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Clinical and morphological characteristics of ectopic thyroid gland in children: A series of 24 patients from Northwest India

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ABSTRACT

Background: Most adults with ectopic thyroid gland (ETG) are asymptomatic at the time of detection of ETG. Unlike adults, the clinical and morphological spectrum of ETG in children remains less characterized. **Methods:** We retrospectively analyzed data of children with primary congenital hypothyroidism (CH) due to ETG who attended the pediatric endocrinology clinic of our hospital between April 2004 and March 2016. The diagnosis of ETG was based on combined scanning with technetium-99m (99mTc) pertechnetate thyroid scintigraphy and high-resolution thyroid ultrasonography. **Results:** Twenty-four (18.7%) of the 128 children with primary CH were identified to have ETG. Their mean age at diagnosis was 64.3 ± 47 months (range, 0.5–144 months); 17 (70.8%) were girls. The mode of diagnosis was based on symptoms in 20, newborn screening in 3 and incidental in 1. Symptoms included poor weight gain in 13 (54%) developmental delay in 9 (37.5%), constipation in 8 (33%), decreased activity in 6, excessive sleepiness in 5, anorexia in 1 and decreased sweating in 1. The anatomical location of ETG was lingual in 17 (71%) and sublingual in 5 (21%). Thyroid function tests showed overt hypothyroidism in 16 and subclinical in 8. The mean duration of follow up was 16 ± 27.5 months (range 0.5–114 months). **Conclusion:** We found symptomatic hypothyroidism to be common at presentation in our children with ETG. This is the largest series of children with ETG based on the diagnosis by combined scanning.

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Introduction

Thyroid gland develops from the median thyroid anlage that appears on the floor of pharynx on day 20–22 of fetal development and starts migrating caudally by day 26 to reach its final pretracheal location by day 48–50 of development. Aberrations in embryogenesis during the delicate migration process may result in presence of ectopic thyroid gland (ETG) at locations other than its normal pretracheal position [1]. The usual prevalence of ETG is approximately 1 per 100 000–300 000 persons [2]. Worldwide, ETG is considered to be the most frequent form of thyroid dysgenesis (TD) accounting for 48–61% of all patients with TD [1–3]. However, recent reports indicate that ETG is not the dominant form of TD in certain patient populations of CH [4–7]. The reasons for differences in spectrum of TD are however unknown at present. Similarly, the reasons for a marked sexual dimorphism in ETG with a female preponderance (almost 80% cases occur in females) are not fully known [1–3, 8–10]. Of the different forms of ETG, the lingual location is the most common followed by supra and infra hyoid locations [2, 8, 9]. ETG due to developmental aberration is occasionally found in locations outside the neck area [2]. Some recent reports indicate a different location spectrum with lingual location as one of the least common [10, 11]. All the previous studies have been conducted in adult patient populations often including a small number of children in their cohorts. The clinical and morphological spectrum of ETG in children, therefore, remains largely understudied.

The investigation of choice for detection of ETG is combined scanning using high-resolution ultrasonography and technetium-99m (99mTc) pertechnetate thyroid scintigraphy [12]. Other investigation modalities such as plain CT and magnetic resonance imaging have also been used successfully to localize ETG (10). Most of the previous series used a single imaging modality for confirmation of ETG [2, 9, 10]. We have been using combined scanning for morphological characterization of TD in a large population of pediatric patients with CH since 2004. With an aim to study the clinical and morphological spectrum of ETG, we analyzed the clinic data of these patients.

Materials and methods

A retrospective analysis of data extracted from files of children with primary CH due to ETG who attended the pediatric endocrinology clinic of our hospital between April 2004 and March 2016 was performed. The data included age at diagnosis, sex, mode of diagnosis, clinical features, thyroid function tests, anti-thyroid peroxidase antibodies, imaging studies and treatment details. Children with incomplete data or syndromic diagnoses were excluded from analysis. The diagnosis of hypothyroidism was based on low serum total thyroxine (T4), and elevated serum thyroid stimulating hormone (TSH) levels according to reference ranges [13, 14]. Serum total T4, TSH and anti-thyroid peroxidase (anti-TPO) antibodies concentrations were measured by Electrochemiluminescence immunoassay on Elecsys 2010 analyzer using specific kits (Roche Diagnostics, Germany).

The diagnosis of ETG was based on the results of combined imaging with technetium-99m (99mTc) pertechnetate thyroid scintigraphy and thyroid ultrasonography done routinely at initial evaluation of CH. Thyroid scintigraphy was performed using a gamma camera fitted with low energy high-resolution collimator (Siemens, Germany). Static planar images of head, neck and chest region were acquired in the anterior projection 20 min after intravenous injection of 74–111 MBq of 99mTc pertechnetate. A focal tracer uptake in midline from tongue to the suprasternal notch in the absence of eutopic gland was identified as ETG. Ultrasonography was done using ultrasound machine equipped with a 3–12 MHz high frequency linear transducer, 3–8 MHz sector array and 2–5 MHz convex array probes (Philips HD11XE). Images were obtained in transverse and longitudinal planes and anterior cervical area was systematically viewed for presence of ETG. Absence of gland in the normal location and presence of any thyroid tissue in the midline was labeled as ETG. The thyroid imaging modalities were used as suggested by the guidelines of the European Society for Pediatric Endocrinology [14].

Results

A total of 253 children with primary CH were registered in the clinic during the 12-year study period; complete information was available for only 128. Twenty-four (18.7%) identified as ETG were further analyzed. The mean age at diagnosis was 64.3 ± 47 months (range, 0.5–144 months); 17 (70.8%) were girls. The mode of diagnosis was based on symptoms in 20, newborn screening in 3 and incidental in 1. In 15 children, the parents could recollect the age at onset of symptoms; during infancy in 6, 1–5 years of age in 5 and after 5 years in 4 children. Poor weight gain was the most common parental complaint noted in 13 (54%) children followed by developmental delay in 9 (37.5%) and constipation in 8 (33%) children. Other symptoms included decreased activity in 6, excessive sleepiness in 5, anorexia in 1 and decreased sweating in 1. Physical examination revealed growth retardation in the majority (15, 62.5%) of children. Goiter was noted in 1 child. Four children had abdominal distension while umbilical hernia, hypotonia, small head size, open posterior fontanellae were seen in 2 each and pseudo muscular hypertrophy in 1 child. Maternal hypothyroidism was noted in 4 cases.

The gland location was lingual in 17 (71%) and sublingual in 5 (21%). The sublingual location was at the suprasternal notch, thyroglossal cyst, submental, anterior cervical region and posterior cervical region along C5-C7 vertebrae in one patient each. Two children had dual ectopia; 1 of these had ectopic tissue in both lingual and hyoid regions, and the other in lingual region and thyroglossal cyst (Fig. 1). Eight of the 17 children with lingual thyroid had additional thyroid tissue; 7 showed dysplastic or non-functional additional tissue in the eutopic location and 1 showed thyroid tissue in the thyroglossal cyst.

Thyroid function tests showed subclinical hypothyroidism in 8 and overt hypothyroidism in 16 patients. The

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