

Case Report

Subhyoid ectopic thyroid with a solitary colloid nodule in a 10-year-old girl: a rare diagnostic entity

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ABSTRACT

Ectopic thyroid is a rare clinical entity, of which its presence in a subhyoid location is uncommon. It is equally rare to find a solitary thyroid nodule in the paediatric age group. We report a case of subhyoid ectopic thyroid in a 10-year-old girl, and its course over a span of 3 years. It was associated with a solitary colloid nodule, which eventually became symptomatic and did not respond to suppressive levothyroxine therapy. Hence surgical excision was done, and the child was put on replacement doses of levothyroxine.

Keywords: Colloid nodule, Ectopic thyroid, Subhyoid ectopic thyroid, Solitary thyroid nodule

INTRODUCTION

Aberrations in the embryogenesis of the thyroid gland during its migration from the floor of the primitive foregut to its final pre-tracheal position results in an ectopic thyroid.¹ The overall prevalence reported of this condition is 1 per 100,000 -300,000.^{1,2} Moreover, thyroid nodules are less frequent in children, and recent studies based on ultrasound evaluation in the paediatric age group have detected a prevalence ranging from 0.2 to 5.1%.³ Current literature suggests that the incidence of malignancy in paediatric thyroid nodules is 25% compared to 5% in adults.^{3,4} We report a case of an ectopic thyroid gland, which was mainly subhyoid in location with a colloid nodule in a 7-year-old girl on presentation and its course over three years, which posed a unique diagnostic challenge.

CASE REPORT

The case described is of a girl who presented to the paediatric endocrine clinic initially at the age of 7 years with a midline neck swelling noticed since the last one-

year. There was no history of pain, dysphagia or hoarseness of voice. There was no radiation exposure or family history of similar complaints.

Her weight was 19kg (z-score-0.87) and height was 114cm (z-score -1.09). She was growing according to her target range. On examination, a midline neck swelling measuring 2.5 x 2.5cm was seen which was non-tender with a smooth surface. It was not moving with deglutition or protrusion of the tongue. There was no cervical lymphadenopathy. Tanner's stage was prepubertal and her systemic examination was normal. She was investigated and her thyroid function tests were within normal limits - TSH-4.62µIU/ml (Normal-0.60-6.30µIU/ml), T₄- 6.97µg/dl (Normal-6.40-13.30µg/dl). Antithyroid antibodies were negative. Ultrasound neck showed a well-defined hypoechoic tissue measuring 2.8 x 2.2 x 0.8cm in the midline below the hyoid bone within which a nodule measuring 2.1 x 0.9 x 2.8cm was noted which was partly cystic and partly solid with minimally increased vascularity and no calcifications. The thyroid fossa was empty [Figure 1 (A)]. ^{99m}Tc pertechnetate scan showed an ectopic thyroid at the subhyoid level with no uptake noted in the region of the

thyroid [Figure 1 (B)]. The findings were confirmed by computed tomography scan of the neck [Figure 1 (C)].

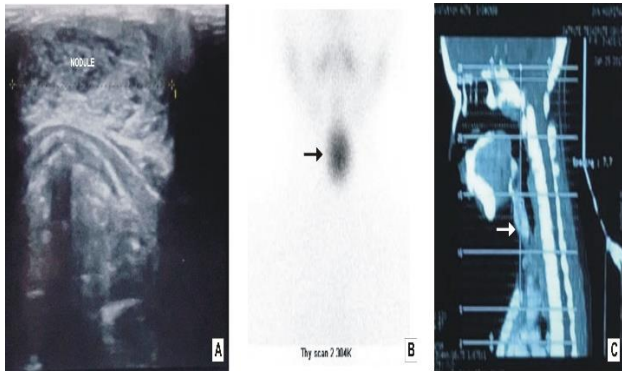


Figure 1: (A): Ultrasonography of neck showing a hypoechoic mass in the subhyoid region with a heterogeneous nodule (B): ^{99m}Tc pertechnetate thyroid scan showing an area of increased uptake in the subhyoid region with no uptake in the region of the thyroid fossa (C): Sagittal computed tomography of the neck showing a heterogeneously enhancing mass predominantly in the subhyoid region.

Fine Needle Aspiration Cytology of the swelling revealed thyroid follicular cells filled with colloid with no evidence of atypical or malignant cells. She was started on Tablet levothyroxine for 6 months, however on follow up it was noted that the swelling increased in size and repeat ultrasound after one year showed an increase in size to 3.5 x 2.8cm. She however remained euthyroid. Serum thyroglobulin levels were high (106ng/dl Normal - 1.6-60ng/dl). Patient was lost to follow up and presented again after around one year at the age of 10 years with failure to gain adequate height. Her growth velocity was 3cm in the last 1 year. The swelling had increased to 3.8cm x 2.8cm and she additionally had complaints of discomfort while swallowing since the last 3 months. She had Tanner's stage 2 of pubertal development. A repeat thyroid function test suggested hypothyroidism (TSH-22μIU/ml, T4-4.2μgm/ml) and was thus started on levothyroxine replacement. After parental informed decision and consent, it was decided to surgically remove the thyroid nodule in view of increase in size of the nodule despite medical management, discomfort during swallowing and an increased risk of malignant transformation of the nodule in this age group. After ensuring a euthyroid status, surgical extirpation was done. Intraoperatively, a nodule was present which was predominantly subhyoid. It also had adhesions to the hyoid bone and a small suprahyoid extension was noted as well. A well-encapsulated greyish brown mass was seen measuring 4.5 x 3.0 x 1.5cm with a few congested areas. The thyroid fossa was empty. The entire nodule along with the median part of the hyoid bone was removed (Figure 2).

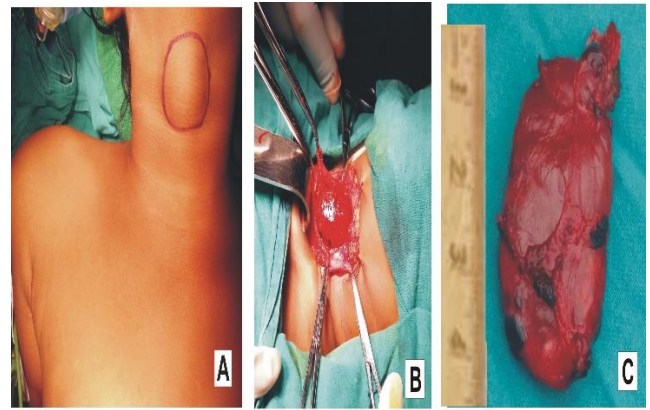


Figure 2: (A): a preoperative midline neck mass (B): intraoperative picture showing a predominant subhyoid nodule (C): postoperative specimen with a few congested areas.

Histopathological examination (Figure 3) showed a thyroid tissue with varying sized thyroid follicles filled with colloid. There were also a few areas of fibrosis and calcification seen.

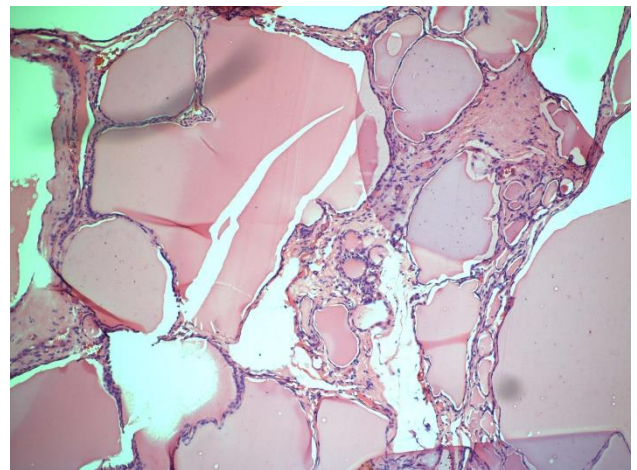


Figure 3: Histopathological slide showing colloid filled follicles.

Postoperatively, her symptoms of discomfort while swallowing were relieved. She is currently on replacement doses of levothyroxine, maintaining a euthyroid status, has not shown any recurrence in the last 12 months and has documented a normal growth velocity.

DISCUSSION

The presence of thyroid tissue anywhere other than the anterior neck region between the 2nd and 4th thyroid cartilages is called ectopic thyroid.⁵ It is the most common form of thyroid dysgenesis accounting for 48-61% of the cases.⁶ Embryologically, during the third or fourth week of gestation, an endodermal diverticulum from the median plate of the floor of the pharyngeal gut is formed. This diverticulum descends in the midline

beginning at the 24th day of gestation to its final pretracheal position, resulting in the formation of a thyroglossal duct.¹ A number of transcription factors play a key role in morphogenesis of the thyroid, of which FOXE1 is required for thyroid migration.⁶ Failure of this migration results in the formation of ectopic thyroid tissue.

Lingual thyroid is the most common type accounting for 90% of cases, whereas the sublingual types which are suprahyoid, subhyoid or at the level of the hyoid bone are less frequently encountered.⁷ Our case described a rare occurrence of a midline neck swelling which was mainly subhyoid in location with an unusual occurrence of a thyroid nodule. A multicentric study on 120 paediatric patients with thyroid nodules not associated with risk factors such as autoimmune thyroid disease or radiotherapy revealed occurrence of thyroid carcinoma in 16% of cases, of which 12% were papillary, 2.5% follicular and 1.7% medullary. Benign tumours accounted for around 10% of all nodules as was also seen in our case.⁸

In a study reported by Corris A and group, 78 euthyroid children and adolescents were evaluated to assess the effectiveness of levothyroxine therapy in benign thyroid nodules. They found a reduction in nodule diameter from 2.24 ± 0.94 to 1.86 ± 1.17 cm ($p=0.039$) in treated patients as well as a decrease in symptoms as compared to untreated patients. However, our case did not show a reduction in size or symptoms despite a trial of 6 months of levothyroxine as mentioned above. In fact, as seen here, an increase in nodule size during ultrasound follow-up, particularly during a trial with suppressive levothyroxine therapy has also been advocated as an index of malignancy.^{8,9}

For the category of children who have a benign cytopathology, surgery may be considered due to increasing size, compressive symptoms, cosmetic reasons or patient's choice.¹⁰

CONCLUSION

A differential diagnosis of ectopic thyroid should be considered in all cases of a midline neck swelling. Thyroid imaging and scintigraphy are essential to diagnose ectopic thyroid. A close observation of the possibility of a nodule in the thyroid should be carefully looked for during ultrasonography, as well as the high prevalence of malignant transformation in the paediatric age group should be kept in mind, as described in our case. Careful monitoring for height velocity, size of the swelling, associated symptoms, as well as response to medical management should be assessed regularly, so

that surgical intervention can be done at an appropriate time if required as in the case described.

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