows a thyroglossal cyst and the foramen caecum.

The nasopharyngeal and intra-oesophageal sites must be ascribed to the displacement of thyroid anlage cells into the dorsal wall of the primitive pharynx, with growth processes carrying the latter into the esophagus. The intratracheal site is similarly explained by thyroid cells being carried away from the thyroid anlage by the developing lung bud. The intra-thoracic site represents a persistence of juxta-cardiac embryonic conditions. The other sites epitomize the essential stages of thyroid development.

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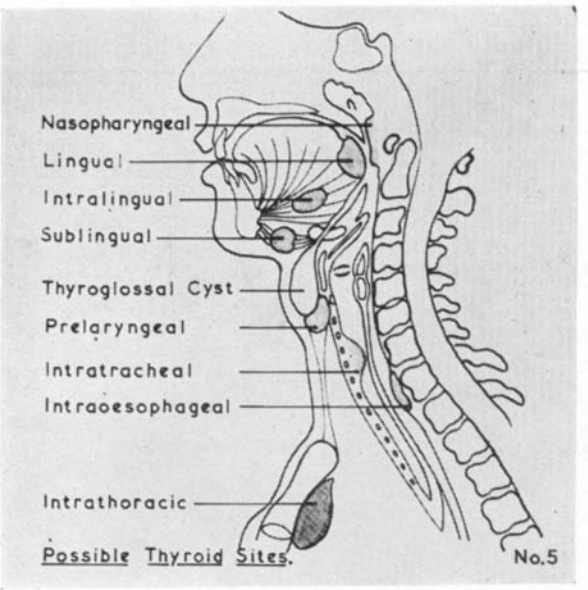


FIG. 5.

Lantern Slide 6

This last slide, Fig. 6, provides us with a synopsis of thyroid development. We see the median thyroid primordium arising from the ventral aspect of the embryonic pharynx and becoming bilobed as it descends. Finally on either side we see the fourth pharyngeal pouch giving rise to the lateral thyroid primordium.

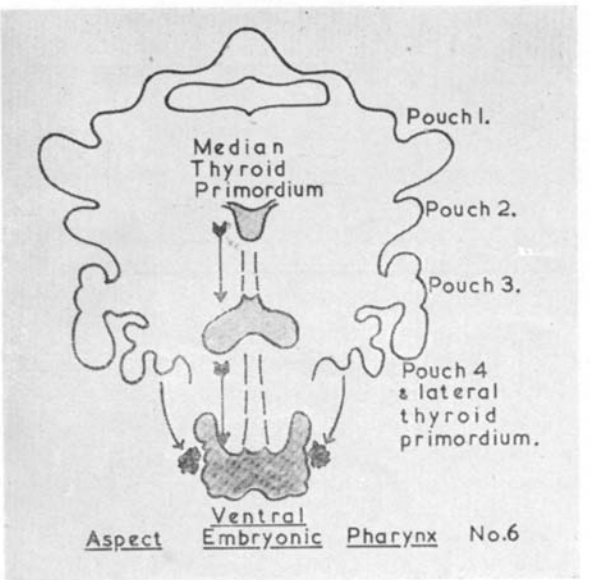


FIG. 6.

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