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Case Report
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Cavernous Cutaneous Angioleiomyoma: A Case Report and Literature Review

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Abstract

Cutaneous angioleiomyoma is rare in the clinic, and cavernous cutaneous angioleiomyoma is rarer in its classification. However, there are few relevant literature reports on it. People have insufficient clinical cognition of the disease, making it easy to be misdiagnosed and then causing delayed treatment. Because the disease symptoms are not unique, the non-invasive examination (ultrasound, MRI, and alike) cannot make exact conclusions, and the preoperative diagnosis is challenging. Therefore, the study and summary of clinical cases are necessary to improve the cognition and diagnosis of the disease. In addition, we report a rare case of cutaneous angioleiomyoma located above the medial malleolus of the patient's right foot. After surgery, according to the histopathological findings, the tumor was diagnosed as cavernous cutaneous angioleiomyoma.

Keywords: Angioleiomyoma; Cavernous; Cutaneous; Clinical; Diagnosis

Introduction

Cutaneous angioleiomyoma is a kind of cutaneous leiomyoma classified according to the tissue source of cutaneous leiomyoma. It is a benign soft tissue tumor. Skin leiomyoma accounts for about 5% of all leiomyomas [1] rare in the clinic. The pathological feature of angioleiomyoma is the proliferation of smooth muscle cells and prominent vascular components. It can occur all over the body, especially in the lower limbs. After searching the literature database, we found few reports on cutaneous angioleiomyoma. Here, we present the case of a patient with large cavernous cutaneous angioleiomyoma above the medial malleolus of the right foot, which is adjacent to the joint, has a long growth life but is not accompanied by pain. It may be confused with lipoma, schwannoma, neurofibroma, pseudoaneurysm, epidermal cyst, giant cell tumor of the tendon sheath, and other diseases. This report is hereby made.

Case Presentation

The patient was a 42-year-old male who had seen a lump above the medial malleolus of his right foot for more than ten years, and he went to the dermatology department of Shuguang Hospital Affiliated with the Shanghai University of traditional Chinese medicine on September 02nd, 2021. The patient had no obvious inducement ten years ago and found a soybean size tumor above the right medial malleolus, without pain, ulceration, and other discomforts, so he ignored it. In recent years, the tumor has gradually increased and is asymptomatic. No previous medical diseases, no history of surgical trauma and no particular family history were reported. Physical examination: The general condition is good, and the superficial lymph nodes of the whole body are not palpable and swollen. Dermatological examination: the upper part of the medial malleolus of the right foot can be felt with a 5 cm-diameter lump. The boundary is clear, soft, with no pain, no change in skin temperature, acceptable mobility, and slightly darker surface skin color (Figure 1). Laboratory examination: Blood routine, coagulation, and infectious diseases (hepatitis B, hepatitis C, syphilis, HIV) were regular. Ultrasound showed the mass of the right leg: a hypoechoic area was found under the skin, with a size of about 31.5 mm × 11.6 mm × 24.6 mm, clear boundary, regular shape, uniform internal echo, CDFI: abundant blood flow signals (Figure 2).

Treatment: the lesion was completely removed by operation, and a pathological examination was performed.

Histopathological examination: The tumor has a clear boundary and contains abundant

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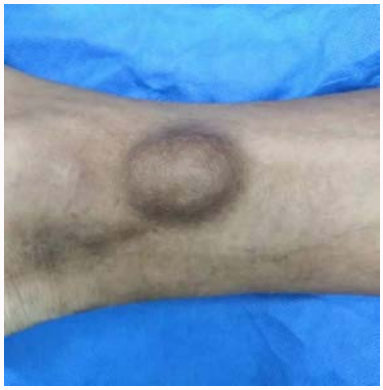

Figure 1: A hemispherical mass with a diameter of about 4cm can be seen above the medial malleolus of the patient's right foot. The boundary is clear, soft and the skin color on the surface is slightly dark.

bloodstream and smooth muscle cells. The blood vessel wall is thin and filled with red blood cells. The smooth muscle cells arranged in bundles surround the blood vessel wall.

Tip: conforming to cavernous angiomyoma (Figure 3a, 3b).

Diagnosis: Cavernous cutaneous angioleiomyoma.

Discussion

In skin tissue, smooth muscle cells are distributed in three parts: Arrector pili muscles, blood vessel walls, genital skin of the scrotum, vulva, and nipple. Cutaneous angioleiomyoma is a benign tumor in vascular smooth muscle. Skin angioleiomyoma can be divided into three types from the perspective of histology [2]: Solid, venous, and cavernous. Solid is the most common, and cavernous is the least. There are many dilated vascular cavities and less smooth muscle components in cavernous tumors. The reported case is a sponge-like type from a pathological point of view. Because vascular smooth muscle is in the middle layer of blood vessels, vascular leiomyoma is a pathological change in the vascular layer. It occurs everywhere in the body, as long as it contains the vascular distribution in the middle membrane. Literature studies have found that leiomyoma is more common in limbs, especially the calf, commonly seen in middle-aged patients [3]. Most articles have mentioned that this disease is more common in women, and the ratio of males to females is about 1:1.5~1.7 [4]. As for the factors that cause skin leiomyoma, we suppose tracing back to the history of trauma is helpful. Therefore, it is necessary to ask whether there was any trauma at the site of onset and whether it was caused by trauma that stimulates the blood vessel wall. If the history of trauma cannot be traced due to various factors, it is necessary to consider the nervous and endocrine system that dominates smooth muscle. Such as the theory of venous stasis proposed by some and the theory of hormonal changes, especially changes in estrogen, are considered one of the etiological factors. The presence of chronic inflammatory cell infiltration was also found in some lesions supporting the theory of venous stasis [2,5]. There is no specific clinical feature, so the rate of misdiagnosis is high, and the clinical manifestations are mostly normal-complexion, independent subcutaneous nodules that are palpable, mobile, temperature-sensitive, and slow-growing, generally no larger than 2 cm, with about half of the patients experiencing pain [5,6]. The pain is pointed out to be the result of local tissue hypoxia [7]. Besides, some studies also found tiny nerve fibers in pain parenchyma. It is speculated that pain results from the tumor compression of the cutaneous nerve [8]. The patient in our case

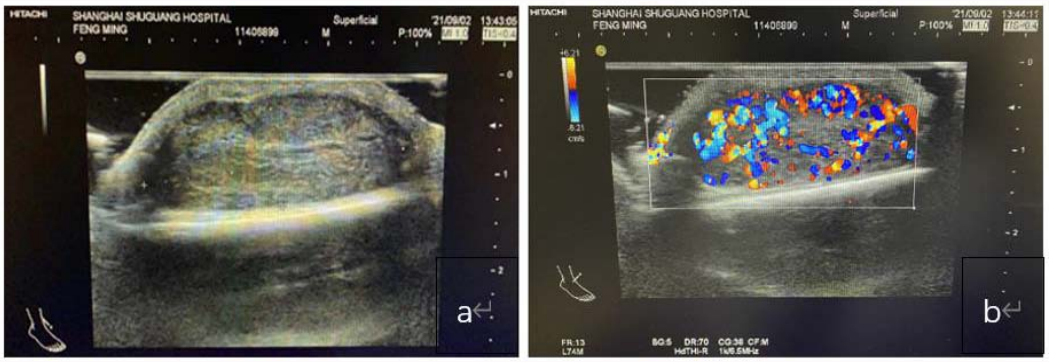

Figure 2: The ultrasonic image and hemodynamic boundary of the patient's skin lesions are clear, the shape is regular, and there are abundant blood flow signals.

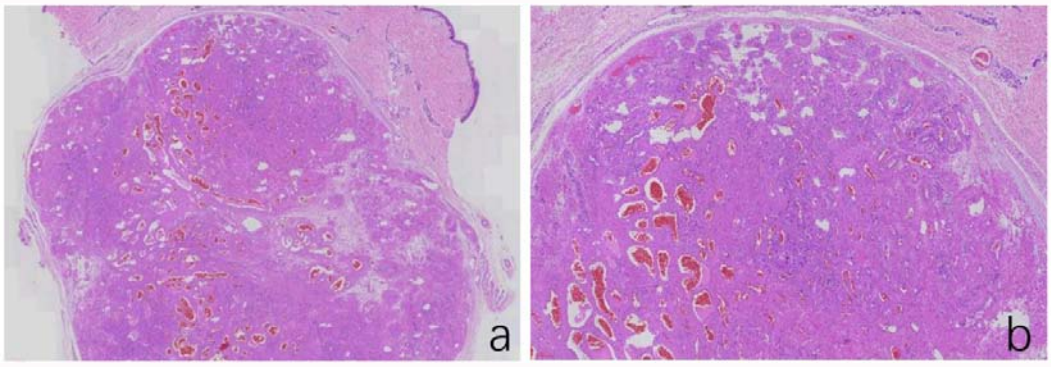

Figure 3: (a) The tumor has a clear boundary and contains abundant bloodstream and smooth muscle cells (HE, x10); (b) The blood vessel wall is thin, filled with red blood cells, and smooth muscle cells arranged in bundles surround the blood vessel wall (HE, x30).

had a long disease duration of more than ten years, a large mass, and a growth site close to the ankle joint without pain sensation for reasons that are not known. Encountering such cases, we need to be differentiated from all nodular lesions of the extremities. However, it is often challenging to give the diagnosis accurately just according to the clinical manifestations documented in teaching materials, and the clinical diagnosis is mainly determined by histopathological features. As we know that smooth muscle contraction could occur when the cold stimulus is encountered, so if a piece of ice is placed on the skin, shrinkage on the surface of the fibroid can be observed soon, which can assist the clinical diagnosis of this disease. Non-invasive ultrasound can also help exclude some diseases that need to be identified. For example, it can be preliminarily distinguished based on the location of the tumor, whether the boundary is clear, the intensity of echo, the degree of echo uniformity, and the blood flow signal. Angioleiomyomas can be treated by surgery. After the operation, it can be diagnosed clearly by pathological examination, and it is usually not easy to recur.

Conclusion

Cutaneous angioleiomyomas are rare in the clinic, cavernous cutaneous angioleiomyomas are the rarest histologic type, and there is no consensus on their etiology. As clinicians, we pay more attention to its clinical diagnosis and treatment. To raise awareness of such tumors, we should consider the possibility of a diagnosis of vascular leiomyoma when clinically encountering such diseases. Use ultrasound or MRI to help diagnose, skilled and complete removal of such tumors, and reduce the recurrence of the disease. To the best of our knowledge, this is the second case to report Cavernous cutaneous angioleiomyoma.

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