Parathyroid adenoma on the ipsilateral side of thyroid hemiagenesis

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We present a case of a parathyroid adenoma on the ipsilateral side of thyroid hemiagenesis—which, to our knowledge, is the third reported case of this entity. A 41-year-old man with nephrolithiasis was found to have elevated calcium and intact parathyroid hormone levels. Both ultrasound and technetium sestamibi scintigraphy with single photon emission computed tomography confirmed left thyroid hemiagenesis and an adenoma in the left inferior thyroid bed. The patient underwent left neck exploration, which confirmed left thyroid hemiagenesis and a left inferior parathyroid adenoma. The left inferior parathyroid gland was resected. The patient was discharged home the same day of surgery and has remained normocalcemic for 14 months without evidence of hyperparathyroidism.

Thyroid hemiagenesis is a very rare anomaly. Dr. C. Handfield Jones, a British physician and histologist, first described this anomaly in 1852 (1). Since then, very few cases of hemiagenesis as well as adenomas associated with hemiagenesis have been reported. We describe a case of a parathyroid adenoma on the ipsilateral side of thyroid hemiagenesis, which has been previously reported only twice (2, 3).

CASE REPORT

A 41-year-old man with a history of nephrolithiasis and fatigue and no prior thyroid surgery was found to have a calcium level of 11.2 mg/dL (normal, 8.5–10.1 mg/dL) and an intact parathyroid hormone level of 79 pg/mL. Examination revealed no masses in the neck. The left thyroid lobe could not be palpated. Ultrasound showed the absence of the left thyroid lobe and two hypoechoic nodules on the left side, one measuring 1.3 cm in the thyroid bed and one measuring 1 cm inferior to the thyroid bed. The right thyroid lobe was present and appeared normal with the exception of a 3-mm nodule. Technetium sestamibi scintigraphy with single photon emission computed tomography showed the absence of the left lobe and a focus of increased tracer accumulation in the left neck just inferior to the inferior margin of the left lobe. A second small focus with only minimal tracer accumulation was identified in the left supraclavicular region.

The left side of the neck was operatively explored. The preincision parathyroid hormone level was 106 pg/dL. There was no evidence of a left thyroid lobe, and a discrete nodule was seen inferiorly with a tongue of tissue extending into the thryothymic track. The adenoma was excised. No other nodules were seen in the left side of the neck. The 5- and 10-minute postexcision parathyroid hormone levels were 28 and 7 pg/dL, respectively. Histologically, the parathyroid gland, which weighed 1.308 grams, was hypercellular with thymic tissue attached to its inferior portion *(Figures 1 and 2)*. The patient was discharged home the same day of surgery in improved condition, and he has remained normocalcemic for 14 months with no symptoms of hyperparathyroidism since surgery.

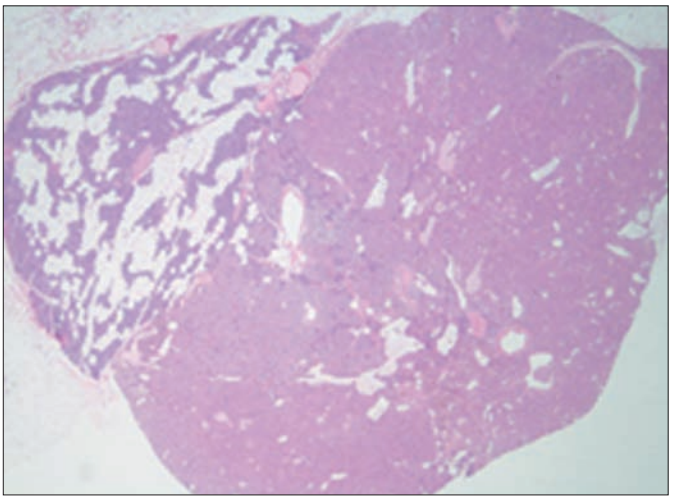

Figure 1. Low-power view shows a well-defined mass (at right) surrounded by adipose tissue and normal parathyroid glandular tissue (left).

DISCUSSION

Few cases of thyroid hemiagenesis have been described since Jones' report in 1852. Mikosch et al found 256 cases reported up to 1999 (4). The diagnosis is usually incidentally made in patients with contralateral thyroid abnormalities. In 1895, Marshall found

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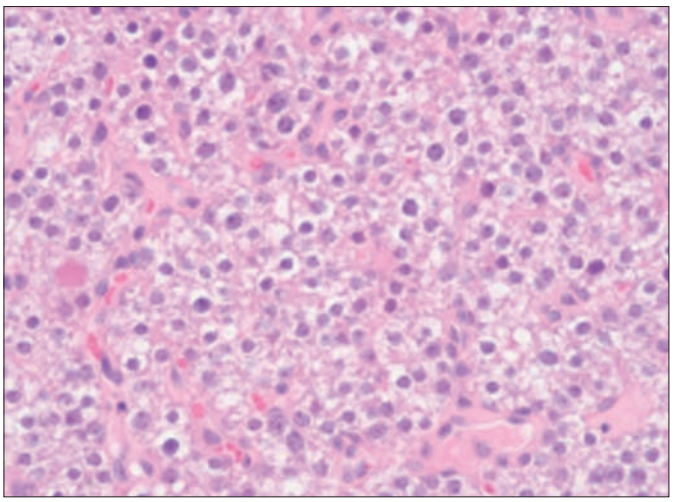

Figure 2. High-power view shows a solid growth of chief cells with a delicate capillary network and no adipose tissue within the adenoma. A few microfollicles are present, resembling thyroid follicles.

one case in 60 pediatric autopsies (5). Harada et al reported no cases in 1007 autopsies in 1972 (6). Melnick and Stemkowski performed 300 autopsies and found 15 cases of hemiagenesis, reporting a 3:1 female predominance and a 4:11 predominance of left over right lobe involvement (7). Russotto and Boyar reported an incidence of one in 1700 cases using an I-131 scan for detection (8). Hamburger and Hamburger discovered an incidence of 4 in 7000 patients using a thyroid scan (9).

The cause of thyroid hemiagenesis is unknown, but most likely the thyroid diverticulum fails to divide into separate lobes. The thyroid diverticulum results from the invagination of a median endodermal thickening in the floor of the primitive pharynx. Normally, this diverticulum divides into two lobes and passes ventral to the laryngeal cartilage and hyoid bone. The thyroid diverticulum is connected to the tongue by the thyroglossal duct, and the thyroid gland reaches its final resting place by the seventh week of gestation. Embryologically, parathyroid glands develop from a separate area, the dorsal epithelium of the third and fourth pharyngeal pouches. The inferior parathyroid gland, from the third pharyngeal pouch, migrates with the thymus caudally to lie on the dorsal surface of the inferior aspect of the thyroid gland. The superior parathyroid gland, from the fourth pharyngeal pouch, migrates very little before it rests on the dorsal and superior aspect of the thyroid gland. This explains why parathyroid adenomas can be found in association with thyroid hemiagenesis in an anatomically normal position (10).

Only two other reports of a parathyroid adenoma on the ipsilateral side of thyroid hemiagenesis have appeared. Maganini and Narendran were the first to report a left inferior parathyroid adenoma in a patient with left thyroid hemiagenesis in 1977 (2). In 1992, Woods and Loury reported a left superior parathyroid adenoma in a patient with left thyroid hemiagenesis (3). One case of an adenoma on the contralateral side of thyroid hemiagenesis was reported by Sakurai et al in 2007 (11). Although a very rare occurrence, this is an interesting case of a left inferior parathyroid adenoma associated with left thyroid hemiagenesis.

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