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A Case of Subhyoidal Median Ectopic Thyroid Associated with Lingual Thyroid

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A case of subhyoidal median ectopic thyroid associated with lingual thyroid was observed in a 14-year-old girl. The subhyoidal median ectopic thyroid was excised for cosmetic reason.

The postoperative course has been satisfactory without hypertrophy of the lingual thyroid due to periodic administration of triiodothyronine.

INTRODUCTION

The thyroid is often involved by abnormal development such as thyroglossal duct or cyst, pyramidal lobe, or ectopic thyroid gland. In recent years, many cases of abnormal thyroid have been found due to application of I^{131} to the diagnosis of thyroid gland. Recently, we experienced a case of subhyoidal median ectopic thyroid associated with lingual thyroid. Since such a case is extremely rare, it is herein reported together with litarature observation.

CASE REPORT

On 24 July 1968, a 13-year-old junior high school girl visited the clinic with the chief complaint of a mass in the anterior neck. Her family history or past history was not remarkable.

At the age of 8, a mass of the size of the tip of the thumb was noted on the thyroid bone in the median line of the anterior neck. The mass gradually increased in size and became walnutsize at the age 12. On 5 August 1967, the mass removed but partially because of accident. The mass was parenchymatous and microscopic examination revealed normal thyroid tissue.

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About half a year after the operation, a swelling was noted again at the same site and became walnut-sized when the patient visited the clinic for the second time.

Clinical findings

The patient was a girl with moderate structure and good nutrition. The pulse rate was 72 without irregularity but with good tension. The blood pressure was 110/70 mmHg.

No abnormal findings was noted in the thoracic and abdominal regions. There was no symptom of hyperthyroidism such as heart acceleration, hyperhydrosis, exophthalmus or tremor of fingers.

A swelling measuring $4\times3\times1$ cm was noted in the anterior neck but no reddening in the same region. (Photo. 1.)

The mass was not adhered to the skin and showed movement at the time of swallowing but was not tender. It was elastic soft in consistence without fluctuation. The pharynx and oral cavity were not remarkable. No lymph node was palpated in the neck and submaxillary region.

Laboratory tests :

The findings of stool and urine tests were normal.

Hematological test revealed RBC 422×10^4, WBC 6000, Hct 41%, Hgb 13.4 g/dl. Biochemical test of blood revealed icterus index 5.0, Kunkel 6.5, TTT 3.5, Alkaline P-ase 7.5 PH/h, S-GOT 27 SFU, S-GPT 22 SFU, serum protein 7.1 g/dl, A/G ratio 1.15, and negative

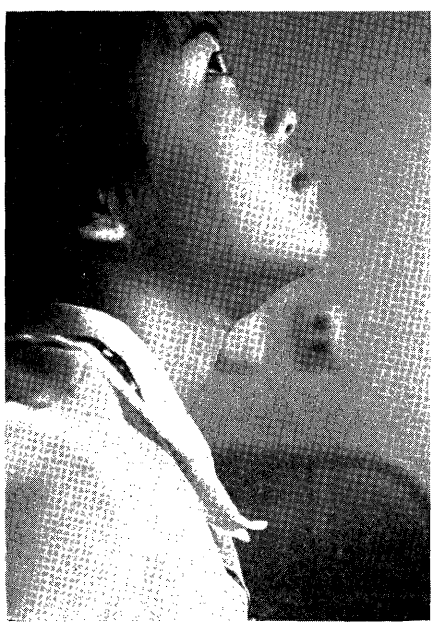

Photo. 1.

serological reaction. ECG was normal.

Thyroid function test revealed basal metabolism 2%, triosorb resin uptake 30.3%, and thyroid radioiodine (I^{131}) uptake 8.1% (24h). (Photo. 2.)

Thyroid scintigram indicated a thyroid gland in conformity with the mass in the anterior portion of neck and another thyroid tissue nearby but none in the normal portion. (Photo. 3.)

Accordingly it was known that the mass in the anterior portion of the neck was an aberrant thyroid gland in front of the hyoid bone

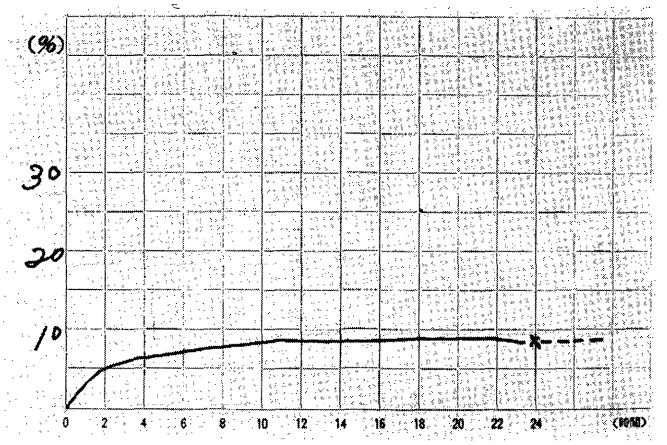

Photo. 2.

Photo. 3.

as observed in the previous examination, and that there was another thyroid gland at the base of the tongue. The former was removed surgically on 19 August 1968.

Operative findings:

The skin was cut approximately 4 cm in length at the upper end of the thyroid cartilage. The mass was located in front of the hyoid bone and was partially adhered in slight degree to the hyoid bone but it was easily detached and removed.

Histological findings:

The mass was dark reddish in color mesuring 3.5×2.5×2.0 cm in size and 10 gm. in weight. (Photo. 4.)

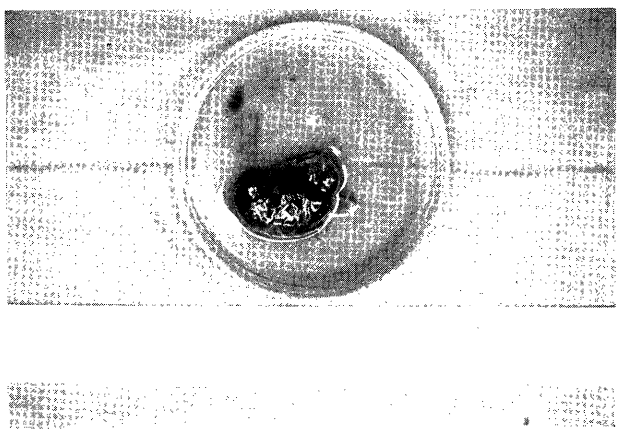

Photo. 4.

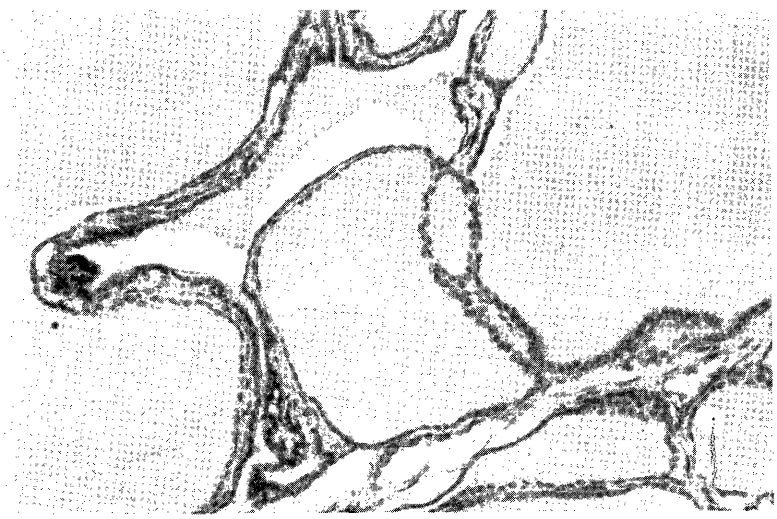

Photo. 5.

The surface was smooth and the cut surface was parenchymatous.

Microscopic examination revealed that the mass was normal thyroid tissue. (Photo. 5.)

Postoperative progress:

Following the operation, the patient was administered every other day with 30 mg. of triiodothyronine. The basal metabolism was −12% on the 28 th day after operation but 4% on the 63 rd day and thus there was no sign of depression in thyroid function.

ECG was also normal. Thyroid scintigram indicated no other tissue than that at the base of the tongue, which was not enlarged.

DISCUSSION

The thyroid develops as a downgrowth from the region of the primitive pharynx. A mass forms at the base of the tongue and extends downward as a long tube, the thyroglossal duct, its final position being in front of the trachea and thyroid cartilage.

The upper end of the thyroglossal duct is marked by a small depression at the root of the tongue, the foramen caecum.

The elongated duct losses its lumen, is transformed into a cord, and normally is obliterated during fetal life, beginning at about the fifth week.

The most frequent abnormal development of the thyroid is persistent duct or cyst which is often observed immediately under the hyoid bone. The residue of the thyroglossal duct sometimes contains thyroid tissue on the wall[1].

STAHL[2] reports 3 out of 90 cases and WARD[3] and Hendrick 1 out of 105 cases. Midline ectopic thyroid tissue caused by failure in descent is located between the tongue and the diaphragm.

The location and symptoms of ectopic thyroid tissue have been reported by FISH et al.[4] as shown Table 1.

Teble 1. Ectopic thyroid tissue; Location and Symptomatology

Location	Symptoms
Lingual	Dysphagia, bleeding, dyspnea
Suprahyoid, infrahyoid	Midline mass in neck
Thyroglossal duct, cyst	None, or midline mass
Pyramidal lobe	Usually none
Intratracheal, intralaryngeal	Respiratory obstruction
Intraesophageal	Dysphagia
Aortic, pericardial, cardiac	None

There have been 40 reported cases of lingual thyroid in Japan[5] and the addition of the author's case results in a total of 41 cases.

Lingual thyroid was first reported by HICKMAN in 1869 and a detailed study was made on 144 cases by MONTGOMERY[6] in 1936.

Since then, fairly numerous cases have been due to the application of radioactive iodine method.

Symptoms are usually slight and hardly subjective except for abnormal pronounciation, sensation of alien substance, bleeding and dysphagia which are occasionally complained.

BAKER[7] experienced a case of bleeding so heavy as to be misunderstood as gastric ulcer. In cases of lingual thyroid, a proper thyroid are often absent. MONTGOMERY[6] reported that approximately 75% of the cases had no thyroid in its normal position.

In the author's case, the proper thyroid was not found but two thyroids were each located both on and under the hyoid bone.

Reported cases with subhyoid median ectopic thyroid gland are very few in comparison with those of lingual thyroid.

In Japan, the one reported by TANAKA[5] et al. is the only reported case. Even in european countries, there is no other than the 19 cases[8-15] reported. However, if the cases in which the residue of thyroglossal duct or cyst contains thyroid tissue on the wall are included, the number of cases increases remarkably.

The median ectopic thyroid gland hardly has any subjective symptom and the mass in the anterior neck is the only symptom in most cases. Accordingly, it is difficult to give a proper diagnosis before operation. QUIGLEY[9] states that 8 cases have been operated under the diagnosis of thyroglossal cystoma.

Though it is a matter of course in the application of radioactive iodine method is helpful in the establishment of diagnosis, distinction from the thyroglossal cyst with thyroid tissue on the wall is difficult.

Cases of median ectopic thyroid associated with lingual thyroid are very rare being reported only NACHLUS[16], TANAKA et al.[5] and MEYEROWITZ et al.[15]. The case reported by NACHLUS was a 36-year-old woman who had lingual thyroid and thyroid tissue in the thyroglossal cyst. The one reported by TANAKA et al.[5] was a 14-year-old girl who had aberrant thyroid glands at the base of the tongue and both on and under the hyoid bone.

MEYEROWITZ[15] et al. reported a 8-year-old girl who had ectopic thyroid associated with lingual thyroid tissue in the neck.

The value of BMR was −21% in the first case, 1% in the second case and unknown in the third case. In the author's case the value was −2%. In general, the value of BMR shows trend to decrease when the aberrant thyroid gland is present.

When the thyroid gland is absent in its proper position, its entire functions are performed by the aberrant thyroid gland.

Aberrant thyroid gland are after removed for cosmetic reason or because of dysphagia or dyspnea. However, since perform the entire function of the proper thyroid as stated above, it is not rare that mucous edema or other disorders are caused as result of the removal [3)4)16)18)]. Because of this, thyroid gland is sometimes attempted so as to eliminate such troubles[17)].

In the present case, only the subhyoidal median ectopic thyroid was removed while leaving the lingual thyroid. Following the operation, 30 mg. of triiodothyronine has been administered every other day to avoid compensatory hypertrophy of the lingual thyroid.

The postoperative course has been satisfactory without any thyroid deficiency symptom and hypertrophy of the lingual thyroid.

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