

An unusual cause of hypothyroidism (following excision of lingual thyroid) in a Chinese boy

by *J. S. Cheah*

MRACP
Senior Lecturer

and

F. K. Loh

MBBS
House Physician

} Department of Medicine (Unit 1),
University of Singapore

THE FREQUENCY OF lingual thyroid is very uncommon (Means, De Groot and Stanbury, 1963; Cheah et al., 1969). Lahey (1923) reported 2 cases in 7,800 cases treated surgically for thyroid diseases while Ulrich (1932) found only one in 4,000. In 70% of patients with lingual thyroid, the normally placed thyroid in the neck is absent (Montgomery, 1935 and 1936); in such instances, removal of the lingual thyroid would result in hypothyroidism. In this paper we report a Chinese boy who developed hypothyroidism following the excision of his lingual thyroid.

Case Report

This patient, O.P., a 17-year-old Chinese boy, was referred to us when he underwent a routine medical examination for National Service and was found to be stunted in growth and had no secondary sexual development. He is the sixth child in a family of 3 boys and 5 girls. His brothers attained puberty at about the age of 14 years. He left school at the age of 10 years because he did not do well in school.

In 1967, he was seen in hospital because of

irritation at the back of his throat. This was found to be due to a mass at the base of his tongue. This mass was removed and histology showed it was a lingual thyroid. He defaulted from follow up after the operation.

Clinical examination showed that his height was 57 inches (below the 3rd percentile) and his weight was 85 pounds (at the 25th percentile). Clinically, he was hypothyroid (dry and coarse skin, pulse rate of 60 per minute, delayed ankle jerks). There was early sexual development of the external genitalia but there was no axillary or pubic hair. He was of average intelligence and his blood pressure was 90/65 mm Hg. The other systems were normal.

Investigations showed that his basal metabolic rate (Dubois) was minus 36% (normal: $\pm 15\%$); the serum protein-bound iodine was 3.4 ug% (normal: 4 to 8 ug%); the resin uptake of triiodothyronine was 74% (normal: 75 to 115%); the serum total thyroxine was 1.4 ug% (normal: 3 to 7 ug%) and the free thyroxine index was 104 (normal: 225 to 805). A thyroid scintiscan showed that there was some uptake of radioiodine over the base of the tongue but none over the neck (see figure). This showed that there was some thyroid tissue at the base of the tongue and there was no thyroid at its usual site over the neck. The uptake of radioiodine over the tongue was 9.2% at 4 hours, 14.6% at 24 hours and 12.3% at 72 hours (normal: 15 to 50%). Thyroid antibodies (antithyroglobulin and antimicrosomal) were absent. The ankle reflex time (tap to half-relaxation) was 320 msec. (normal: 230 to 310 msec.). The electrocardiogram showed typical features of hypothyroidism. The bone age was 14 years. The chest



Anterior-posterior and lateral scintiscan showing concentration of radioiodine (^{131}I) at the base of the tongue. Note the absence of the thyroid gland in the neck.

X-ray was normal while X-ray of the skull showed that the pituitary fossa was on the large side of normality.

The patient was started on thyroxine replacement therapy (0.1 mg thrice daily) and he has progressed well.

Discussion

The usual causes of childhood and juvenile hypothyroidism include dysgenesis of the thyroid gland, deficiency of iodine, dyshormonogenesis, ingestion of goitrogens, primary thyroid diseases and pituitary disorders (Hutchinson, 1969; Means, De Groot and Stanbury, 1963). Juvenile hypothyroidism due to excision of a lingual thyroid as is seen in our patient is very rare as there are about 200 cases of lingual thyroid reported in the literature (Cheah et al., 1969).

In our patient, like 70% of patients with lingual thyroid, the normally placed thyroid is absent (Montgomery, 1935, 1936). In such instances, as this patient illustrates, removal of the lingual thyroid results in hypothyroidism. In 15% of cases of lingual thyroid, spontaneous hypothyroidism has been reported (Goetsch, 1948). This suggests that the function of an ectopically placed thyroid gland is not as efficient as the normally placed gland.

Our patient has retarded physical, gonadal and skeletal growth. These are the hallmarks of hypothyroidism in children (Hubble, 1956). Although delayed puberty is often found in hypothyroidism as in our patient, precocious puberty has also been reported (Hubble, 1969).

Thyroid antibodies are absent in this patient as was in the case previously reported (Cheah et al., 1969). The occurrence of thyroid antibodies in lingual thyroids has not been previously reported (Means, De Groot and Stanbury, 1963; Cheah et al., 1969).

Like other thyroid disorders, lingual thyroid is commoner in females; about 75% of cases occur in females (Schilling et al, 1950). Our first patient was also a female (Cheah et al., 1969); the present patient is a male.

Irritation of the throat was the symptom that brought the present patient to have his lingual thyroid detected and removed. The symptomatology and treatment of lingual thyroid have been discussed in detail previously (Cheah et al., 1969).

Summary

A 17-year-old Chinese boy with hypothyroidism due to excision of a lingual thyroid five years previously is described. He has delayed physical,

AN UNUSUAL CAUSE OF HYPOTHYROIDISM

gonadal and skeletal growth. There is still some thyroid tissue in the tongue but there is no thyroid tissue in the neck. The clinical features and treatment of lingual thyroid are discussed. The literature on lingual thyroid is briefly reviewed.

References

- Cheah, J.S., Tan, B.Y., Goh, E.H. and Chew, C.H.: "Lingual thyroid: report of a case and review of the literature", *Med. J. Aust.*, 2, 403, 1969.
- Goetsch, E.: "Lingual goiter: report of 3 cases", *Ann. Surg.*, 127, 291, 1948.
- Hubble, D.: "Thyroid disorders in childhood", *Brit. Med. J.*, 1, 875, 1956.
- Hubble, D.: "Paediatric endocrinology", edited by Hubble, D., 1st. ed., Blackwell Scientific Publications, Oxford and Edinburgh, 343, 1969.
- Hutchinson, J.H.: in "Paediatric endocrinology" edited by Hubble, D., 1st. ed., Blackwell Scientific Publications, Oxford and Edinburgh, 119, 1969.
- Lahey, F.H.: "Lingual goitre", *Surg. Gynae. Obstet.*, 36, 393, 1964.
- Means, J.H., De Groot, C.J. and Stanbury, J.B.: "The Thyroid and its Diseases", 3rd. ed., McGraw-Hill, New York, 526, 354, 1963.
- Montgomery, M.L.: "Lingual thyroid: comprehensive review", *West. J. Surg.*, 43, 661, 1935.
- Montgomery, M.L.: "Lingual thyroid: comprehensive review", *West. J. Surg.*, 44, 54, 122, 189, 237, 303, 373, 442, 1936.
- Schilling, J.A., Karr, J.W. and Hursh, J.B.: "The treatment of a lingual thyroid with radioactive iodine", *Surgery*, 27, 130, 1950.
-