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Letter to editor

A diagnosis you wouldn't expect in a supraclavicular mass

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Dear Editor,
A 50-year-old woman, undergoing chest X-rays in preparation for urinary incontinence surgery, was found to have multiple lung lesions, suspicious for metastasis of a tumor of unknow origin. Her medical history was positive for type II diabetes, grade I obesity, hypertension, fatty liver disease, dyslipidemia, non-toxic multinodular goiter, and glaucoma. Computed tomography (CT) scan confirmed the presence of multiple bilateral solid lung nodules, with round and well-defined margins. Most cranial scan sections showed a single, solid neck mass measuring 5 x 2.6 cm, located in the left supraclavear fossa. The patient was referred to our ENT unit for further evaluation. At objective examination, the lesion presented as a single, parenchymatous left supraclavear mass, and was apparently mobile with respect to superficial tissue without clinical evidence of tissue infiltration. Examination of the oral cavity and fibroscopy of the upper airways did not identify any possible primary tumor. Total body CT-positron emission tomography (PET) scan showed minimal uptake in the neck mass (SUV 3.1) and no significant uptake of 18 fluorodeoxyglucose in lung nodules. Ultrasound-guided fine needle biopsy of the lesion was performed at another institution, providing the diagnosis of a spindled cell proliferation, suggestive for leiomuscular origin; the available tissue did not allow further diagnostic investigations.
The patient underwent surgical resection of the neck mass. The histological examination confirmed an expansile proliferation of well differentiated leiomuscular cells, positive for desmin, actin, and H-caldesmon. Cytological atypia, mitotic activity or necrosis were not observed (Fig. 1). The lesion was not associated with lymphoid tissue. Findings were coherent with a localization of leiomyoma. The extreme rarity of primary neck leiomyomas and the presence of concomitant lung nodules suggested an origin from the female genital tract. The patient's history was positive for hysterectomy at the age of 44 years for uterine fibroids. After the multidisciplinary discussion the patient was taken in charge from our oncological department. Gynecological evaluation supported the hypothesis of possible intravascular spreading leiomyomatosis and gave indication to the revision of the hysterectomy histological samples, ant repeat thoracic CT scan after 3 months to monitor the pulmonary lesions. Therapy with gonadotropin releasing hormone analogues was indicated in case of diagnostic confirmation. The patient did not comply to the prescription and was lost to follow-up.

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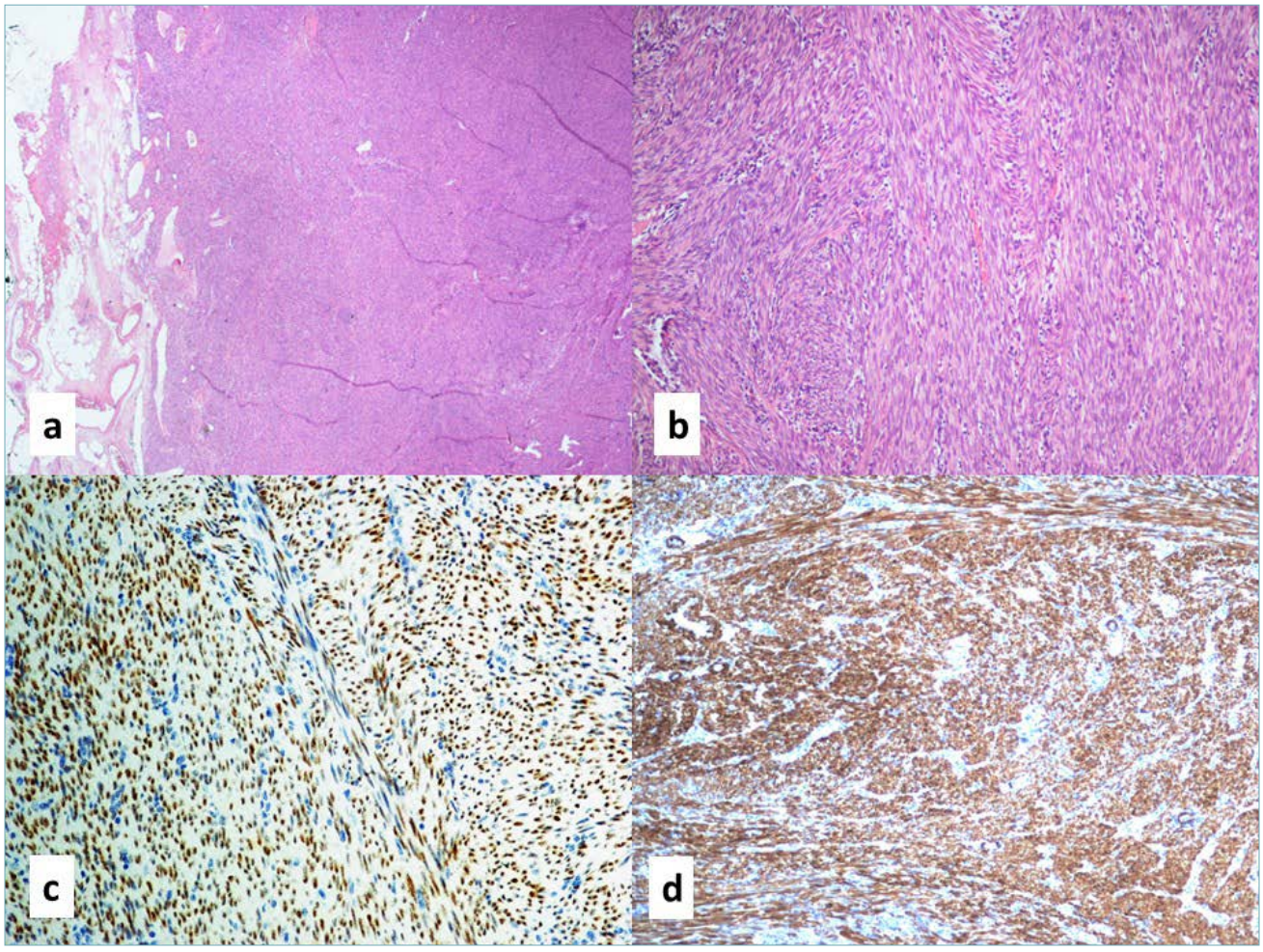

Figure 1. Microscopic images of the surgically resected neck mass. (a) Low-power view, showing a cellular lesion with sharp borders, and no peripheral lymphatic tissue; (b) high power view documenting spindled cell proliferation, fascicular architecture, and monotonous, mild cellularity (H&E, 10x); (c) immunostain documenting homogeneous nuclear expression of estrogen receptor (DAB chromogen, 10x); (d) immunostain documenting diffuse cytoplasmic expression of H-caldesmon (DAB chromogen, 10x).

Neck masses are common findings in adults with a wide range of possible differential diagnoses. When approaching a lateral neck mass it is important to keep in mind that it may develop from inflammatory, infectious, benign or malignant neoplastic, congenital or traumatic processes [1]. It is well recognized that persistent neck masses in adults may be the initial or the only clinical manifestation of head and neck malignancies, and should always be considered as malignant until proven otherwise. Histological evaluation is necessary to precisely characterize the nature of a potentially malignant lesion, but it may not help in defining the site of origin of a metastatic localization. Accurate collection of patient's history and thorough ENT evaluation and imaging are crucial in defining the possible site of origin of the a neck metastasis. Rare causes of lateral neck mass should be considered in case of atypical presentation.

As far as we know, no other case of cervical benign metastasizing leiomyoma (BML) has been reported in literature. BML originate form hematogenous spread outside of the genital area of leiomuscular cells during surgical resection of uterine fibroids (myometrial leiomyomas, the most common type of female genital tumor) [2], is considered rare. Other possible sources of BML are intravenous leiomyomatosis and metastatic low-grade leiomyosarcoma [3]. BML are most commonly found in the lungs, but heart, lymph node, mesentery, liver, bladder, central nervous system, skeletal muscle and bone localizations have been reported as well [4].

BML are often incidental findings on chest imaging in asymptomatic patients Symptoms, if present, are always due to size and burden, and change according to disease location, including cough, chest pain, hemoptysis, hemothorax, pneumothorax and dyspnea [5]. Disease course is indolent, with slow growth rate and possible spontaneous regression over time in response to hormonal changes associated with pregnancy and menopause. A palpable mass or nodule could be the first manifestation of BML in soft tissue or skin [2]. FDG PET-CT can distinguish BML from malignant tumors, which show high 18F-FDG uptake [3]. The diagnosis of BML is established with biopsy and histopathology, showing nodules composed of circumscribed fascicles and whorls of well-differentiated spindle cells positive for smooth muscle actin (SMA), desmin, and calponin [2]. Tumor cells are hormone-sensitive as they express nuclear estrogen and progesterone receptors. The histopathological diagnosis should always rule out a low grade malignancy (uterine leiomyomsarcoma) [3]. Besides mild cell morphology and the absence of obvious nuclear atypia, lack of mitoses and necrosis and low proliferative state (Ki-67 index) are useful to differentiate BML from its malignant counterpart.

No standard treatment for patients with BML is available due to its rarity. Surgery is indicated in case of a single and localized lesion that can be completely eradicated with resection [2]. Due to their slow growth, asymptomatic BML can be monitored without treatment. The suppression of sex hormone production through long-acting GnRHa avoids progression or recurrence.

The present case is a rare example of atypical laterocervical neck mass that ENT surgeons may come across to. The diagnosis of less common causes of benign neck masses can be challenging, and a histolopathological diagnosis together with accurate evaluation of clinical data is crucial in order to reach the right diagnosis.

Conflicts of interest

The authors declare no conflict of interest.

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