Invasive Squamous Cell Carcinoma Secondary to Chronic Osteomyelitis

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Statement of Purpose

Chronic osteomyelitis of the tibia with malignant transformation from superficial ulcerations is a rare occurrence. This case highlights a rare neoplasm that formed due to a chronic ulceration secondary to chronic osteomyelitis. Our patient was taken to the operating room multiple times for debridement's and cultures. Each time cultures were different. The Infectious disease team provided their expertise and treatment each time cultures were taken. After all treatment modalities were exhausted, patient was scheduled to received a below knee amputation for definitive management.

Literature Review

Squamous cell carcinoma is an uncommon complication of chronic osteomyelitis with an associated sinus tract. The site most commonly associated is the tibia, followed by the femur. When the tumor invades the bone, osteolysis or a pathological fracture is evident. A recent study in 2011 reported seven cases of chronic osteomyelitis related squamous cell carcinoma between 1993-2005. The pathological examination showed five cases of a well differentiated squamous cell carcinoma with bone invasion and 2 cases of invasive squamous cell carcinoma. Definitive treatment consisted of amputation of the affected limb. The amputees did not experience local recurrence of metastasis dissemination within a period of five years. [I]

Due to the possibility of lymphonodular metastasis radical resection is the goal of treatment. The prevalence of malignant transformation from a chronic ulceration in the setting of chronic osteomyelitis ranges from 1.6% to 23%. The increase in fistulous drainage, as well as persistent exophytic growth of an ulcer can be warning signs of malignant transformation. [II]. Reactivation of osteomyelitis, even after a 50 year disease free interval has been reported in literature. The reoccurrences are usually by the same organisms. A different bacterium responsible for a second infection at the same anatomical site is proof positive for a true re-infection. Uckay et al 2016 showed case reports of patients who has osteomyelitis reactivated by different bacteria. Previously infected bone must be considered a lifetime focus of diminished resistance. (III)

Case Study

A 50 year old male presented to our facility with a chief complaint of a chronic non-healing ulceration to his right ankle with an increase in purulence and erythema. He had a history of a right ankle fracture with open reduction and internal fixation in 1993 at an outside hospital. (figure 1) An MRI was ordered that showed early abscess formation, but unable to exclude osteomyelitis due to hardware interference. Patient was scheduled for operative debridement and irrigation with removal of hardware. Intraoperatively abscess formation was confirmed with an additional finding of a large bone void filled with caseous material. Soft tissue and bone cultures were taken and sent for pathology and microanalysis. Intraoperative pathology was positive for osteomyelitis. Patient was started on IV abx for 6 weeks by the infectious disease team targeted towards culture results. Xrays were taken every other visit to assess osseous erosions. Osteolysis was noted along the medial and anterior distal tibia. (figure 2) Patient was followed weekly in the clinic for local wound care. (figures 3 and 4) Patient was taken to the Operating room for a second irrigation and debridement. At that time new cultures were taken. Three new organisms were grown with the addition of yeast. Patient was started on a new course of long term abx with 3 months of Fluconazole. Due to the chronicity of the ulceration a second MRI was ordered to evaluate the extent of the infection (figure 5) MRI showed a large 3 cm x 3 cm x 3 cm erosion of the tibial plafond extending into the tibial metaphysis and an intramedullary abscess extending to the talus. At that time due to the extent of tibial erosion and failure of abx therapy patient was scheduled for below knee amputation. Surgical pathology from the Below knee amputation showed an invasive keratinizing squamous cell carcinoma located within a Marjolins ulcer. The amputation site healed uneventfully, the patient did not have any evidence of metastasis of carcinoma.

Discussion

This case highlights a rare malignant pathology secondary to chronic osteomyelitis after an ankle fracture. When a malignant soft tissue carcinoma invades deep, osteolysis is observed. With this case in particular, multiple times bone was sent for gross analysis. Each time was shown to have chronic osteomyelitis. The malignancy was observed when the limb was sent for gross analysis.

Figures

Figure 1

Figure 2

Figure 3

Figure 4

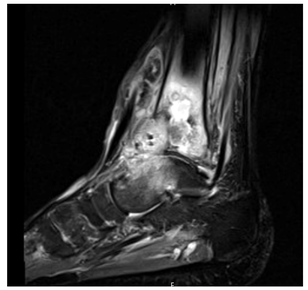

Figure 5

References

I) Qinghu Li-Haomin, et al. "Squamous cell carcinoma resulting from chronic osteomyelitis: a retrospective study of 8 cases" *International Journal of clinical and experimental pathology* (2015): 10178-10184.

II) Tozzi, James E., et al. "Squamous Cell Carcinoma Secondary to Chronic Osteomyelitis" *The Iowa Orthopedic Journal* (1985): 103-106.

III) Uckay Ilker, et al. "Recurrent Osteomyelitis Caused by Infection with Different Bacterial Strains withouto obvious Source of Reinfection" Journal of Clinical (2006): 1194-1196.