Perforating osteoma cutis: a case study

Key words:
- *Congenital dermal sinus*
- *Foot deformity*
- *Osteoma cutis*
- *Non-healing ulcer*

Abstract: Perforating osteoma cutis is a rare condition. Here we report a case of a woman with multiple perforating osteoma cutis skin lesions. Only four cases of perforating osteoma cutis have been reported to date; one female, located on the breast (Kim and Ahn, 2015) and three males, two of them located on the forehead (Kluger, 2016; Cohen, 2018) and one on the pretibial region, within a tattoo (Basu et al, 2019).

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Osteoma cutis is a rare, benign disorder of bone formation in the dermal or subcutaneous layer of the skin that presents as a stony hard nodule (Hong and Kang, 2003). Although the mechanism of bone formation remains unclear, it is speculated that resident fibroblasts or nests of pluripotent mesenchymal cells differentiate into osteoblasts to form bone tissues (Duarte, 2010). Osteoma cutis can be classified into primary and secondary, the primary type is where there is no preexisting lesion or predisposing factors. Whereas, osteoma cutis is classified as a secondary or acquired when it is associated with inflammatory processes, scars, dysembryoplasia, or malignancy. Some authors have demonstrated a correlation between osteoma cutis and GNAS1 gene mutations, which act as a regulatory gene in progressive osseous heteroplasia and Albright hereditary osteodystrophy (Pignolo et al, 2015).

Most of the reported literature has documented osteoma cutis to be found in patients between the age of 40–60 years old. Lesions are typically found on the body at areas such as the head, face, scalp, back, chest and occasionally fingers. Osteoma cutis is commonly found on the face for females and scalp for males.

Case report

A 73-years-old lady was referred by the Dermatology clinic for painful lesions, present on her left leg for four months, she also had hypertension and dyslipidaemia. The patient had previously had a similar wound at the same site, but claimed it was healed spontaneously on that occasion.

Physical examination revealed two ulcers on the left leg. The first ulcer was located at the anterior compartment of the shin, 5.0cm (length) x 3.5cm (width) x 0.3cm (depth) *(Figure 1a)*; the second one was located at the lateral aspect of the leg. The wound bed for the first ulcer had slough, and granulation tissue was seen with notable calcification over the peripheries of the wound bed. The surrounding skin was erythematous, warm to touch, shiny and had circumferential post-inflammatory hyperpigmentation (PIH) surrounding the wound. The second wound over the lateral aspect of the shin, was around 5cm (length) x 3.5cm (width) x 0.5cm (depth) *(Figure 1b)*. The base was noted to be sloughy with exudative discharge. It also exhibited calcium deposit mostly around the peripheries, with macerated edges. There was no significant lymphadenopathy and systemic examination was otherwise normal.

Routine investigation and biochemical parameters showed slight elevation of uric acid (454µmol/l), low 25-hydroxy-Vitamin D level: 41.53 mmol/l (76–250nmol/l); otherwise serum calcium, phosphate, alkaline phosphatase, thyroid function test, tumor markers eg AFP, CEA, Ca 19.9, CA 125, iPTH were all within the normal range. An x-ray of the left leg revealed multiple infiltration of bone deposits surrounding the circumferential aspect of the leg *(Figure 2 a–b)*. Similar deposits were found over the right leg, but to a lesser extent *(Figure 2 c–d)*.

The edge and base of both ulcers were excised and sent for biopsy. Microscopic examination showed ulcerated surface epithelium replaced by fibrino-purulent exudate. The remaining epidermis showed acanthosis and spongiosis. The epidermis takes on a sponge like look due to the shrinkage

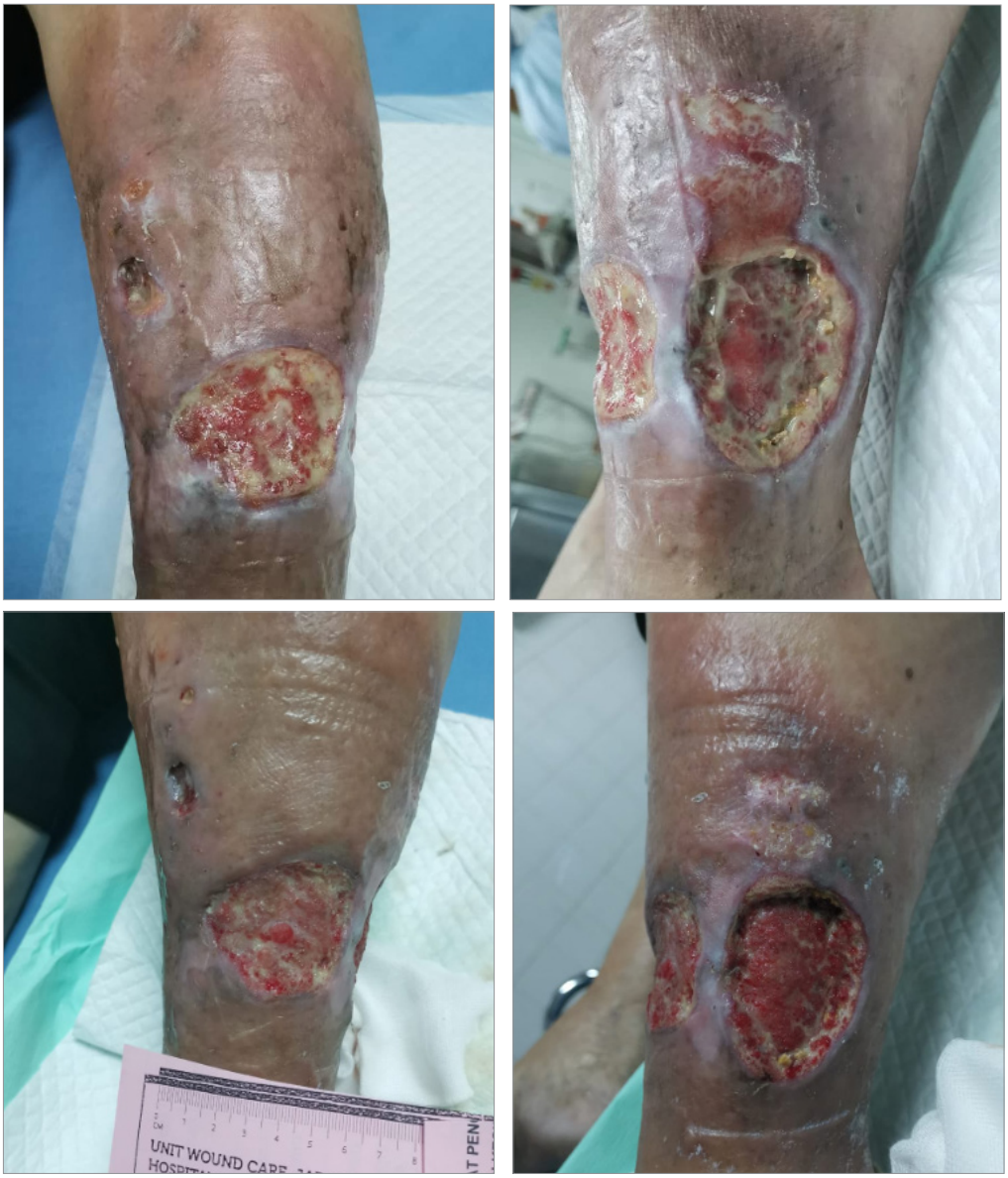

Figure 1. The wounds, both anterior (a) and lateral (b) view, upon first review in our clinic. Shown below are after 4 weeks of treatment (c–d). The wound base still had minimal slough and the surrounding skin appeared less eryhtematous compared with the first visit.

and rounding of keratinocytes (condensation) and the widening of intercellular gaps, which can lead to the formation of tiny intraepidermal vesicles. The dermis is infiltrated by lymphocytes, plasma cells, and neutrophils. There was dermal fibrosis, ossification and calcium deposition. No evidence of malignancy observed. Histopathological features were consistent with an benign ulcer with primary osteoma cutis.

In view of the wound was infected initially, a few courses of antibiotics was started to eradicate the infection. Furthermore, Nano copper solution was used to soak the wound for 5 minutes and after bone deposits were removed as much as possible. An adequate amount of Nanogen was applied on the wound and RTD foam was used as a secondary dressing. Finally Calmoseptine was used as a barrier cream to prevent wound maceration. Additionally, this case was referred to our endocrine team for further investigation and for vitamin D supplement. In this case, surgical approach was not recommended as the area of involvement was large.

There was improvement in the wound bed in this hard to heal wound, the wound began to heal and good granulation was noted after 4 weeks. The wound was treated and details recorded for four weeks, after which the patient

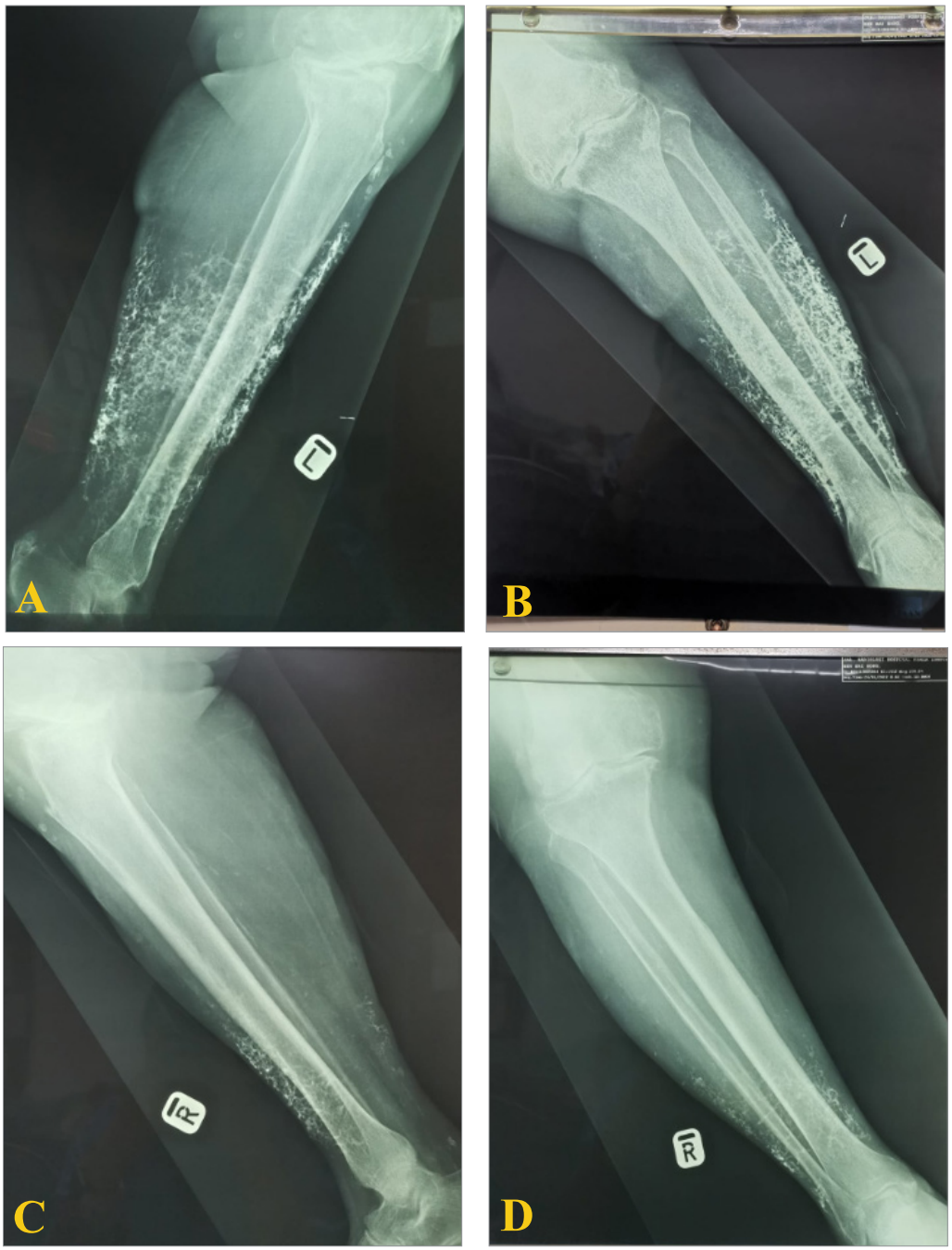

Figure 2. Picture on the left showed X-ray of lateral (A) and anterior (B) view of left leg of the patient respectively, while the X-ray of lateral (C) and anterior (D) view of right leg of the patient

was followed up by the endocrine unit who were to investigate the causes of the osteoma cutis for further management.

Discussion

The patient had presented to us after multiple visits to general practitioners and surgeons and had initially been mistaken as ulcerated gout because of hyperuricaemia. A diagnosis of gout is typically made using the American College of Rheumatology criteria; the primary criteria are joint swelling/pain/tenderness, monosodium irate crystals in the synovial fluid, and the presence of tophi (Hainer et al, 2014; Neogi, et al, 2015). Although it is uncommon for gout to be ulcerated, it might occur in patients with poorly controlled hyperuricaemia, especially those who have limited access to healthcare or are non-adherent to medical advice (Patel et al, 2010). Ulcerated gout has a higher prevalence in the elderly and in males. There was no other evidence to support gout in this case apart from hyperuricaemia.

Another diagnosis taken into consideration was a chronic venous leg ulcer as the patient

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had noticeable varicose vein and skin changes suggestive of possible stasis eczema. However, because her ulcers were painful and the atypical location of the ulcer (lateral gutter), this diagnosis was excluded. Because of patient's long-standing hypertension, age, location, and characteristics of the ulcers, another differential diagnosis was Martorell hypertensive ischaemic leg ulcer (HYTILU also known as Martorell ulcer). However, the biopsy didn't show any stenosis of arteriole lumen or thickened arterioles walls, arteriolar thrombosis, or skin infarction.

Osteoma cutis is a benign condition defined as the eruption of an osseous structure in the skin and is distinguishable from calcinosis cutis. The former arises from membranous ossification without cartilage as a precursor; also can be called cutaneous osteoma. The latter is described as the deposition of calcium salt in the absence of osteoid, which constitutes osteoblasts, osteoclasts, and hydroxyapatite as essential components (Jatana et al, 2012). Calcinosis cutis was one of the differential diagnosis that was considered in this case as there were multiple subcutaneous nodules on the right anterior shin of the patient. Similar nodules were initially seen on the left leg of the patient before it turned into an ulcer. The skin was initially hypopigmented, hardened and had some irregularity upon palpation. Subsequently, after the skin broke an ulcer formed. However, the biopsy showed that the nodules consists of bony component instead of calcium deposits hence the diagnosis of Calcinosis cutis was eliminated.

The treatment of osteoma cutis can be divided into invasive and non-invasive modalities. The author has observed cases of non-invasive osteoma cutis, located on the face, which were successfully treated with application of tretinoin cream, but had limited results essentially in small and superficial lesions (Cohen et al, 2001).

Invasive treatment options for single or multiple lesions of osteoma cutis include complete excision along with less traumatic techniques, such as curettage, which had excellent cosmetic results, or may use ablative laser procedures e.g., CO_2 and erbium. Yttrium aluminium garnet (YAG) lasers may induce cutaneous pigmentary changes. Successfully non-ablative YAG laser treatments for military osteoma, has been performed, which did not induce pigmentary changes (Barolet et al, 2020).

In a Case secondary osteoma cutis, the associated metabolic abnormalities should be investigated and treated accordingly.

Conclusion

In conclusion, our patient presented with recurrent wounds on bilateral lower limbs which were mistaken for other common pathologies, such as diabetic foot ulcer or a venous leg ulcer. A detailed history taking and physical examinations with appropriate blood and radiological investigations resulted in a correct diagnosis. In our experience, Osteoma cutis with an ulcerated wound is not easy to treat hence secondary causes (if any) should be identified and treated accordingly. WAS

References

Barolet AC, Litvinov IV, Barolet D (2020) Multiple miliary osteoma cutis treatment response to Q-switched Nd:YAG laser: A case report. *SAGE Open Med Case Rep* 8:2050313X20910562. https://doi.org/10.1177/2050313x20910562

Basu P, Erickson C P, Calame A Cohen PR (2019) Osteoma cutis: an adverse event following tattoo placement. *Cureus* 11(3):e4323. https://doi.org/10.7759%2Fcureus.4323

Cohen PR (2018) Perforating osteoma cutis: case report and literature review of patients with a solitary perforating osteoma cutis lesion. *Dermatol Online J* 24(3):13030/qt6kt5n92w.

Cohen AD, Chetov T, Cagnano E, (2001)Treatment of multiple miliary osteoma cutis of the face with local application of tretinoin (all-trans-retinoic acid): a case report and review of the literature. *J Dermatolog Treat* 12(3):171–3. https://doi.org/10.1080/09546630152607925

Duarte IGL(2010) Multiple injuries of osteoma skin in the face: therapeutical least invasive in patients with acne sequelacase report. *An Bras Dermatol* 85(5):695–8. https://doi.org/10.1590/s0365-05962010000500016

Hainer BL, Matheson E, Wilkes RT (2014) Diagnosis, treatment, and prevention of gout. *Am Fam Physician* 90(12):831–6

Hong SH, Kang HY (2003) A case of perforating osteoma cutis. Ann Dermatol 2003;15(4):153. http://dx.doi.org/10.5021/ad.2003.15.4.153

Kim BK, Ahn SK (2015) Acquired perforating osteoma cutis. *Ann Dermatol* 27(4):452–3. https://doi.org/10.5021%2Fad.2015.27.4.452

Kluger N (2016) Cutaneous and systemic complications associated with tattooing. *Presse Med* 45(6 Pt 1):567–576. https://doi.org/10.1016/j.lpm.2016.02.016

Jatana SK, Negi V, Das SA (2012) A case of idiopathic calcinosis cutis. *Med J Armed Forces India* 68(4):383–5. https://doi.org/10.1016%2Fj.mjafi.2012.07.006

Neogi T, Jansen TL, Dalbeth N et al (2015) Gout classification criteria: an American College of Rheumatology/European League Against Rheumatism collaborative initiative. *Arthritis Rheumatol* 67(10):2557-68. https://doi.org/10.1002/art.39254

Patel GK, Davies WL, Price PP, Harding KG (2010)Ulcerated tophaceous gout. *Int Wound J* 7(5):423–7. https://doi.org/10.1111/j.1742-481X.2010.00700.x

Pignolo RJ, Ramaswamy G, Fong JT et al (2015) Progressive osseous heteroplasia: diagnosis, treatment, and prognosis. *Appl Clin Genet* 8:37–48. https://doi.org/10.2147%2FTACG.S51064