A rare etiology of oropharyngeal obstruction

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Clinical history

A 22-year-old woman, without previous history, presented with impaired swallowing, altered timbre and nocturnal snoring, of gradual onset over the previous year. Direct laryngoscopy found a reddish, firm sessile mass at base of the tongue. Head-and-neck MRI was performed, and your interpretation of the images is requested (Fig. 1).

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Questions

Question no. 1
What is your radiological interpretation?

Question no. 2
Which complementary examinations do you prescribe?

Question no. 3
What is your diagnosis?

Question no. 4
What is your treatment attitude?

What is your diagnosis?
Replies

Reply no. 1: What is your radiological interpretation?

Sagittal head-and-neck MRI shows an oblong oropharyngeal mass at the base of the tongue. The lesion is hypointense on T1-weighted sequences (A: sagittal slice) and hyperintense on T2 (B: axial slice). The mass is intensely and homogeneously enhanced on gadolinium injection (C: sagittal slice; D: coronal slice).

Reply no. 2: Which complementary examinations do you prescribe?

Cervical ultrasound finds an empty thyroid space. There are no clinical signs of dysthyroidism. US-thyroid stimulating hormone (TSH) level is normal, at 1.80 mIU/l, as are T3 and T4 (respectively, 4.4 and 11.2 pmol/l).

Reply no. 3: What is your diagnosis?

Basilingual ectopic-thyroid goiter.

Reply no. 4: What is your treatment attitude?

Surgery is indicated by clinical signs and tumor volume. Following tracheotomy, the lesion was completely removed by medial pharyngotomy on a suprathyroid approach, conserving the hyoid bone. Histopathology confirmed the benign thyrohyoid nature of the tongue-base mass. The patient was decannulated on day 5, and the nasogastric probe was removed on day 15 after clinical check-up confirmed resolution of swallowing disorder and dyspnea. Hormone replacement therapy (100 µg Levothyroxine®) was initiated. At 3 months, healing was good and there was no more swallowing disorder.

Comments

Basilingual ectopic thyroid is a rare clinical entity of embryological origin, first described by Hickman in 1869, since then less than 400 cases have been reported [1,2]. It is due to defective intrauterine thyroid gland migration in the thyroglossal duct. Ninety percent of thyroid ectopias are basilingual. Prevalence is reported to range from 1/100,000 to 1/300,000 [3], but is probably underestimated: autopsy studies found thyroid cells at the tongue base in 10% of corpses [1]. Incidence is elevated four-fold in females in their fourth decade [1–4]. Etiopathogeny is unclear, but antithyroid antibodies secreted in the womb may prevent thyroid precursor migration to the thyroid space from the second branchial pouches [4]. Female predominance is due to hormonal changes during puberty, menstruation and pregnancy. Thyroid hormone synthesis by the ectopic tissue may be sufficient in physiological conditions, but not when the organism’s metabolic needs increase, inducing TSH hypersecretion, leading to hypertrophy of the ectopic tissue, which may compensate the hormonal deficit, resulting in the euthyroidism found in two thirds of the cases reported in the literature [1–5]. Pathology is in that case revealed by the ectopic glandular mass, causing oropharyngeal obstruction syndrome [1,3]. Recurrent or cataclysmic bleeding was reported, although malignant degeneration of the ectopic goiter is exceptional [1].

An ectopic thyroid may be located anywhere along the thyroglossal duct, but most frequently at the base of the tongue. Exceptionally, mediastinum, esophagus, heart and diaphragm sites have been reported [1].

Clinically, basilingual ectopic-thyroid goiter may be asymptomatic [4]. Revelation is often by oropharyngeal obstruction syndrome, associating dysphagia, dyspnea, oropharyngeal foreign-body sensation and, more rarely, hemoptysis. Hypothyroidism is observed in one third of cases [2]. Ultrasound and especially contrast-enhanced cervical CT usually enable diagnosis. Head-and-neck MRI may be used in first intention when ectopic goiter is suspected due to a basilingual mass. T1- and T2-weighted (w) sequences show high sensitivity and specificity. The ectopic thyroid may appear on T1-w as a well-contoured mass in iso-hypersignal as compared to muscle, with moderate and sometimes heterogeneous enhancement by gadolinium, and in slight hypersignal on T2-w, without signs of invasion of neighboring tissue. MRI also determines the size and exact location and relation to tongue muscles, and thus the appropriate surgical approach [5]. Lingual thyroid ectopia has been shown to be associated with in situ cervical thyroid in 30% of cases [1]. Iodine-131 or technetium-99 scintigraphy, however, can locate the goiter, demonstrate the empty thyroid space and explore for other ectopic sites [2]. A basilingual mass showing the above MRI features associated with an empty thyroid space is highly suggestive of ectopic-thyroid goiter. There are, however, differential diagnoses: palatine tonsil lymphoid tissue hyper trophy, accessory salivary gland tumor, or tongue-base tumor [2].

Treatment is not systematically surgical. Abstention with surveillance is indicated for small asymptomatic or weakly symptomatic lesions [2]. In goiter showing clinical and biological signs of dysthyroidism without obstruction or threat of hemorrhage, medical management alone may obtain regression of goiter volume and of obstructive symptoms [1,2]. Surgery is indicated for obstructive forms and forms without dysthyroidism. Complete exeresis may be performed on an intrabuccal, cervical suprahyoid or combined approach. Technique depends on tumor volume and cervical extension. Oral or nasal intubation entails risk, and requires the presence of a surgeon; nasal intubation facilitates surgery using an oral approach. Preoperative tracheotomy avoids the risk of respiratory distress caused by ptosis, hematoma or lingual hemorrhage [5]. Postoperative nasogastric feeding reduces the risk of pharyngostoma.

If surgery is not indicated, regular follow-up with clinical, endoscopic, biological (TSH) and radiological (MRI) assessment is required to detect signs of dysfunction or increased volume of the thyroid. Malign transformation, known to be
frequent in ectopic thyroid, has been exceptionally reported in basilingual locations [1,2].

**Conclusion**

Basilingual goiter is a rare pathology, but nevertheless to be suspected in case of a basilingual mass of benign radiological aspect. Surgery is indicated only in obstructive forms without signs of dysthyroidism. Clinical evolution following well-conducted treatment is good.

**Disclosure of interest**

The authors declare that they have no conflicts of interest concerning this article.

**References**