Colloid goiter in ectopic thyroid tissue presenting as submandibular swelling with a coexisting functional orthotopic thyroid gland in a pregnant female

Sir,

The thyroid tissue is defined as ectopic when not located in the pretracheal region between the second and the fourth tracheal cartilages. The ectopic thyroid gland is very rare, with a prevalence of approximately one in 100,000 to 300,000 in the population, with a female to male ratio of about 4:1, the age ranging from 5 months to 40 years but is usually seen in adolescence, pregnancy, and menopause due to increased physiological demand of thyroid hormones and marked endocrine activity. Most of the patients of ectopic thyroid are asymptomatic.[1,2] Lingual endotracheal tube (ETT) is the most common site, accounting for more than 90% of cases, and patients usually have hypothyroidism. The simultaneous finding of the submandibular ectopic thyroid tissue and a functional orthotopic thyroid gland is an extremely rare event.[3] As in many cases, the ectopic thyroid gland is the only functioning thyroid. The incidence of malignancy arising in ETT is approximately 1%. [1]

A 19-year-old pregnant female presented with a firm, nontender swelling measuring 2.0 cm × 1.5 cm in the right submandibular region [Figure 1a]. The thyroid gland was not enlarged. Ultrasonography revealed a hypoechoic lesion measuring 2.4 cm × 1.4 cm in the right submandibular region with cystic changes, separate from submandibular gland anterior to bifurcation of the common carotid artery [Figure 1b]. No significant lymphadenopathy was seen. Sonography features were in favor of the ectopic thyroid tissue. Thyroid function tests were normal; thyroid-stimulating hormone (TSH) level was 2.0 mU/L (normal range 0.3-4.5), FT4 1.5 ng/dL (0.8-2), FT3 4.5 pg/mL (1.8-5). Calcitonin, calcium, and parathyroid hormone (PTH) levels were normal. Because of pregnancy, Tc99m pertechnetate scan and computed tomography (CT) scan were not performed.

Fine needle aspiration yielded blood mixed colloid, smears prepared showed abundant thin colloid over a background of blood. A few follicular epithelial cells arranged in micro and macro follicles, revealed involutional and hyperplastic changes, with no features of malignancy. Abundant foamy and pigment-laden macrophages and multinucleated giant cells were additionally seen along with a few lymphocytes [Figure 1c]. Cytological findings suggested ectopic thyroid tissue with colloid goiter with cystic change.

Cases of ectopic thyroid, though rare, should be considered as a differential diagnosis of the submandibular mass. An ultrasound and fine needle aspiration cytology (FNAC) will help in confirming the thyroid tissue and diagnosing any existing pathology. Hence, the present study illustrates the potential for rare sites of ectopic thyroid gland lateral to the midline that enters into the differential diagnosis of metastasis from well-differentiated thyroid cancers.

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Conflicts of interest
There are no conflicts of interest.

Figure 1: (a) Firm, nontender swelling measuring 2.0 cm × 1.5 cm in right submandibular region (b) Ultrasonography revealed a hypoechoic lesion measuring 2.4 cm × 1.4 cm with cystic changes (c) A few follicular epithelial cells revealing involutional changes along with abundant foamy and pigment-laden macrophages in a background of thin colloid and RBCs. (Giemsa stain, x200)
Sir,

Pheohyphomycosis is caused by brownish-black colored dematiaceous fungi characterized by melanin in cell wall. Clinically, it presents as localized infections of skin, subcutis, or systemic involving the central nervous system, eyes, or bones.\[1,2\] It is considered to be an opportunistic fungus commonly reported in immunocompromised patients which may spread from the primary infected site to other organs resulting in increased mortality. Therefore, its early diagnosis is essential to prevent further complications.

A 55-year-old female, a housewife and a resident of Uttarakhand, a north Himalayan state of India, presented with a gradually increasing 2 × 2 cm, soft, tender swelling over the dorsum of the right hand for last 2 months \[Figure 1a\]. There was no history of trauma, diabetes, and tuberculosis; her human immunodeficiency virus (HIV) status was also negative. The patient was subjected to fine needle aspiration cytology (FNAC); the smears revealed fungal hyphae which were septate showing branching with irregular constrictions along with conidia in background of inflammation and necrosis. The wall of fungal hyphae, at places, showed brownish-black pigment on Giemsa stain, and hence a diagnosis of pheohyphomycosis was suspected \[Figure 1b\]. Smears were then subjected to periodic acid-Schiff (PAS) and silver methanamine (SM) stains which showed positivity for fungal hyphae \[Figure 1c and d\]. Fungal hyphae also showed brownish-black pigment in their cell wall on Masson Fontana staining and a diagnosis of pheohyphomycosis was confirmed. This was followed by surgical excision; histopathology was consistent with subcutaneous pheohyphomycosis. The patient was treated with itraconazole for a period of 3 months, and she responded well to the treatment.

Pheohyphomycosis are usually found in soil infected subcutaneous tissue mostly on the extremities and may spread to distant sites particularly in immunocompromised individuals. The present case also demonstrated pheohyphomycosis in subcutaneous tissue of hands, which may have been implanted from soil as the patient was actively involved in gardening. Pheohyphomycosis has been reported very rarely from this area and literature search shows that it is the second reported case from this north Himalayan Uttarakhand region. The previous reported case was of pheohyphomycoic keratitis which also presented in an immunocompetent patient without any history of trauma.\[3\]

Deposition of melanin in the cellular wall of fungus is a characteristic of dematiaceous fungi. The present case on vigilant cytomorphological examination also clearly showed the presence of brownish-black pigment in fungal walls on Giemsa stain, which raised suspicion.