

# A Rare Case of Bleeding Ectopic Lingual Thyroid Presenting as Hematemesis

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To the Editor:

Lingual thyroid is a rare embryological anomaly caused by failure of the gland to descend from the foramen cecum to its normal site in the pre-laryngeal area. Its precise pathogenesis is unknown, and has been found to be more prevalent in females (female:male ratio, 7:1).<sup>1</sup> Although bleeding from an ectopic lingual thyroid is rare, life-threatening massive hemorrhage is possible, since the lingual thyroid may have prominent large blood vessels on its surface.<sup>2-4</sup> We present a rare case of bleeding from an ectopic lingual thyroid presenting as hematemesis.

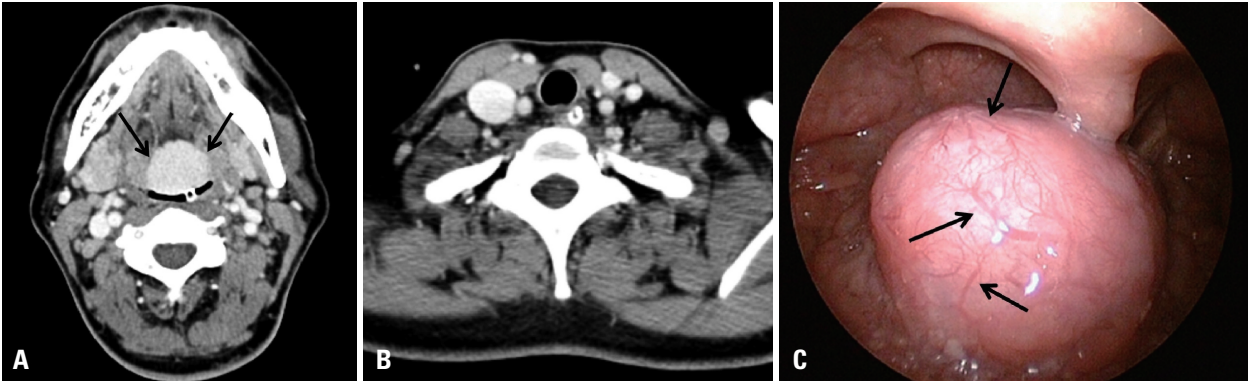
A 42-year-old woman visited the emergency room of our hospital complaining of hematemesis. Her blood pressure was 110/70 mm Hg and pulse rate was 80 beats per minute. Given a history of recent drinking and vomiting, the initial impression was Mallory-Weiss syndrome. Initial emergency gastroendoscopic examination revealed fresh blood in the stomach, without any evidence of mucosal abnormalities in the esophagus and stomach. Under supportive care, the hematemesis spontaneously stopped two days later, and a follow-up gastroendoscopic examination showed no blood in the stomach. Neck computed tomography (CT) examination revealed a 3-cm enhancing mass on the midline base of the tongue and no presence of thyroid tissue in the normal thyroid position. On laryngoscopic examination, the mass had a smooth surface with engorged vessels, and the lesion was more clearly visualized when the tongue was pulled forward, which are characteristic findings of an ectopic lingual thyroid (Fig. 1). Thyroid ultrasonography revealed absence of the thyroid gland. Technetium (Tc99m) thyroid scan was performed and demonstrated isotope uptake at the base of the tongue and no uptake in the normal thyroid location (not shown here). A thyroid function test showed subclinical hypothyroidism: free thyroxine (fT4), 1.52 ng/dL (normal range, 0.89–1.78 ng/dL); T3, 95 ng/dL (normal range, 60–180 ng/dL); and thyroid-stimulating hormone, 6.43 mIU/mL (normal range, 0.17–4.78 mIU/mL). Although surgical removal was recommended due to the risk of re-bleeding, the patient refused surgery and opted for a wait and see approach.

Ectopic thyroid is a rare developmental anomaly and is caused by aberrant embryogenesis during the descent of the thyroid gland to the neck. Thyroid tissue may be found anywhere along the course of the thyroglossal duct (i.e., oropharynx, infrahyoid area, trachea, mediastinum, esophagus, and cervical lymph nodes).<sup>2,5</sup> Among these, lingual thyroid, complete arrest of thyroid tissue at the base of tongue, is the most common form, with a reported prevalence of 1 in 100000 to 1 in 300000 (90% of all cases of ectopic thyroid).<sup>6</sup> Many patients are asymptomatic and a diagnosis

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**Fig. 1.** (A) CT image showing a 3.0×2.5 cm enhancing mass (arrows) at the midline base of the tongue, above the epiglottis. (B) CT image showing no thyroid tissue in the neck or pretracheal area of the second to fourth tracheal rings (normal thyroid position). (C) Laryngoscopic examination reveals a well-defined protruding mass with a smooth surface with prominent vessels (arrows) at the midline base of the tongue.

is made incidentally as a result of neck imaging for localization of an ectopic thyroid tissue and to confirm the absence of thyroid tissue in the normal position (the pretracheal region of the second to fourth tracheal rings). In symptomatic patients, the lingual mass may result in dysphagia, foreign body sensation in the neck, dyspnea, and oropharyngeal obstruction. Although bleeding from an ectopic lingual thyroid is rare, it can be life-threatening, as massive hemorrhage is possible, since the surface of a lingual thyroid may be covered by engorged and irregular blood vessels.<sup>2-4</sup> Mean age at clinical presentation is 40 years, with two peaks at 12 and 50 years of age.<sup>2,3</sup> In our case, neck CT helped demonstrate that the lingual thyroid was the only functioning thyroid tissue, and a thyroid function test indicated mild hypothyroidism. Accordingly, we recommended thyroxine suppression therapy to prevent subsequent enlargement of the lingual thyroid and to reduce the risk of malignancy.<sup>3</sup> Treatment of a lingual thyroid depends on the size, the presence of symptoms, and the presence of complications, such as ulceration, hemorrhage, and malignancy. Treatment of an ectopic thyroid is not necessary when the mass is asymptomatic and thyroid function and cytology are normal. However, surgery is considered when a mass produces obstructive symptoms or bleeding, demonstrates a sudden increase in size, or

if malignancy is suspected.<sup>2,3,6</sup> In patients being considered for surgical excision, it is essential to establish if any normal thyroid tissue is present elsewhere, because removal of a lingual thyroid will in most cases render the patient profoundly hypothyroidal.

After ruling out common causes of acute gastrointestinal bleeding by gastroendoscopic examination, bleeding ectopic lingual thyroid could be considered as a rare cause of hematemesis in emergency situations.

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