

Case Report

Multiple cutaneous miliary osteomas of the face: a case report

Osteomas cutâneos miliares múltiplos da face – relato de caso

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ABSTRACT

Summary: Multiple cutaneous miliary osteoma is a rare condition characterized by ectopic bone tissue in the dermis and/or hypodermis. It usually occurs on the face, in individuals of 17 to 79 years old. Its etiology is still unknown. The present article reports the case of a female patient affected by a hardened, poorly delimited, papular plaque of dark brown hue and irregular surface, affecting the malar and mentonian regions. The patient sought medical care two years before the publication of this study, describing untreated acne since the age of 16. The papular plaque was composed of hyperchromic papular nodular lesions – some with white-yellowish hue. The diagnosis was confirmed histopathologically. Satisfactory therapeutic results were achieved with the use of 0.1% tretinoin cream.

Keywords: osteoma; decalcification, pathologic; ossification, heterotopic.

RESUMO

Introdução: Osteoma cutâneo miliar múltiplo é doença rara, caracterizada por tecido ósseo ectópico na derme e/ou hipoderme. Usualmente ocorre na face, em pessoas entre 17 e 79 anos. A etiologia é ainda desconhecida. Relata-se caso de paciente do sexo feminino, apresentando placa papulosa de coloração castanho-escura, endurecida, acometendo regiões malaras e mentoniana, mal delimitada, com superfície irregular, formada por lesões papulonodulares, hiperocrômicas, algumas branco-amareladas, há dois anos, referindo acne, desde os 16 anos de idade, não tratada, tendo o diagnóstico sido confirmado histopatologicamente e apresentado resultado terapêutico satisfatório com uso de tretinoína creme 0,1%.

Palavras-chave: osteoma; descalcificação patológica; ossificação heterotópica.

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INTRODUCTION

Multiple cutaneous miliary osteomas (MCMO) is a rare disorder characterized by the presence of ectopic bone tissue in the dermis and/or in the hypodermis.¹ It usually occurs on the face, affecting people between 17 and 79 years old (mean = 47 years old). Its etiology is still unknown.² Cutaneous ossification can be classified as primary when bone tissue develops in the skin and is observed early after birth or during childhood, with the absence of preexisting skin lesions or calcium and phosphorus metabolic abnormalities. This condition may possibly be associated with Albright's hereditary osteodystrophy, fibrodysplasia ossificans progressiva, progressive osseous heteroplasia and Gardner's syndrome. It may also arise in isolation, with no association to other diseases, manifesting as a single small osteoma, a single large plate-like osteoma, multiple disseminated osteomas, or multiple miliary osteomas on the face.^{1,4-6} Secondary cutaneous osteomas are more common, accounting for 70-85% of cases and occurring when bone tissue develops in preexisting skin lesions. Secondary cutaneous osteomas can be of neoplastic origin (such as melanocytic nevi, pilomatricoma and basal cell carcinoma) or inflammatory origin (such as scleroderma, systemic erythematosus lupus and dermatomyositis). It can also appear on post-operative scarring, or following folliculitis, chronic venous insufficiency, calcinosis, or trauma.^{1,5} Multiple idiopathic osteomas are most commonly reported on the faces of women with no history of acne. Nonetheless, the precise correlation between cutaneous osteoma and acne remains unclear.⁷

CASE REPORT

A 47-year-old female patient came to a medical appointment complaining of facial darkening (which had been taking place for about 24 months), and a history of untreated acne

(since the age of 16).

Dermatological examination showed a hardened, poorly-defined papular plaque of dark-brown hue and an irregular surface affecting the malar and mentonian regions. The papular plaque was formed by papulonodular hyperchromic lesions, some of them of yellowish-white hue (Figure 1). Following an incisional biopsy, histological examination revealed fragments of mineralized tissue with an osteoid characteristic located within the dermis, in addition to the presence of an epidermal cyst (Figure 2). Histopathologic diagnosis suggested an epidermal cyst associated with cutaneous osteoma. Serum levels of calcium/phosphate, alkaline phosphatase and parathyroid hormone proved unaltered. Following the initial consultation, the patient was instructed to apply 0.1% tretinoin cream. Four months later, there was significant improvement in hyperpigmentation (Figure 3).

DISCUSSION

The term osteoma cutis was originally described by Wilkins in 1858, corresponding to the presence of mature bone tissue, in the dermis and/or hypodermis. The condition is a rare, benign dermatosis, classified as primary when arising early in life in healthy skin, and secondary when associated with preexisting neoplastic or inflammatory skin lesions (representing 70-85% of cases).^{1,6} Secondary ossifications of the face occur almost exclusively in women with a history of long-lasting inflammatory acne, as in the present case. However, the precise correlation between cutaneous osteoma and acne remains unclear.⁷ MCMO arise in locations that coincide with those of the onset of acne vulgaris lesions (most often on the face) and when there is a history of this condition (55% of cases). It has been suggested that osteomas are secondary to dystrophic alterations in acne



Figure 1: **A** - Confluent hyperchromic lesions presenting small yellowish papules, involving the mandibular, malar and mentonian regions; **B** - Presence of hyperchromic plaque with irregular surface in the left hemiface, with brownish nodule and small whitish dots

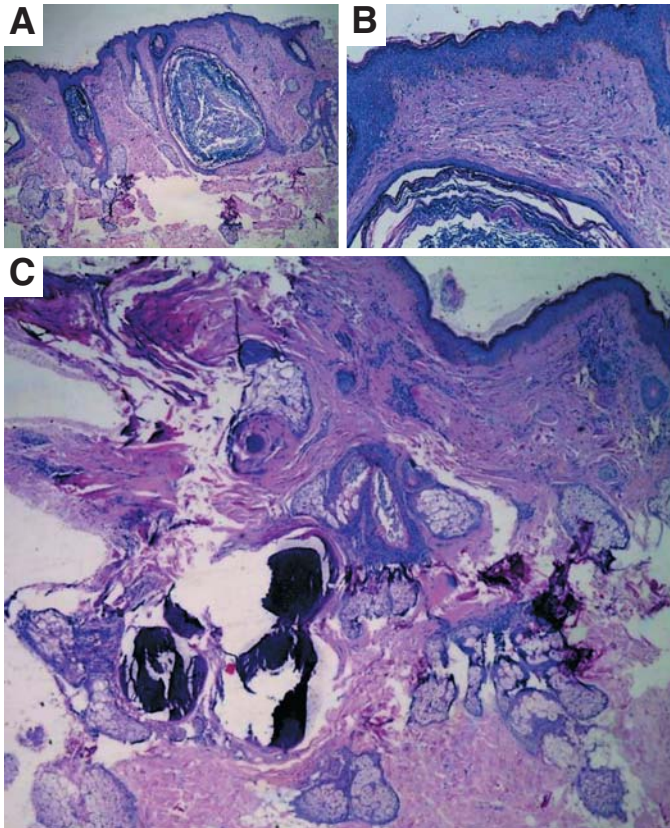


Figure 2: A - Cutaneous fragment indicating the presence of epidermal cyst located in the dermis (HE 40x); B. Greater magnification showing the cystic cavity filled by keratin blades (HE, 100x); C. Mineralized tissue of osteoid appearance located in dermis, close to the sebaceous glands (HE, 40x)

scars. The role of estrogen has been discussed, however it does not seem to be crucial in the formation of osteoma, since postmenopausal women, and some men also develop such lesions. 2 Several MCMO cases have been reported in patients without a history of acne or other inflammatory conditions, such as that of a 75-year-old female patient with an incidental histological finding of exogenous ochronosis, resulting from the use of hydroquinone.^{3,7}

In the present case, the patient sought medical care due to hyperchromia in the affected areas. No confirmation was available as to whether she had previously used any medication to treat acne, or used skin lighteners containing hydroquinone.

The main histological features are multiple bone spicules of varying shapes and sizes in the dermis, extending as far as the subcutaneous tissue. Such bone spicules contain numerous osteocytes, as well as cement lines. Havers ducts containing blood vessels and connective tissue can be observed in some areas. Osteoblasts with elongated nuclei can be seen along the margin of some osseous spicules. Some osteoclasts with multiple hyperchromatic and elongated nuclei can be found in the so-called Howship lacunae, while some osseous spicules also contain mature adipocytes aggregates.^{1,8}

The pathogenesis of cutaneous ossification remains uncertain. It has been suggested that chronic inflammation leads to the development of small calcifications and metaplastic ossification. Stimulation of mesenchymal cells by different factors may induce their differentiation into osteoblastic cells, resulting in bone formation. While it can occur in isolation, the presence of ossification should be seen as a possible indication of the presence of associated diseases, including fibrodysplasia ossificans progressiva (McCune-Albright syndrome), hereditary osteodystrophy, progressive osseous heteroplasia and Gardner's syndrome.



Figure 3:
A, B, C - Initial picture
D, E, F - Whitening and regression after treatment with 0.1% tretinoin

In this case report there was no evidence of clinical, metabolic characteristics or endocrine abnormalities corresponding to those conditions, with only a history of acne being confirmed.

Facial MCMO treatment methods are limited. The surgical technique involving incision and curettage of bone fragment, followed by suture, has been shown to be less invasive, and provides excellent results. The technique of extirpation of bone fragments after needle microincisions, followed by primary closure, is straightforward, effective, and cost effective.⁶ Dermabrasion or CO₂ laser or Erbium:YAG lasers are mentioned

as epidermis ablation processes.⁹ The topical application of tretinoin may in some cases have a favorable effect by promoting the transepidermal elimination of osteomas.⁶ The patient studied responded satisfactorily to use of 0.1% tretinoin, in daily and nightly applications, which proved to be an efficient, safe and non-invasive method for treating MCMO. The potential development of osteoma cutis should be considered in all patients with chronic inflammatory acne, because the identification of the first condition plays an important role in the treatment of the MCMO. ●

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