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Toe Amputation After Minor Surgery in a Patient with Behçet's Disease: A Case Report

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ABSTRACT

Behçet's disease is a systemic autoimmune vasculitis. Although various clinical findings can be observed depending on the pathologic features caused by the blood vessels involved, the classic triad of the disease includes oral aphthae, genital ulcers, and uveitis. Although complications involving the aorta or the vena cava inferior can prove fatal, thrombophlebitis in the superficial veins of the lower extremities are more commonly observed. Some patients can remain asymptomatic for a long period after the diagnosis. In patients with positive pathergy test findings, trauma can trigger the inflammatory cascade. This case report presents a patient with vasculitis that occurred subsequent to minor surgery and led to amputation of the great toe in a female patient with a 14-year old history of Behçet's disease.

Behçet's disease (BD) is a multisystem vasculitis of unknown etiology, first described in 1937 by Dr. Hulusi Behçet (1,2). Although the classic triad of the disease includes oral aphthae, genital ulcers, and uveitis, it can show a wide variety of clinical presentations depending on the extent of the vasculitis. The most commonly observed vascular pathologic feature is thrombophlebitis. The vasculitis more commonly affects veins than arteries (3,4), and vascular lesions are observed 3 times more frequently in males than in females and follow an undulating course with clinical exacerbations and remissions (4). In the present report, we describe the case of a young adult female who experienced a venous circulation disorder after biopsy of a papular lesion localized to her left great toe, which ultimately led to amputation of the toe.

Case Report

A 22-year-old female presented to our clinic with a black and nonhealing papular and necrotic mass in the nail bed of her left great toe (Fig. 1). The nail plate had fallen off spontaneously owing to the mass under it at the initial presentation. Consideration of her history revealed that the mass had been present for approximately 3 months, and it was insidious in onset without a history of trauma or any concurrent diseases that could readily explain the mass. Because the lesion was painful, discolored, and nonhealing, the decision was made to perform an excisional biopsy of the nodule to accurately identify the nature of the tissue.

The surgery was performed with the patient under local anesthesia and through a digital nerve block using 2 mL of plain prilocaine hydrochloride infiltrated at the base of the hallux on the left and right sides and 1 mL on the dorsum of the toe at the metatarsophalangeal joint level. A digital tourniquet made using a sterile nonlatex glove was placed at the base of the digit during the surgery. After the wound was cleaned and dried blood removed, the papular necrotic lesion, which measured approximately 8 mm in diameter, was totally resected. The cortical bone had been invaded 3 mm in diameter. The surface of the bone cortex was curetted, and the surgery was completed through the application of a local rotational flap from the adjacent nail bed. The flaps were raised from the both sides of the defect, and the donor areas were brought closer using 7-0 polyglactin suture to sew the flaps to each other. The duration of tourniquet application was approximately 30 minutes, and no gross circulation complication was observed in the toe during the biopsy and flap coverage procedure. We applied a soft wound dressing with a gauze bandage, and we offered an open-toe box shoe to avoid pressure on the operation field. Because the patient had come from outside the province in which our hospital is located, she was advised to have her dressings changed at a local health facility, and an initial postoperative follow-up examination was scheduled for 3 weeks after the procedure.

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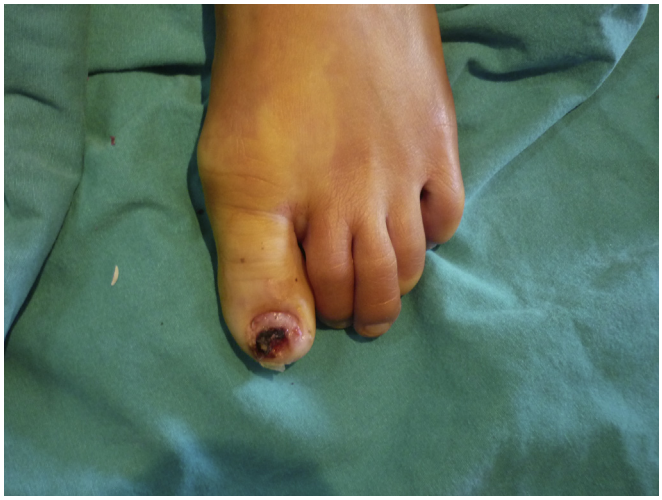

Fig. 1. A black and nonhealing nodular mass was present in the nail bed of her left great toe.

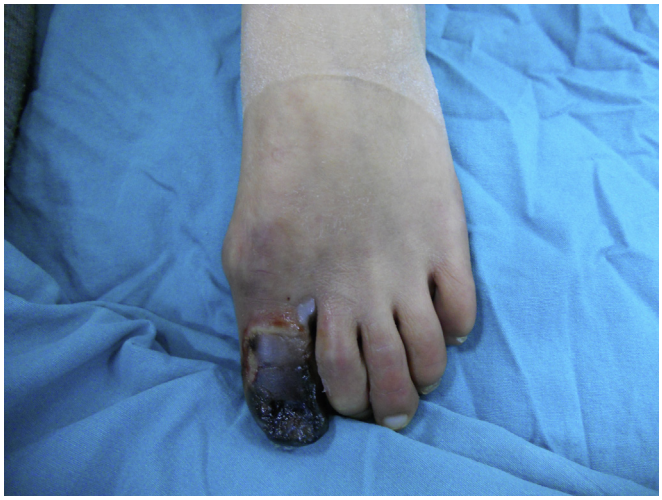

Fig. 2. The cyanotic and necrotic digit on the seventh day of local intervention.

At 1 week after the surgery, the patient returned to our hospital complaining of pain and dark discoloration localized to the operated toe. The examination at that time revealed her left hallux to be cyanotic, and the skin between her proximal phalanx and the tip of her toe was necrotic (Fig. 2). When the patient was again questioned about her history, she revealed that she had been taking colchicine for approximately 14 years for the treatment of BD. She also admitted that she was a nurse by profession and that she had not mentioned her disease or medication use before the biopsy because the surgeons to whom she had previously presented had refused to operate on her hallux when she told them about her BD.

The patient was admitted to our hospital and a regimen of enoxaparin sodium 4000 anti-Xa international units twice daily was started. We also applied a topical anti-inflammatory pomade that consisted of nitrofurazone (Furacin® pomade; Zentiva, İstanbul, Turkey), and her leg was elevated until the necrotic demarcation was ascertained. The pathologic examination demonstrated that the biopsy of the nail bed lesion was consistent with necrotic tissue (Fig. 3). After dermatology and rheumatology consultations and confirmation of her positive pathergy test findings, she was advised to use colchicine 1 mg/day and prednisone 1 mg/kg/day orally. Local wound care, consisting of cleaning with povidone iodine and saline solution, was also initiated. We observed the necrotic tissue without any surgical intervention for the first week to determine clear demarcation lines. After 1 week of follow-up medical treatment and local wound care, the patient's necrotic demarcation line was determined, and the left hallux was amputated at the mid-diaphyseal level of the proximal phalanx, preserving the metacarpophalangeal joint. The surgery was performed with the patient under general anesthesia without a tourniquet and using an approach to cause minimal tissue trauma. The skin was closed primarily using simple interrupted sutures of nonabsorbable monofilament, and a sterile bandage was applied. She was instructed to resume the use of her open surgical shoe. After surgery, infusion of the plasma expander Rheomacrodex® (dextran 2.5 mmol, 150 NaCL mmol; Medisan Pharmaceuticals, Parsippany, NJ) 500 mL/day was started to support the capillary circulation. Until the fourth postoperative day, no pathologic features other than edema were observed at the wound site. However, on the fifth postoperative day, ischemic necrosis became apparent in the amputation stump. Angiography of the left lower extremity showed normal circulation in the dorsalis pedis and posterior tibial arteries, with perfusion extending into the common and proper digital arteries. Furthermore, no evidence of venous pathologic features was observed using Doppler ultrasonography of the left lower extremity. The patient was followed up with daily dressing changes, and 20 days after the first amputation surgery, she underwent revision amputation at the level of the metatarsophalangeal joint, preserving the head of the first metatarsal (Fig. 4). Thereafter, the patient healed unremarkably, with the exception of vague amputation stump pain. She resumed wearing regular shoes at 4 weeks after the revision amputation, and she had resumed her regular activities at 8 weeks after the revision

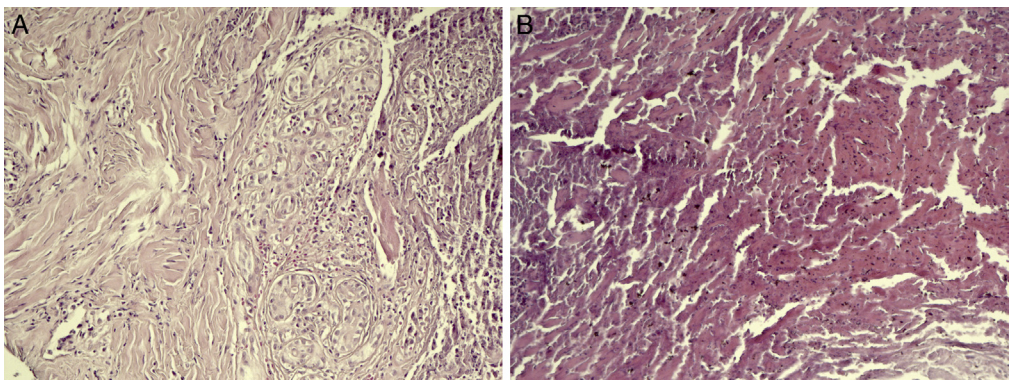

Fig. 3. Hematoxylin and eosin-stained sections were examined using a high-power light microscope. (*A*) Dense neutrophil invasion is seen in all areas of the specimen (magnification ×80). (*B*) View of nonspecific necrotic tissue with inflammatory cells spreading throughout all the tissue (original magnification ×100).