A Case of Angioleiomyoma on the Lip

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It is known in general that neoplasms of smooth muscle appear very rarely in the oral region, probably because of the absence of smooth muscle except in the blood vessel walls. In the literature, a few cases in the oral region with histopathologic investigations have been reported. This report presents an unusual case of angioleiomyoma that developed on the lower lip of a 48-year old woman over the course of one year and the immunohistochemical findings of the angioleiomyoma are described. (Ann Dermatol 8:(1)57~60, 1996)

Key Words : Angioleiomyoma, Lip

The angioleiomyoma is a rare, usually benign tumor arising from the muscle of veins and is characterized by a painful solitary tumor occurring most frequently in the extremities[1-4].

Since the tumor appears infrequently in the other anatomical sites[9,10], it is difficult to make an initial diagnosis of angioleiomyoma when a lesion develops at a location other than the predilection site.

We report a case of angioleiomyoma occurring on the lip of a 48-year-old woman in which the histologic examination of a non-tender solitary nodule is consistent with angioleiomyoma.

REPORT OF A CASE

An asymptomatic semitranslucent pinkish nodule developed on the lower lip of a 48-year-old woman over the course of one year (Fig. 1). It was a dome-shaped, firm nodule and 7 mm in diameter. Based on the physical examination, initially a clinical diagnosis of mucous cyst was made and the lesion was surgically removed.

Histopathologically, there was a tumor mass which consisted predominantly of smooth muscle fibers and vascular channels (Fig. 2). The muscle fibers encircled with variable sized blood vessels were arranged in concentric layers. Our biopsy specimens were subjected to Masson's trichrome (Fig. 3).

The results of immunohistochemical staining were as follows (Table 1): tumor cells were positive for desmin and vimentin but not for neuron specific enolase, S-100 protein and actin (Fig. 4,5). Factor VIII related Ag was detected in the endothelium of blood vessels (Fig. 6).

At present the tumor lesion was not recurred 3 months after surgical excision.

Table 1. Results of immunohistochemical stain

Antigen	Results
vimentin	+
desmin	+
NSE	−
S-100 protein	−
factor VIII-related Ag	+
actin	−

DISCUSSION

Leiomyomas can be classified into four types in origin; multiple piloleiomyomas of arrector pilar origin,

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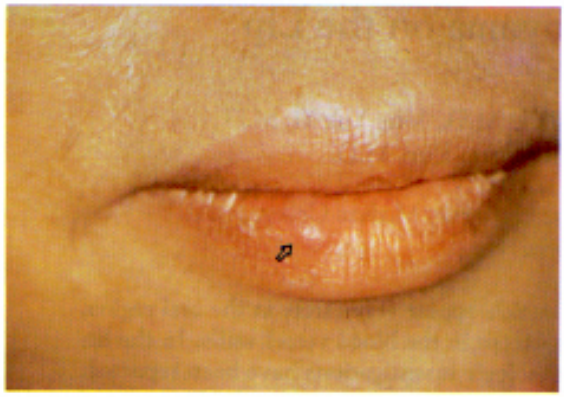

Fig. 1. Semitranslucent pinkish, dome shaped nodule on the lower lip (arrow).

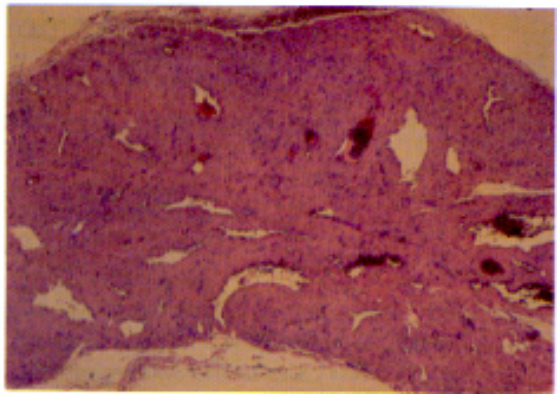

Fig. 2. The tumor mass is encapsulated and showes bundles of smooth muscle fibers and the vascular channels (H&E, ×20).

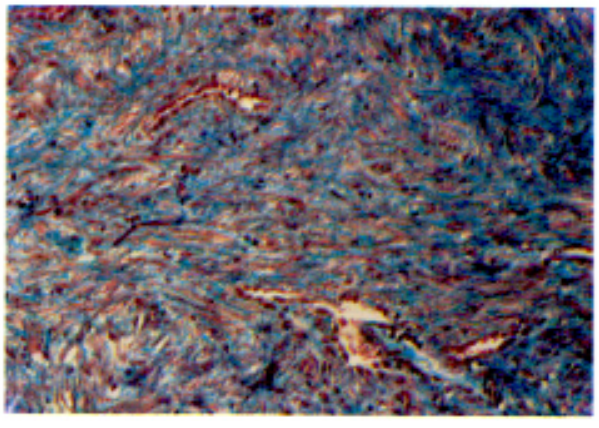

Fig. 3. Smooth muscle bundles are irregularly separated by strands of collagen fiber(Masson's trichrome, × 100).

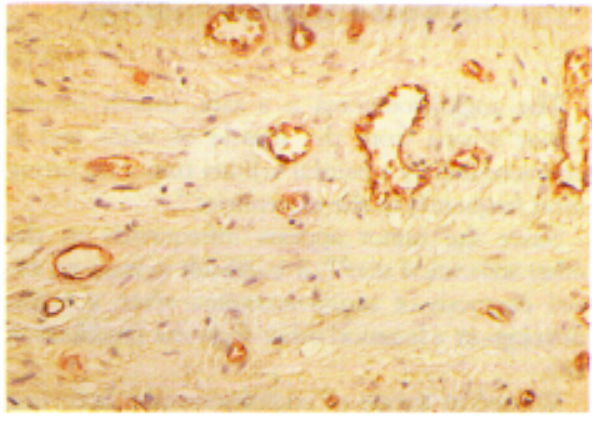

Fig. 4. Smooth muscle cells positive for desmin are seen in the tumor mass(Immunohistochemical stain, ×200).

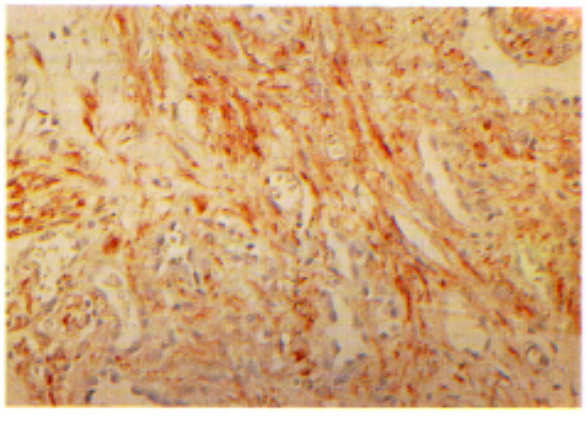

Fig. 5. Smooth muscle cells and the basement membrane of endothelial cells positive for vimentin are seen in the tumor mass (Immunohistochemical stain, ×200).

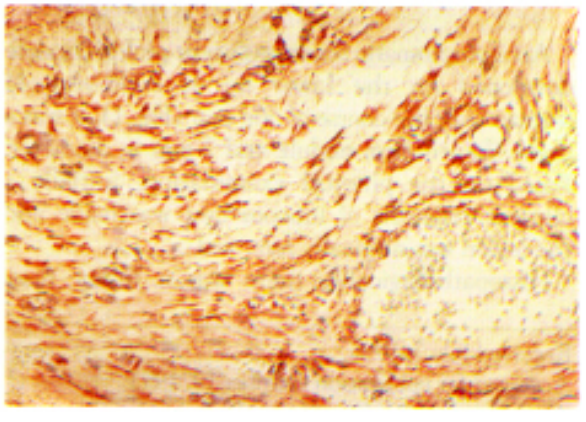

Fig. 6. Endothelial cells positive for Factor VIII-related Ag. are seen in the tumor mass (Immunohistochemical stain, ×200).

solitary piloleiomyoma arising from the smooth arrector pili muscle, solitary genital leiomyoma arising from the dartoic, vulvar, or mamillary muscle, and solitary angioleiomyoma arising from the muscle of veins[1].

Of them angioleiomyoma is usually a solitary tender lesion and it is most commonly reported on the lower extremities of middle-aged women[1-4]. Its differential diagnosis includes tender tumors such as neuroma, neurilemmoma, and eccrine spiraderma[5], because pain is the most characteristic subjective symptom in a patient with angioleiomyoma. Therefore, it is difficult to make a clinical diagnosis in a case when a non-tender lesion occurs at a location other than the predilection sites. Hachisuga[9] described that the angioleiomyoma occurring in the head regions were not usually accompanied by pain and our case involving an unusual site, the lower lip was without pain.

In the Korean literature, there have been 3 reported cases of leiomyoma[6-8] on the lower extremities, cheek and multiple locations involving both shoulders, right arm, right side of the neck, upper chest and upper back, respectively, and no case of angioleiomyoma on the lip has been reported yet.

With the appearance of a mucous cyst and the absence of pain, we could only confirm the diagnosis of angioleiomyoma by histopathological findings.

The pathogenesis of angioleiomyoma remains obscure. The cause of the frequent pain or tenderness has not been satisfactorily determined. Morimoto[11] suggested that pain would follow contraction of vessels giving rise to local ischemia. Also Mann et al[4] have observed damage to nerve fivers through distortion and disruption of the myelin sheath, which might induce the pain in the angioleiomyoma.

Morimoto[11] separated these tumors into two groups; the larger group of extremity tumors and the smaller group of head tumors. In the former group, tumors were mainly of the solid type and often painful, whereas the latter tumors were usually the venous type and painless.

Histopathologically, the leiomyoma is composed of bundles and masses of smooth muscle fibers[3]. The muscle bundles are irregularly separated by strands of collagen fibers. The finding of our biopsy is consistent with that of angioleiomyoma (Fig. 2) with the aid of Masson's trichrome staining (Fig. 3).

Histopathological differentiation must be made from neurofibroma and other spindle cell tumors as well as leiomyosarcoma. Angioleiomyoma differs from dermatofibroma when stained with Masson's trichrome[4]. It can be also distinguished from neurofibroma by being encapsulated and contains numerous veins[4]. Leiomyosarcoma can be also discriminated with the appearance of atypical cells[1-4].

In addition, we performed a variety of immunohistochemical stainings such as desmin, vimentin, S-100 protein, neuron specific enolase, actin and factor VIII-related protein in order to distinguish angioleiomyoma from any other tumor. The presence of factor VIII-related antigen (Fig. 6) and desmin (Fig. 4) in the immunohistochemical study of our case is an indication of muscle origin and endothelial cell[4,10].

Excision is the treatment of choice for solitary angioleiomyoma if a patient complains of pain or cosmetic concerns[1-4]. In our patient, surgical excision of the lesion was performed and there was no evidence of recurrence after 3 months.

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