

CASE REPORT

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Adenosquamous Cell Carcinoma Secondary to Chronic Osteomyelitis of the Tibia - Rare Case Report

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1 Background

Osteomyelitis is an infection with progressive destruction of bone tissue involving the bone and articular cartilage with its medullary canal. Chronic osteomyelitis treatment is challenge for the orthopaedician. The bone microscopic anatomy and also the microorganism biofilms and their ability to adapt to their surrounding environment. It's tough for the antibiotics and disinfectants to breach the biofilm barrier and kill these microorganism colonies.

Cutaneous neoplasm are a rare complication developing after a chronic bone infection. The incidence and prevalence of these cutaneous tumor/neoplasm following chronic bone infection continues to increase despite the increasing awareness to the disease process, and medical advances in early diagnosis and early treatment. Most common neoplasm after a chronic bone infection are squamous cell carcinoma and rarely sarcoma or lymphoma.¹

In our study we report adenosquamous cell carcinoma arising from chronic osteomyelitis of tibia for which above knee amputation was done.

2 Case

Patient sustained open injury to the left leg following Road traffic accident in 1992 and was treated with ilizarov fixator and later skin grafting was done. Patient noticed discharge sinus from the left leg in 2000, which was insidious in onset, gradually progressive, on-blood tinged, non-foul smelling. In 2010 the patient underwent debridement and antibiotic bead application for chronic osteomyelitis of left tibia, following which the discharge initially reduced and later again aggravated and foul smelling. Patient noticed abnormal mass growth of left leg from wound site which was increased in size since 4 months.

On examination Cauliflower like growth with Ulcer of size 8x9 cm present over the anterior, middle third aspect of left leg. Ulcer contains hypertrophied soft tissue soft with everted edges with slough over the base which bleeds on touch. Active seropurulent discharge present, foul smelling, blood tinged, greyish yellow in colour.

Superficial Inguinal lymphatic nodes are palpable over the left inguinal region. Initial blood investigations and radiographs were done confirmed chronic osteomyelitis of left tibia. Initial biopsy reported as adenosquamous cell carcinoma. Inguinal lymph

node FNAC showed features suggestive of reactive lymphadenitis. Patient condition and treatment plan was explained. Patient underwent left side above knee amputation with superficial inguinal lymphatic nodes removal in January 2021. Histopathological report confirmed Adenosquamous cell carcinoma. Amputation stump healed well. 6 months later the patient developed multiple inguinal lymph node enlargements in the left thigh. Biopsy revealed to be adeno squamous cell carcinoma. Patient was not willing for chemotherapy and later in the same month patient demised.



Fig 1.



Fig 2.



Fig 3. Post-operation image

3 Discussion

Neoplasm is an outcome of chronic insults to the normal tissue. Aulus Cornelius Celsius observed tumoral neoformations in correspondence to chronic wounds.² During the middle of 19th century, Hawkins with Marjolin correlated skin diseases with osteomyelitis.³ A Mayo Clinic study in which their investigators analysed around 4000 cases of chronic osteomyelitis, out of which malignant lesions were noted in 23% of the patients.⁴ The incidence of cutaneous lesions varies in between 0.2% and 1.7% of total patients who are affected with chronic osteomyelitis.^{5,6} Trauma is the most common cause of osteomyelitis, in developing countries, there is increase in percentage with delayed diagnosis and inadequate treatments.⁷ Infection rates vary from 4% to 64% in open fracture of the long bone, and their incidence is still increasing.⁸ The time from onset of osteomyelitis is most likely to be the main factor related to carcinogenesis onsetting, with latency period of 20 years (minium) or more.⁹ Males are more often than females in incidence and prevalence of the disease, with a predominance of 85%, and are typically aged in between 50 - 60 years old.¹⁰ To avoid local invasion/metastatic spread, treatment should never be delayed. The pathogenesis of neoformation is still under discussion, but the most widely and commonly accepted theory is based on the chronic inflammatory state in which inflammatory mediators and cytokines which are expressed by the immune system to regulate the genic expression of various proteins, including p53.¹¹ Gram-positive organisms can be replaced by predominant gram-negative flora that produce endotoxins associated with cancer.¹² Squamous cell carcinoma is characterized by an intraepidermal proliferation of atypical keratinocytes¹³ when it is associated with osteomyelitis which is usually of a low-grade malignancy.¹⁴ Magnetic resonance imaging can be helpful in differentiating squamous-cell-carcinoma with soft tissue neoplasm of other origins.¹⁵ CECT can justify the suspicion of metastasis, commonly to the lungs. Biopsy can confirm the diagnosis. Patients with chronic osteomyelitis with recurrent exacerbations under-go frequent admissions to hospital, pharmacological agent's exposure, and multiple surgical procedures in their entire lifetime. Multiple authors based on their experience have suggested that amputation is the definitive treatment.^{5,16,17} In the absence of metastasis, it may be possible for evaluation with a wide excision of the lesion. Our patient attenders did not accept the amputation at first as he is the sole earner in the family but after explaining the tumour risks of metastasis, they agreed for it. Our patient underwent a superficial inguinal lymph node dissection of the affected side to rule out metastasis which was reported as reactive lymphadenitis.

4 Conclusion

Adenosquamous cell carcinoma is one of the rare complications following chronic osteomyelitis with unknown etiological features requiring utmost attention in diagnosis and plan of management for better earlier outcome.

Amputation is the most accepted and appropriate treatment for adenosquamous carcinoma occurring in patients with chronic osteomyelitis. To avoid risks of lymphonodular and organ metastasis, this lymphadenectomy surgical procedure should not be delayed. Early diagnosis with timely therapy could have prevented further progression and amputation.

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