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Squamous Cell Carcinoma Arising from Chronic Osteomyelitis Case Report and Review of Literature

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SUMMARY

Squamous cell carcinoma arising from discharging sinuses of chronic osteomyelitis is a rare entity. With proficient use of appropriate antibiotics in the acute stage of osteomyelitis, and adequate management of chronic osteomyelitis, this dreadful complication has almost disappeared in the developed world. In countries of the third world, we still come across discharging sinuses associated with complications. Amputation followed by radiotherapy is the usual management. Limb salvage is possible in lesions with limited extent.

CASE REPORT

A fifty year old male, laborer by profession, presented with pain and discharging sinuses, left thigh for the last thirty years. The problem started after he sustained a trivial injury while wrestling. He developed pain, swelling and fever off and on. Incision and drainage was performed by a local bone-setter. This lead to chronic discharging sinus. He took irregular treatment at different hospitals. The sinus did not heal.

One year back, he fell down and sustained injury to the left thigh. He had pain and inability to bear weight on the left lower limb. He was admitted in a hospital and was diagnosed as a case of pathological fracture after osteomyelitis. Debridement and curettage was performed and left femur was stabilized with AO tubular external fixator. There was no other significant illness in the past. The wound did not heal and foul smelling discharge continued after repeated dressings. He was referred to our hospital for further management.

On examination, he was thin built, pale looking, with normal vital signs. The cardiovascular, respiratory, and abdominal examination was unremarkable. Examination of left lower extremity revealed multiple discharging sinuses at the posterolateral aspect of middle third of thigh and foul smelling pus coming out. The central sinus was 3-4 cm in size with raised margins. There were scar marks of previous surgery. AO tubular external fixator was on the anterior aspect of thigh. Neurovasular was intact in the left lower extremity and lymphnodes were not palpable. Range of movement was normal at left hip while left knee was stiff in extension.

Routine investigations were performed which revealed hemoglobin of 8.3 gm/dl, WBC 11500 and ESR of 45 mm Ist hour. The blood urea, serum creatinine, blood sugar, serum electrolytes, routine urine examination, and x-rays chest were normal. In the x-rays of left femur (Fig. 1), there was a radiolucent area in the middle third of shaft with a pathological fracture. Diagnosis of chronic osteomyelitis with pathological fracture of left distal femur was made. Blood transfusions were given and patient was scheduled for debridement and curettage.

At the time of surgery, the involvement of shaft of left femur looked extensive. About 20 cm of bone was resected and sent for histopathology. The stabilization was performed with the help of AO tubular external fixator with Ilizarov attachment (Fig. 2). The plan was to perform an osteotomy after the infection was controlled and fill the defect with segment transport. The report of histopathology demonstrated well differentiated infiltrating keratinising squamous cell carcinoma with extensive

Fig. 1: X-rays of left femur showing radiolucent area in the distal third of shaft with a pathological fracture.

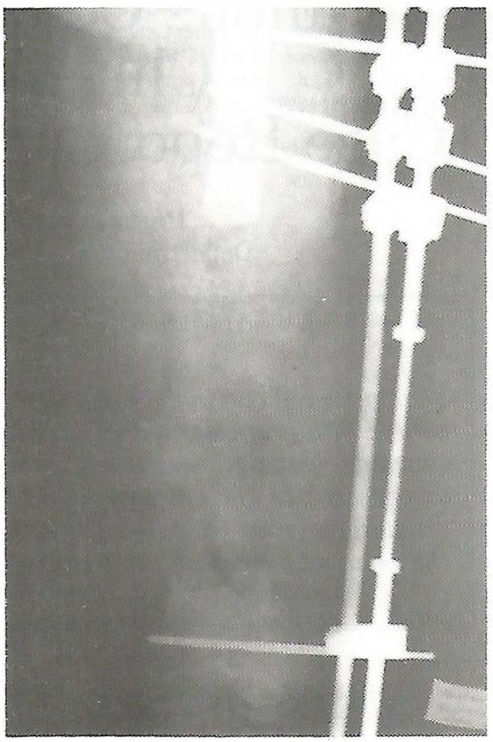

Fig. 2: X-rays after resection of 20 cm of femur and stabilization the help of AO tubular external fixator with ilizarov attachment.

bony involvement. The plan was changed and high above knee amputation was performed. On careful histological examination, the proximal end of resected specimen was found free from tumor tissue. The recovery was smooth and uneventful. The patient received radiotherapy after healing of stump wound.

DISCUSSION

Squamous cell carcinoma or Marjolin's ulcer is the malignant lesion which develops in a burn scar or chronic fistula. It usually develops on surfaces covered with squamous epithelium particularly as a result of chronic irritation, chronic ulcer or chronic discharging sinus. In fistulous tracts, it also develops in fistulas lined by stratified squamous epithelium. Due to the low prevalence of this lesion, there has been disagreement regarding its clinical features, methods of treatment, and prognosis. This rare complication (0.2 per cent-1.6 per cent)[1] most frequently involves the lower limb, although unusual sites like sacrum[2], toe[3], ankle[4], mandible[5] have also been reported. Most patients are men between 50 and 60 years of age. It is associated with long-standing osteomyelitis. Mean reported latent period is thirty years, and it varies from 4.5 to 50 years[6]. It is most frequently a squamous cell carcinoma or rarely a sarcoma. Other tumor reported in long standing osteomyelitis are malignant fibrous histiocytoma, epidermoid cancer, Ewing's sarcoma, and angiosarcoma.

The features indicative of malignant change are previous innocuous sinus tract and foul smelling persistent discharge. However, the diagnosis is confirmed by biopsy of the lesion. The biopsy should be from various depths including bone marrow.

Hahn et al[6] evaluated 19 patients with Marjolin's ulcer from 1970 to 1985. The results were as follows: The previous lesion was a burn scar in 52% of the cases and a fistula of chronic osteomyelitis in 32%. The mean latent period was 31.5 years. The initial symptoms were increased pain (74%), discharge with foul odor (68%) and bleeding (58%). Upon histological examination, all of the cases

were squamous cell carcinoma. The rate of metastasis at the time of diagnosis was 32%. Of the 16 patients treated by surgery, local recurrence was noted in 4 cases. Three of these cases were patients who had been treated by excision and split thickness skin graft. The time interval for local recurrence ranged from 6 months to 11 months (average 8.8 months). The conclusion was that, the squamous cell carcinoma of marjolin's ulcer has a worse prognosis than other squamous cell carcinomas and it requires aggressive treatment.

Retrospective analysis of seven cases by Su-JI et al[7] of chronic osteomyelitis complicated with squamous cell carcinoma (SCC), who were treated at Chang Gung Memorial Hospital Taiwan from 1983 to 1991 revealed 6 males and one female, with the age between 43 and 80 years (average 57 years). The distribution of these lesions was 5 in the lower legs and 2 in the thighs; one had another hip lesion. The duration of discharging sinus presented before malignant change was from 4.5 years to 50 years (average 30 years). The pathological findings were all well-differentiated squamous cell carcinoma. Amputation was performed in 5 of them. Limb salvage procedure was selectively done in one case. Chemotherapy and regional irradiation without surgery was given in the remaining case. Follow up examination showed that the short term results were satisfactory in all cases. Six cases had at least one year of follow-up; the longest follow-up was 8 years.

Many authors have recommended amputation[8,9] or disarticulation[10] as the treatment of choice for locally invasive disease. Limb salvage is possible when tumor is diagnosed early and it has limited extent. Recently, good results with microvascular techniques used for the management of squamous cell carcinoma arising as a complication of chronic osteomyelitis have been reported by Ueng-WN et al[11]. A 61-year-old man with 42-year history of chronic osteomyelitis of the left tibia was found to have malignant change. Local wide excision was followed by staged microvascular latissimus dorsi muscle flap and fibular osteoseptocutaneous flap transfer. The patient remains well 36 months later with no clinical or radiological evidence of local recurrence or distant metastasis. This kind of limb-sparing treatment is an excellent alternative treatment for malignancy.

Cases of squamous cell carcinoma arising in the draining sinus tracts of chronic osteomyelitis have been reported[12,13] that were treated successfully with Mohs micrographic surgery (MMS). After ten year follow-up the patient remained tumor free and continued to enjoy use of his leg. MMS as a therapeutic option to amputation for control of locally invasive disease has also been recommended.

Squamous cell carcinoma may be associated with hormone changes in the human body. Fenton et al.[14] have reported a case of chronic osteomyelitis complicated by the development of squamous cell carcinoma which was itself associated with the development of humoral hypercalcemia of malignancy. Common misconceptions relating to the interpretation of serum parathyroid hormone levels led to the misdiagnosis of hyperparathyroidism and parathyroid exploration. Forced saline diuresis, mithramycin and oral phosphate supplements were not able to provide long-term control of the hypercalcemia but the patient was subsequently managed successfully with intravenous (3-amino-1-hydroxypropylidene)-1,1-bisphosphonate (APD) and resection of the causative tumor.

The frequency of metastatic spread is about 30 percent[1]. Metastases should be excluded by x-ray of the chest, scintigram and CT of the regional lymphnodes. Malignant degeneration appears to be a local phenomenon, and if diagnosed early, has a rather favorable outcome following complete excision. The burn scar or chronic fistula that occurs in elderly patients especially requires more adequate treatment and close observation.

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