Primary Osteoma cutis : A Rare Case with Unusual Presentation

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Abstract
Osteoma cutis (OC) is a term applied to localized bone formation in dermis or subcutaneous tissue. This lesion is exceedingly rare with no well defined data on an incidence. Patients develop multiple cutaneous nodules histologically composed of mature bony trabeculae with enclosed fatty marrow and seldom haemopoietic tissue. We present a case of OC in a 30 year old male with multiple cutaneous nodules on trunk with no other systemic illness nor any evident family history.

Introduction
Synonyms include miliary osteoma, (osteodermia, osteosis and cutaneous ossification).1-6 OC was 1st reported by Hopkin’s in 1928. OC differs from metaplastic reaction (wherein enchondral ossification occurs) by presence of mesenchymal (membranous) ossification without cartilage precursors or models.1-3 It occurs de-novo or in association with Albright’s hereditary osteodystrophy (AHO) as a primary form, or as secondary form following various benign and malignant condition, commonest being long standing acne formation, trauma and infection. Pseudohypoparathyroidism, naevi, pilomatrioma and basal cell carcinoma are less common culprits.1-3,6 Recently, a new disorder Progressive osseous heteroplasia has been described in orthopaedic literature. Primary cutaneous ossification beginning in infant age and presenting as secondary form could be presenting sign of this progressive disabling disease.3

Case Summary
A 30 year old male was referred to skin OPD for multiple, small, painless cutaneous nodules around nipple, waxing and waning but now, progressing in size, since past two years. Biopsy of nodule was performed.
We received a tiny skin covered firm tissue bit measuring 0.8x0.5cm. Histological examination showed skin with unremarkable epidermis. The dermis underneath showed a well circumscribed, circular bony tissue with entrapped mature adipose tissue. No haemopoietic elements were seen. We rendered a preliminary diagnosis of OC after seeking detailed family history. Investigations pertaining to serum calcium levels, 5hydroxyphenol and PTH were done to rule out pseudohypo/hypoparathyroidism. All parameters were within normal limits. Hence we favoured a diagnosis of primary OC.

Discussion

Pathogenesis of OC is uncertain. Burgdoff and Nasemann\(^4\) theorized two possible processes -1. embryologic process whereby, primitive mesenchymal cells differentiate to osteoblasts 2. more plausible, is osteoblastic metaplasia of mesenchymal elements like fibroblasts. Second theory is more acceptable and has been proposed by many.\(^4\)\(^-\)\(^6\)

OC can occur at any age\(^3\) with a female predilection, male to female ratio being 2:1. Clinically it can occur in various forms. 1) Localised OC as congenital form associated with Albright’s hereditary osteodystrophy (AHO), Gardner’s syndrome. 2) Plaque-like lesions occur congenitally usually on scalp 3) Single lesions unassociated with any systemic illness may occur later in life. 4) multiple OC unassociated with AHO usually occur in young females over face with a history of long standing acne.\(^1\)-\(^6\) Presenting case was unusual as it occurred in 4th form but in a male and on trunk with no evidence of AHO.

Histologically, foci of ossification occur in dermis/subcutaneous tissue, sometimes around extravasated keratin debris, hair shaft or in other tumours like BCC. Enlargement by continuous bony application at the surface with eventual central remodelling gives circular outline with central hollowing which creates a space for fatty marrow.\(^1\)\(^-\)\(^6\) Our case showed similar histologic findings. Some cases show transepidermal elimination i.e epitheliotropism of channels with bony streaks / fragments in breaks of epidermis. Therefore, Osteomyelitis with sinus tract formation is a close D/D. OC must be differentiated from tumoral calcinosis wherein calcium salts are deposited whereas ossification occurs with no calcification in OC.\(^1\)\(^,\)\(^2\)\(^,\)\(^6\)

Surgical excision is most common modality of treatment. In multiple lesions due to cosmetic reasons, laser techniques as well as non-operative modalities including local application of trenitoin or oral intake of etidronate disodium to promote transepidermal elimination have been used with unproven efficacy.\(^1\)-\(^6\)

OC is a benign lesion, however Osteogenic sarcoma has been reported.\(^5\) Squamous cell carcinoma in situ was associated with OC but stated that it could be a collision tumour.\(^5\)

References

5. Poonam Sherma. Squamous cell carcinoma in-situ associated with OC. Internet Jour Dermatology ISSN 2006; 4 (2) : 1531.