

# Chronic osteomyelitis in the lower extremity predisposing to the unusual formation of keloids

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## ABSTRACT

**Keloids commonly occur in black and pigmented races as well as in young adults, but rarely affect Caucasians and albinos. While they have a predilection for the upper trunk as well as the head and neck regions, they seldom occur in the lower limbs. A six-year-old boy presented with multiple fibrous nodular swellings of the right leg and a discharging sinus over the ankle. Closer evaluation revealed underlying chronic osteomyelitis complicated by multiple huge keloids over the leg and ankle. Associated chronic inflammation had resulted in the huge keloids. Our patient's age, site of occurrence and presentation were not typical. In the event that a keloid is presented in isolation, chronic osteomyelitis should be considered as a differential diagnosis, and a high index of suspicion is required in order to establish the diagnosis.**

**Keywords:** keloid, osteomyelitis, lower extremity

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## INTRODUCTION

Keloids are thick scars of the human skin or cornea that are produced by the deposition of excessive amounts of collagen over the area for prolonged periods.<sup>(1)</sup> They involve excessive overgrowth of abnormal, immature collagenous tissue, resulting in a firm mass of tissue. They are associated with abnormal prolongation of the proliferative phase of wound healing. The trauma that initiates keloid formation may sometimes be insignificant, and some keloids may arise spontaneously.<sup>(2)</sup> In normal wound healing, there is an optimum balance between the deposition and lysis of collagen. This regulation is defective in patients with keloids and hypertrophic scars. Accumulation of collagen may result from excessive collagen synthesis or decreased collagen degradation; the former has been favoured more than the latter.<sup>(3)</sup>

Despite the body of research into the aetiology of keloids, the precise mechanism or initiating factor of keloid or hypertrophic scar formation still remains unknown. Some theories that have been propounded

to explain the aetiology include factors such as race, age, direction of scar, infection, hypoxia, and genetic, immunologic, endocrine and other factors.<sup>(2,4)</sup> A keloid progressively invades the surrounding normal tissue and continues to grow. In hypertrophic scars, the scar tissue remains confined to the area of tissue damaged by the initial injury, and increases in size only by pushing out the margins of the scar and not by invasion of the surrounding normal tissue; the scar tends to regress over time.<sup>(2,5)</sup> For reasons yet unknown, keloid and hypertrophic scar formation is unique to mankind. It has not been observed in albinos of any races, while pigmented races are at higher risks of developing such conditions.<sup>(1,2,4)</sup>

Infection leading to chronic inflammation in the skin could predispose to keloid formation; the infection may arise from the soft tissue or bone. Chronic osteomyelitis is primarily an infection of the bone, but it often progresses to affect the surrounding soft tissue, with spontaneous discharge of pus through the skin in most, if not all, cases. It is an infection that is difficult to treat. A lack of blood supply to the sequestrum and the scarring of the surrounding soft tissues with resultant poor vascularity render antimicrobial treatment less effective in halting the process; hence, there is always associated prolonged chronic inflammation. This chronic inflammation in the sinus track has been reported to lead to malignant transformation into a marjolin ulcer.

There is a paucity of information in the literature regarding the formation of keloid around the sinus track of chronic osteomyelitis.<sup>(6)</sup> We present the case of a six-year-old boy with keloidal lesions on the leg, which were found to be complicating chronic osteomyelitis of the affected part.

## CASE REPORT

A six-year old Nigerian boy was referred by the Plastic and Burns Unit of our hospital. He presented with discharging sinus over the right ankle since the past 14 months and multiple swellings on the right leg since the past eight months. The condition was accompanied with high-grade fever associated with chills and rigor and a painful swelling over the distal third of the patient's right

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leg. The swelling increased in size and subsequently resulted in the discharge of seropurulent fluid. The patient had initially presented to a health centre, where antibiotics and wound dressings were prescribed, which resulted in some improvement in pain and swelling. There was no history of surgical drainage.

Over the following two months, the patient developed two other swellings in the right leg that discharged pus, which subsequently healed. Eight months before presentation, the patient had developed two progressive swellings over the scars of the discharging sinuses, which were initially the size of peanuts, with associated itching. Subsequently, the other sinus scars developed overlying masses. There was recurrent ulceration of the swelling over the ankle that was associated with difficulty in ambulation. There were no swelling in other parts of the body. The patient reported having had minor trauma from a fall in the past, which healed without complications. He had also completed the childhood immunisation programme, without any swelling over the injection sites. His father, however, had a presternal keloid.

Physical examination revealed a healthy-looking boy who was afebrile and had enlarged inguinal lymph nodes. No abdominal organomegaly was noted. Both his upper limbs and lower left limb were normal. He walked with an antalgic gait, with about 5° of equinus in the right ankle. He had four keloid masses on the anterior and medial aspect of the right leg, with the most distal mass overlying the ankle measuring 7 cm in its greatest dimension; it had surface ulceration and was discharging seropurulent fluid. The other masses were not ulcerated (Fig. 1). The patient had a real limb length discrepancy of 2 cm (real length right lower limb = 49 cm, real length left lower limb = 51 cm) and an apparent limb length discrepancy of 1 cm. There was no neurovascular deficit in the limbs, but his right ankle was ankylosed. Radiography revealed features of chronic osteomyelitis of the right tibia and fibula, anterior bowing of the tibia, destruction of the right tibial and fibular distal physes and epiphyses, synostosis of the distal right tibia and fibula, avascular necrotic changes in the right talus as well as a soft tissue mass pushing the mid and fore foot into plantar flexion at the mid foot (Fig. 2).

Laboratory results revealed that the patient's haemoglobin genotype was AA. The packed cell volume was 33% and the white cell count was 13,100 (neutrophil 26%, lymphocytes 64%, eosinophils 10%). The erythrocyte sedimentation rate was 50 mm/hr (Westergren method). The patient underwent sequestrectomy, with removal of the keloid overlying the ankle. Microscopy, culture and sensitivity (MCS) of the intraoperative sample yielded



**Fig. 1** Photograph shows keloid masses overlying the sinus tracks of chronic osteomyelitis of the right tibia and fibula.

*Staphylococcus aureus* and *Proteus* spp. Infection control was achieved, and the patient's wound healed. Following surgical excision of the keloids and healing of the wound, pressure bandaging was the mainstay of treatment in order to prevent recurrence. No radiotherapy or steroid was given locally or systemically. The patient was being followed up at both the Plastic and Orthopaedic Surgery Outpatient Clinics when this paper was being written. A shoe raise has been offered for his limb length discrepancy in the interim.

## DISCUSSION

The patient exhibited several typical manifestations and complications of chronic osteomyelitis. These included synostosis of the right distal tibia and fibula, physis and epiphysis destruction, ankylosis of the right ankle joint and avascular necrosis of the right talus from ankle septic arthritis and limb length discrepancy, which was likely to be progressive. Malignant transformation in sinus tracks is seen in longstanding chronic osteomyelitis, usually in adult patients. Our patient presented with keloidal masses over his sinus scars, which was an unusual manifestation of chronic osteomyelitis in all age groups. The identified predisposing factors in this patient included race, a family history of keloids and the chronic inflammation imposed by chronic osteomyelitis of the affected leg. A family history of keloids is found in up to 50% of keloid patients; such patients have a significantly higher incidence of multiple keloids.<sup>(7)</sup>

Hunt et al<sup>(8)</sup> observed that fibroblasts do not multiply in the absence of macrophages. The chronic inflammation found in chronic osteomyelitis provided an abundant source of macrophages to induce the fibroblast. Tissue hypoxia imposed by the relative avascularity of scarred tissue has also been suggested.<sup>(2,4,8)</sup> The fact that this patient had other minor wounds on the body that were not complicated by keloid or hypertrophic scarring



**Fig. 2** Plain radiograph of the right tibia and fibula shows anterior bowing of the tibia, synostosis of distal right tibia and fibula, avascular necrotic changes in the right talus and soft tissue masses.

underscores the role of chronic inflammation in the aetiopathogenesis of keloid formation in this patient. The leg and ankle are not common sites for the development of keloid in the body.<sup>(2)</sup> Keloids may form at any age, but peak incidence occurs at 10–30 years of age.<sup>(9)</sup> In this patient, the keloids formed over the sites of the discharging sinuses of chronic osteomyelitis quite early, i.e. in the first decade of life. Infection has been proposed as a possible mechanism in keloid formation. This case supports the role of infection in the aetiopathogenesis of keloids. Keloids arising from chronic osteomyelitis in this situation had a poorer prognosis, as chronic osteomyelitis is a difficult infection to cure. As such, the resultant keloid was difficult to treat.<sup>(2,6,10,11)</sup>

Intralesional steroid injection, which is the most commonly used modality of treatment, was not applicable to this patient, as it could flare up the underlying chronic osteomyelitis, with propagation of the keloidal process. The multiplicity and large size of the keloids could also contraindicate steroid injection due to the large steroid dose required, causing a higher risk of developing complications from the steroid treatment. There is also a low risk of developing malignancy following irradiation of keloids, especially in the younger age group. The location of the keloids in the extremity allowed for pressure bandaging after excision. This patient responded well to pressure bandaging of the excised keloid as a preventive measure against recurrence.

The best treatment for keloid is prevention. No single modality of treatment has been effective in all cases. The first line of treatment in most cases is intralesional injection of corticosteroids. Surgical excision, when used in combination with steroid injection or radiation therapy, has been found to be effective. Other treatment modalities that are useful include compression therapy and the use of occlusive dressings with silicon sheet or non-silicone occlusive dressings. Some experimental treatments that are on trial for the treatment of keloid include cryotherapy, laser therapy, imiquimod, 5-fluorouracil, bleomycin, retinoids, calcium channel blockers, mitomycin C and interferon- $\alpha$  2b, with variable success rates. At present, a combination of treatment modalities has been most effective; however, the search for the most optimum treatment continues.<sup>(2,4,10-18)</sup>

Measures aimed at curing chronic osteomyelitis in addition to other multimodality treatments for keloids would be crucial in achieving positive outcomes in this challenging condition. In conclusion, when evaluating a patient with keloids for the underlying cause, chronic osteomyelitis should be considered in the differential diagnosis. A high index of suspicion is required, and treatment can be challenging. The prognosis for cure would depend on adequate treatment of the underlying chronic osteomyelitis.

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