Dear Editor,

Angioleiomyoma is an uncommon benign solitary neoplasm composed of smooth muscle cells that arise from the tunica media of veins. The most prevalent site is the skin of the extremities, in particular the lower legs, and the lip has been reported to be a very rarely involved site. We herein describe a case involving the largest labial angioleiomyoma yet reported, which presented as an asymmetric persistent swelling of the lower lip.

A male patient aged 71 years was referred to our clinic for the evaluation of an asymmetric persistent swelling of the lower lip for the previous 2 years (Figure 1A). He had no subjective symptoms, including pain or tenderness, but reported that the size of the lesion had increased. He did not have a history of trauma, injection, or contact with irritants. Physical examination revealed an ill-circumscribed enlargement of the left side of the lower lip without prominent changes in color. Neither facial paralysis nor abnormalities of the tongue and gums was observed. A computed tomography (CT) scan from the previous hospital that treated this patient showed a solid, highly enhancing ovoid and well-circumscribed nodule that was 2.2 cm in maximum diameter (Figure 1B). The nodule was situated mainly within the subcutaneous fat layer and abutted the dermis and facial expression muscles at the uppermost and lowermost surfaces, respectively, and elevated the skin. There was no pathologic lymph node in either of the cervical areas.

An excisional biopsy revealed a 2.3 × 1.5-cm well-defined firm and round whitish nodule, which was easily dissected from the surrounding tissue. The histopathology of the specimen disclosed a well-circumscribed encapsulated nodule predominantly composed of thick bundles of smooth muscle fibers and numerous vascular spaces (Figure 2A and B). There were also areas of hyaline changes and mature fat cells detected, but no obvious mitosis was observed. Immunohistochemical staining showed that the tumor cells were positive for smooth muscle actin (Figure 2C and D). The lesion showed no recurrence during a 4-month follow up.

Labial angioleiomyomas have thus far been reported rarely. Furthermore, there has been no previously reported case of labial angioleiomyoma greater than 2 cm in diameter to our knowledge. Among various etiological factors proposed, minor trauma may be relevant for angioleiomyoma of the lip, which is subject to repeated minor injuries. The reported cases of labial angioleiomyomas typically presented as a small asymptomatic round or raised sessile submucosal nodule, which therefore appeared quite similar to a mucocele, a hemangioma, a salivary gland tumor, a fibrous hyperplasia, or a pyogenic granuloma. However, the lesion in our case looked more like a swelling of the lip rather than a well-defined round nodule, which may be attributed to its large size. The lip swelling may result from numerous conditions, including trauma, angioedema, chelitis, acromegaly, mucopolysaccharidosis, and bacterial or herpes infection. Uniquely, asymmetric and persistent lip...
swelling, which was observed in our case, raises the suspicion of cheilitis granulomatosa and associated Melkersson–Rosenthal syndrome, Crohn’s disease, or sarcoidosis. Without CT scan examination, we may have suspected cheilitis granulomatosa as a more preferential diagnosis. Other noninvasive imaging tools including ultrasound examination and magnetic resonance imaging may also help make a differential diagnosis. A labial angioleiomyoma is rarely considered an initial clinical diagnosis because it is much rarer than common similar-looking conditions such as mucocele. Furthermore, atypical presentation similar to what was observed in our case makes it even more difficult to include angioleiomyoma in the differential diagnosis. In summary, we here report a case of the largest labial angioleiomyoma recorded to date and recommend considering it as a differential diagnosis of asymmetric persistent swelling of the lip.

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Figure 2 Histopathologic examination from the excisional biopsy showing a well-circumscribed encapsulated nodule predominantly composed of thick bundles of smooth muscle fibers and numerous vascular spaces with areas of hyaline change and mature fat cells: (A) hematoxylin and eosin staining, ×40; (B) hematoxylin and eosin staining, ×400. Positive immunohistochemical staining of the tumor cells for smooth muscle actin: (C) ×40; (D) ×400.

References


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