# SPINA BIFIDA: A Twenty-Year Struggle With Osteomyelitis

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## **INTRODUCTION**

Spina bifida is a congenital disorder caused by the incomplete closing of the embryonic neural tube (1, 2). There are two variations of spina bifida described in the literature: spina bifida cystic and spina bifida occulta (2, 3). Spina bifida occulta refers to a vertebral defect without herniation of the spinal canal contents and is most commonly found in the lumbosacral region (3). Patients with spina bifida commonly develop foot deformities such as clubfoot, calcaneovalgus, metatarsus adductus, and congenital vertical talus (1). These patients also have varying degrees of neurologic and musculoskeletal impairment, which ranges from complete paralysis and/or paresthesia to minimal or no impairment (3). The cavus deformity of the foot, in combination with absent sensation along the lateral border of the foot, makes recurrent ulcerations a problem (1). These ulcerations in turn, put the patient at significant risk for developing chronic, polymicrobial osteomyelitis (4).

### CASE REPORT

A 44-year-old female presented to our clinic with a history of a nonhealing ulcer to her left lower extremity, which had been present for 20 years. The patient had a past medical history that consisted of spina bifida with a sequela of peripheral neuropathy as well as deformity to the left lower extremity. During childhood, the patient had a laceration of her left foot while taking swimming lessons at a local pool. Due to her neuropathy, the patient did not notice the laceration until someone pointed out that her left foot was bleeding. During that same year, the patient underwent her first foot surgery; a bilateral tendon transfer aimed at correcting the position of her feet. The patient was put in a cast and her laceration healed during the recovery process. A year after her first surgery, the patient underwent a second foot surgery that consisted of a tendon transfer as well as a triple arthrodesis to her left foot (Figure 1). The triple arthrodesis was not fixated with internal hardware. However, during the postoperative course, the patient's physician became dissatisfied that the foot was stuck in a varus position, and the patient was taken back to the operating room to undergo a calcaneal osteotomy with internal fixation to the left foot (Figure 2). The patient did not experience any recurrence of her left foot wound during the time period between age 8 and 15 years.

At age 15, the patient had another traumatic wound to her left foot while at a local pool. Following this event, the patient was taken to see a physician, and this physician was concerned that there may have been suspicion of osteomyelitis to the left foot (Figure 3). At this time the internal fixation was removed and the patient was put on intravenous (IV) antibiotics for a total of 6 weeks. The ulcer remained closed for a period of 9 years following the removal of hardware.



Figure 1. Lateral radiograph of left foot after triple arthrodesis. Note the supinated position of the foot.



Figure 2. Lateral radiograph of left foot after calcaneal osteotomy with internal fixation.



Figure 3. Lateral radiograph of left foot after calcaneal osteotomy with internal fixation. Note osseous changes to plantar surface of the osteotomy site.



Figure 5. Clinical view of left foot. Note multiple ulcerations with periwound erythema and 50% necrotic/ 50% granular wound base.

At age 24, the ulcer reopened. At this time, the patient underwent a partial calcanectomy with skin flap as well as a 12-week course of IV antibiotics (Figure 4). This treatment series kept the wound closed for another 4 years. The ulcer reopened again for a third time at age 29, and at this time, the patient was recommended to have a total calcanectomy by her physician. It was at this point that the patient decided to visit our clinic for a second opinion in June 2011.



Figure 4. Magnetic resonance image of left foot after partial calcanectomy.



Figure 6. Lateral radiograph of left foot on first clinical visit. Notice destructive osseous changes to the calcaneus.

The patient presented to our clinic having already undergone a magnetic resonance image of the left foot and ankle, which was negative for osteomyelitis. However, the patient did present with an open ulcer that showed clinical evidence of an infection (Figure 5). There were chronic osseous changes noted on radiographs to her left rearfoot (Figures 6, 7). The patient was admitted to the hospital at this time, and an infectious disease doctor was consulted. During this hospital stay, the patient underwent a surgical procedure that consisted of soft tissue debridement and bone biopsy of the calcaneus. The biopsy report came back with a positive result for osteomyelitis of the calcaneus. IV antibiotic therapy was ordered for a total of 8 weeks. Wound VAC therapy was also implemented. The patient was instructed to be non-weightbearing to her left lower extremity. This series of treatments failed to close the patient's wound.

The patient subsequently underwent multiple bone and soft tissue debridements over the next 3 years. During the healing process, wound VAC therapy was used twice, another session of 8 weeks of IV antibiotics was also initiated. At one point during the treatment course, the patient developed a palmoplantar wart at the ulcer site that healed without any complication. Different attempts for offloading the left lower extremity were utilized, including the use of postoperative shoes, CAM Walker with plastizote inserts, crutches, and also the use of a Toad Anti-Gravity Brace Of the different devices used, the CAM-walker boot was the one that better fit the patient's needs. Before the last surgical procedure, the patient developed a well



Figure 7. Inversion lateral radiograph of left foot at first clinical visit. Notice destructive osseous changes to the calcaneus.



Figure 9. Computed tomography image prior to patient's last surgery. The calcaneus has been almost entirely removed.

circumscribed, invaginated ulcer cavity that prevented the area from filling in with granulation tissue. The last surgery that we performed proved to be successful at closing and healing the ulceration. The last surgery consisted of resection of much of the remaining calcaneus to eliminate the non-viable bone, while still keeping the Achilles tendon insertion intact. The wound was also primarily closed at this time (Figures 8, 9). It should be noted that amputation of the patient's left lower extremity was discussed during the course of treatment. Reconstructive surgery with external fixation was also discussed. The patient remained adamant throughout the treatment process that neither of these options were a consideration. As of six months following her last surgery; the patient's wound has remained closed and her radiographs have remained stable (Figures 10, 11, 12).



Figure 8. Clinical image after first postoperative course of surgical debridement and wound VAC therapy. Wounds are invaginated and have failed to close completely. There is still erythema noted.



Figure 10. Clinical view after last surgery. Primary closure was performed after debridement of all nonviable bone and soft tissue.



Figure 12. Lateral radiograph of left foot demonstrating position of foot and remaining bone 6 months following all surgical procedures.

#### DISCUSSION

Neuropathic ulcerations describe those lesions associated with spina bifida, spinal cord lesions, peripheral nerve injuries and leprosy (5). The abnormal distribution of load, leading to local areas of peak pressures is the cause of many neuropathic ulcers (5). These ulcers are usually slow to heal, and may not respond to treatment. This infection may in turn to spread to the underlying bone (5,6). Osteomyelitis is an inflammatory process accompanied by bone destruction and caused by infecting microorganisms (6,7). Osteomyelitis due to local spread from a contiguous contaminated source of infection follows trauma, bone surgery, or joint replacement, and implies an initial infection that gains access to bone (6-8).

The most significant risk factor for developing osteomyelitis is the presence of a pre-existing ulceration in the insensate foot. A severely dislocated and unstable foot or



Figure 11. Clinical view of the closed ulceration.

ankle may also be a predisposing factor (9). Osteomyelitis may be difficult to treat. Conservative treatment for osteomyelitis includes early administration of broad-spectrum empiric antibiotics. Antibiotic therapy can be tailored by an infectious disease specialist as culture and sensitivity data become available (3,8). Chronic osteomyelitis, however, generally cannot be eradicated by IV antibiotics alone. At this point, surgical intervention is initiated, and the goal of surgery is to achieve a viable vascularized environment and eliminate dead bone (6-8). In patients with adequate blood supply and oxygen tension at the infected site, debridement of bone with a 4 to 6 week course of antibiotics should be adequate treatment for osteomyelitis. However, in those patients with poor vascularity and oxygen tension, the wounds fail to heal and amputation of the infected foot is eventually necessary (7).

In conclusion, spina bifida is a congenital defect of the spinal cord that causes deformities of the foot combined with neurologic impairment. Due to these impairments, these patients are at risk for developing neuropathic ulcerations with underlying osteomyelitis. For our patient, the chronicity of the osteomyelitis combined with her underlying condition, made closure of her chronic wounds difficult. Since our patient had adequate blood flow, it was possible to close her wounds once all nonviable bone was removed. This case was made challenging by that fact that multiple surgeries put the patient at risk for spreading of the infection as well as the possibility of amputation. However, our main goal as foot and ankle surgeons should be to treat the patient with the ultimate goal of preservation of lower extremity function.

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