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PRIMARY MILIARY OSTEOMA CUTIS – A CASE STUDY WITH REVIEW OF LITERATURE.

Pathology

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ABSTRACT

Miliary osteoma cutis is a rare entity, is characterized by the deposition of multiple fragments of mature bone in the dermis. They cause diagnostic, therapeutic and cosmetic concern especially in women who are usually concerned. There are two types of Osteoma cutis ,Primary and Secondary. Primary Osteoma Cutis is characterized by denovo bone formation in the skin. Secondary Osteoma Cutis is more common, seen in 85% of cases, as a sequel of various disorders. We report a case of primary miliary osteoma cutis of the face and also discuss the possible pathogenesis and histopathological findings.

KEYWORDS

Face, Osteoma cutis, Surgical treatment.

CASE REPORT:

A 45 year old female had long standing lesions on face for which she consulted a dermatologist for aesthetic improvement of the lesions. On examination several normochromic papules were grouped in malar regions similar to milia. The lesions have been increasing in size progressively over a period of 3 yrs measuring 0.5cm to 1cm,which were hard on palpation. Skin biopsy was done from the lesion and was send to our lab for histopathological examination. On routine Haematoxylin and eosin staining showed islands of well formed calcified lamellar bone tissue in the dermis with presence of few osteocytes. Epidermis was unremarkable and no underlying pathology was seen. Lab investigations for serum calcium, phosphorus and electrolytes along with LFT and RFT were within normal limits. There was no history of trauma , drug intake and any previous cutaneous lesions .So a final diagnosis of Primary Osteoma Cutis was confirmed.

DISCUSSION:

Miliary Osteoma cutis was first described in 1864 by Virchow[1]. Osteoma Cutis is characterized by formation of morphologically normal bone within the dermis or in the subcutaneous tissue. These lesions are asymptomatic and usually appear as skin coloured papules in the scalp, the face as well as the trunk, the breast, the extremities and the buttocks. [2]

There are two types of Osteoma Cutis Primary and Secondary. Primary Osteoma Cutis is less common and seen in 15% of patients and secondary Osteoma Cutis is commoner and seen in 85% [3]. Primary Osteoma Cutis is characterized by denovo bone formation in the skin without a known associated or pre-existing cutaneous disorder . Primary Osteoma Cutis has four different clinical variants – solitary, widespread, plaque like and multiple miliary osteomas of the face [4]. Four main syndromes associated with Primary Osteoma Cutis are Albrights hereditary Osteodystrophy, Fibrodysplasia of Progressive Ossification, Osseous Progressive Heteroplasia and Plate like Osteoma Cutis[5]. Secondary Osteoma Cutis on the other hand occurs as a sequel to multiple disorders including nevi, scleroderma, pilomatricoma, dermatomyositis, basal cells carcinoma, scars, cutaneous inflammation, trauma, calcification, fibrous proliferation and venous stasis, syringoma ,epidermoid cyst [3].

The pathogenesis of this disease still remains obscure however it is essential to know in order to prevent its occurrence. Various theories have been postulated to explain its pathogenesis. Montgomery regarded the osteomas as hamartomas or nevoid tumours[6] Burgdorf and Nasemann reported two possible processes. One theory assumes a disordered embryologic process, whereby primitive mesenchymal cells differentiate normally into osteoblasts but migrate to the wrong location. The second theory, which appears more compatible with our histological findings, interprets the presence of bone as a result of osteoblastic metaplasia of mesenchymal cells, such as fibroblasts[7]. Levell reported that a long standing inflammation may cause mesenchymal cells differentiation into osteoblasts [8].

Hopkins, in 1928, was the first to suggest the role of acne in the development of multiple miliary osteoma cutis [9]. Some authors reported extensive endometriosis, metrorrhagia and osteoarthritis affections in association with this disease[10]. Most cases have been reported in women with no evidence of the implication of the hormonal status. In some cases transepidermal elimination of fragments of bone within the channels lined by epidermis and leading to the surface is seen.

In the Genetic level, there is heterozygous inactivating germ line mutation of the GNAS gene causing abnormal expression or function of the α-subunit of the stimulatory G-protein adenyl cyclase. This results in reduced inhibitory control of cellular induction to osteoblast differentiation in ectopic sites(11) resulting in heterotrophic ossification originating in fat cells(12). Progressive osseous heteroplasia is characterized by dermal ossification of cutaneous, subcutaneous and deep connective tissues(12).

There are several forms of treatment which are reported in literatures which includes the use of retinoic acid 0.1% to eliminate transdermal elimination of the bone formation to use of dermabrasion (Fulton), needle microincision extirpation method (Baskan et al), Yag laser to ablate the epidermis (Kaufmenn).Best treatment option is with surgical techniques such as curettage and Needle excision[13].

CONCLUSION:

The classification of miliary osteoma cutis is controversial as although it is described traditionally as primary type it is associated with previous history of acne in 50% cases [9] thus placing it under both categories. In our case there was no previous history of acne or any other inducing factors. So more case reports are needed to acess the pathogenesis, its therapeutic management and the inducing factors of this rare entity. Diagnosis of Primary Osteoma Cutis in our case was

based solely on its clinical features ,lab evaluation and distinct histopathological findings.

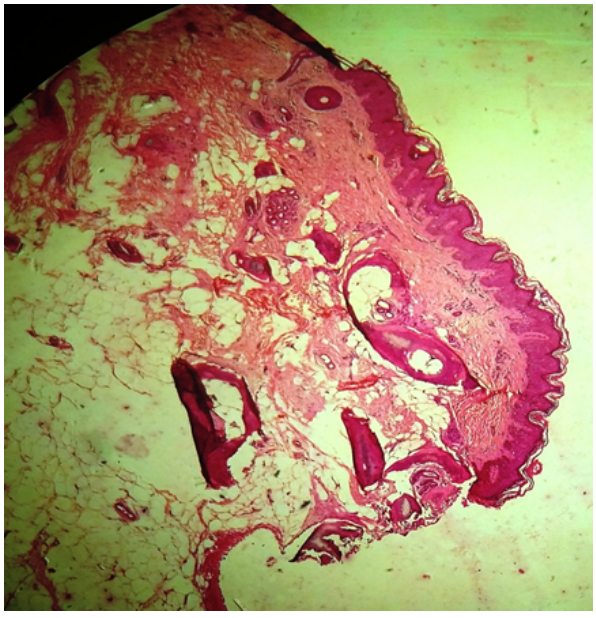

Fig 1-Scanner view 40X- Showing islands of lamellar bone tissue in the dermis.

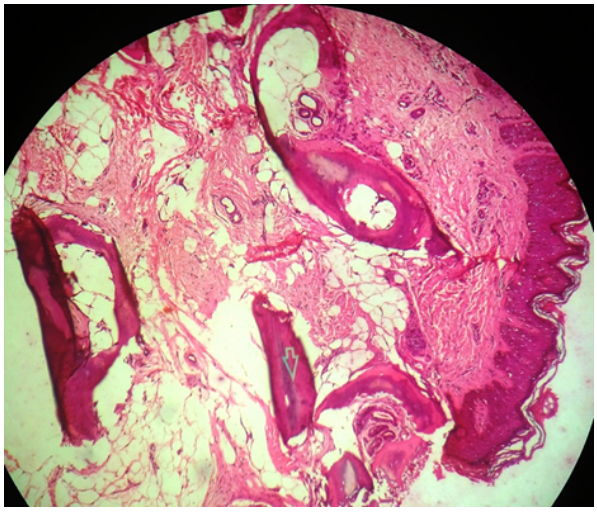

Fig 1(b)LP 100X-Low power magnification.

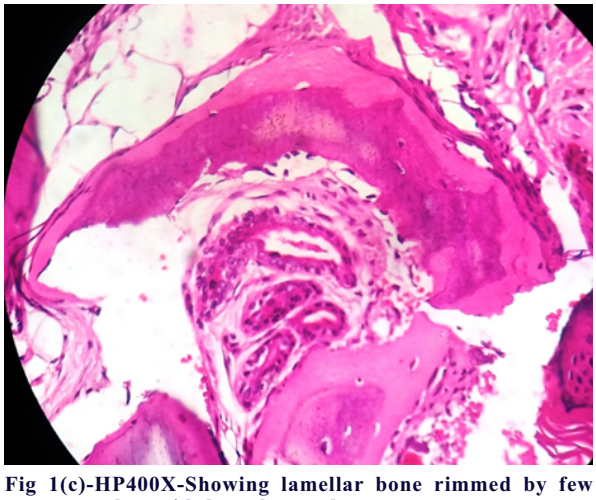

Fig 1(c)-HP400X-Showing lamellar bone rimmed by few osteocytes along with dermal appendages.

REFERENCES:

[1] R. M. Myllyl¨a, K. M. Haapasaari, R. Palatsi et al., "Multiple mil iary osteoma cutis is a distinct disease entity: four case reports andreviewoftheliterature," BritishJournalof Dermatology, vol. 164, no. 3, pp. 544–552, 2011.

2. Boschert MT, Puckett CL. Osteoma cutis of the hand. Plast Reconstr Surg 2000; 105:1017-8.
3. NA Montecionos Ayariri, FX Nahas, MV Jardini Barbosa, AB Farah, J de Arimatéia Mendes, LM Ferreira. Isolated primary osteoma cutis of the head: Case report. Can J Plast Surg 2006;14(1): 33-36.
4. Sethuraman G, Malhotra AK, Khaitan BK, Kumar R, Sharma VK, Kabra M, et al. Osteoma cutis in pseudohypoparathyroidism. Clin Exp Dermatol 2006;31:225-7.
5. Bowman PH, Lesher JL Jr. Primary multiple military osteoma cutis and exogenous ochronosis Cutis 2001; 68:103.
6. Montgomery H. Dermatopathology, p 1131. New York, Paul B. Hoeber, 1967.
7. Burgdorf W, Nasemann T. Cutaneous osteomas: a clinical and histopathologic review. Arch Dermatol Res.1977; 260: 121-135. PMID: 413495.
8. Levell NJ, Lawrence CM. Multiple papules on the face. Mul- tiple miliary osteoma cutis. Arch Dermatol. 1994; 130: 373- 374. PMID: 8129420.
9. A. M. Thielen, L. Stucki, R. P. Braun et al., "Multiple cutaneous osteomas of the face associated with chronic inflammatory acne," Journal of the European Academy of Dermatology and Venereology, vol. 20, no. 3, pp. 321–326, 2006.
10. Ward M, Viraben R. A case for diagnosis: miliary osteoma of the face. Ann Dermatol Venereol. 1993; 120: 713-714. PMID: 8161106.
11. Acquired platelike osteo ma cutis Neelam Vashi MD, Julie Chu MD, Rishi Patel MD, Dermatology online Journal 17 (10):1, 2011
12. Unilateral progressive osseous heteroplasia Eur J Dermatol 2009; 19 (3): 214-5.
13. J.F.Altman,K.S.Nehal,K.J.Busam,andA.C.Halpern,"Treatment of primary miliary osteoma cutis with incision, curettage, and primary closure," Journal of the American Academy of Dermatology, vol. 44, no. 1, pp. 96–99, 2001.