A Case Report on Acquired Tufted Angioma with Severe Pain after Healed Herpes Zoster

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To the Editor: A 43-year-old woman presented with mulberry-like plaques on the left side of her back. At the same site, she had acquired herpes zoster approximately 3 months ago and been healed after treatments. The condition started as small plaques and pimples in irregular shapes, followed by the extension of the new lesions with acupuncture-like pain and tenderness upon palpation. The patient had been treated with oral gabapentin and topical glucocorticoid cream. However, the lesions sustained and the pain exacerbated after drug withdrawal. Physical examination revealed a cluster of plaques sized about 4 cm × 7 cm and underneath were some subcutaneous indurations [Figure 1a]. Excision biopsy of the lesion showed that the characteristic "cannonball appearance" was scattered vascular tufts composed of capillaries and endothelial cells in the dermis [Figure 1d]. In addition, multiple channels and slits with erythrocytes and hemorrhagic areas were present in them [Figure 1e]. On immunohistochemical staining, cells of the vascular tufts were positive for CD34, and the membrane of the endothelial cells was stained brown-yellow [Figure 1f]. In addition, hematoxylin and eosin and immunohistochemical staining revealed that S-100-positive nerve cells were surrounded by the vascular tufts [Figure 1g and 1h]. The patient did not belong to a risk group of sexually transmitted diseases and presented no history of bleeding, ulceration, or infection. Hemogram and liver examination revealed no abnormality. According to the clinical symptoms and histological findings, this patient was diagnosed as acquired tufted angioma (ATA) after healed herpes zoster and accepted excision surgery. However, two years after the surgery, the plaques recurred with aggravated pain [Figure 1b]. The patient has undergone surgical excision for the second time and showed no signs of recurrence after that [Figure 1c].

TA was first described by Wilson Jones in 1976 as a kind of benign hemangioma. TA is characterized histologically with angiomatous tufts and lobules scattered in the dermis. TA is classified into two kinds, which are congenital and ATA. Most cases of ATA occurred in adults 30–60 years old. Typical clinical findings are reddish indurated plaques or nodules on the oral mucosa, neck, and upper trunk, which grow slowly for months to years and may cause tolerable pain. Some cases have been reported with histories of transplant, vaccination, and pregnancy, but the causes of ATA remain unclear.

Several nervous and cutaneous reactions that occurred after herpes zoster have been reported. The most common cutaneous reaction is the granulomatous disease followed by malignant tumors.[1] ATA lesion developed after healed herpes zoster has been reported once and considered as an isotopic response.[2] Our case occurred three months after the herpes zoster history, which excludes the possibility of a virus causing the disease. However, varicella-zoster viruses destroy nerve cells and lead to the production of endogenous neuropeptides, such as substance P and vasoactive intestinal peptide, which play important roles in angiogenesis and promote the proliferation of vascular cell as well as endothelial growth factor expression. The pathogenesis of ATA is unclear and may be associated with some vascular growth factors. Therefore, herpes zoster history can somehow explain the onset of ATA in our case. In addition, the patient complained of severe pain, which we considered was due to the excessive vascular hyperplasia that oppressed the peripheral nerves as shown in the histopathological examination result [Figure 1g and 1h]. We also considered the damage that herpes virus did to the nerve might be the reason of the pain.

Numerous therapies have been used to treat ATA. Surgical excision, cryotherapy, and pulsed dye laser were applied as topical treatments. Systemic administration includes corticosteroid, interferon-α 2a, and vincristine.[3,4] All the treatments exhibited different efficacies. ATAs are mostly self-limiting and are seldom transferred to other sites of the body, and asymptomatic treatment is sufficient in most patients. Our patient suffered severe pain so that we performed excision surgery to reduce the symptom. We consider that the incomplete excision is the main cause of recurrence as the lesion area was large, which reminds us that complete excision over visual range is very important in the treatment of ATA. Furthermore, frozen section can help determine the necessity of expanding the surgery scope as well.

This case was reported for its rarity and to indicate that skin caring and diagnosis of isotopic response after herpes zoster or any other skin diseases should be emphasized.

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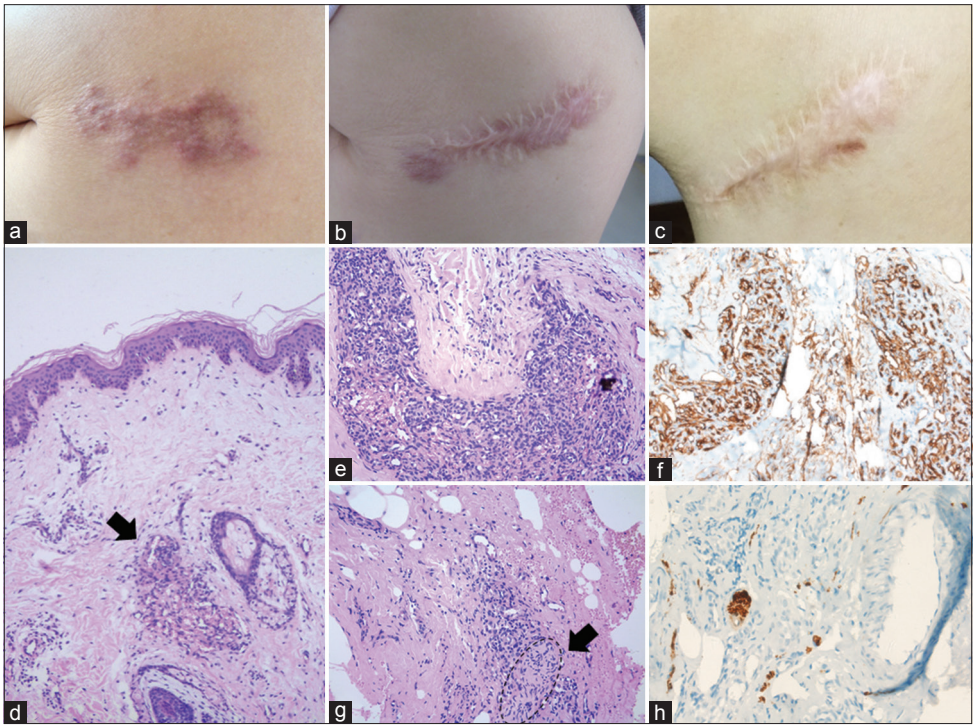

Figure 1: Representative pictures of the lesions of ATA with severe pain after healed herpes zoster. (a) Mulberry-like plaques and pimples on the left back of the patient. (b) Recurred lesions 2 years after the excision surgery. (c) Latest picture collected by return visit examination. (d) Vascular tufts were distributed in the dermis with the characteristic "cannonball" appearance (Hematoxylin and eosin [H and E], ×40). (e) Capillaries and endothelial cells were separated by the slit-like erythrocytes and hemorrhagic areas (H and E, ×200). (f) Vascular endothelial cells were CD34 positive (DAB, ×200). (g) Nerve cells were surrounded by the hyperplastic vascular tufts (H and E, ×200). (h) Nerve cells were S-100 positive (DAB, ×400). ATA: Acquired tufted angioma; DAB: Diaminobenzidine.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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