Miliary Osteoma of the Face: A Report of 4 Cases and Review of the Literature

Fabiane N. Bergonse, MD; Marcello Menta S. Nico, MD; Maria Ines Kavamura, MD; Miriam N. Sotto, MD

Osteoma cutis (OC) is a rare disorder characterized by compact bone formation in the dermis and subcutaneous tissue. It is classified in primary and secondary forms according to the presence or absence of previous cutaneous lesions. Miliary osteoma of the face (MOF) is a form of primary OC that generally occurs in middle-aged and older adult women. We report 3 cases of typical MOF and one additional case in a black patient, which to our knowledge has not been described previously.

steoma cutis (OC) is a benign disease first described by Wilckens in 1858.^{1,4} It is characterized by the presence of well-formed calcified bone in the dermis.⁵ Primary OC occurs in the absence of a previous lesion, leading to bone formation. Secondary OC occurs in some benign tumors (pilomatricomas, chondroid syringomas, basal cell carcinomas, pilar cysts, nevi, sebaceous adenomas).^{1,6-9} Miliary osteoma of the face (MOF) refers to a condition that occurs in middle-aged and older adult women. It presents as multiple asymptomatic papules on the cheeks. Many patients with MOF remember having had acne during their adolescence. We present 3 cases of MOF in light-skinned women and one case in a black woman.

Case Reports

Patient 1—A 61-year-old white Brazilian woman had a 3-year history of lesions on her cheeks. Results of a clinical examination revealed several (not exceeding 20) skin-colored, 1- to 3-mm papules scattered bilaterally on the cheeks that were hard on palpation. The woman had a history of mild acne as a teenager. At presentation, her only medical condition was well-controlled hypertension. Laboratory results (eg, complete blood count [CBC], glucose, cholesterol, sodium, potassium, calcium, phosphate,

Drs. Bergonse, Nico, and Sotto are from the Hospital das Clinicas, São Paulo, Brazil. Dr. Kavamura is in private practice, São Paulo. Reprints: Marcello Menta S. Nico, MD, Rua Itapeva 500, 3° andar, Bela Vista cep: 01332-000, São Paulo-SP, Brazil.

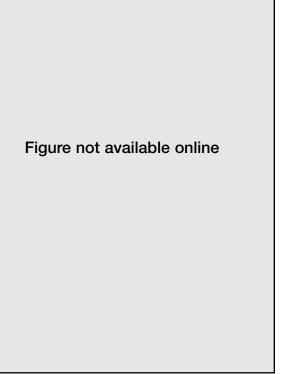
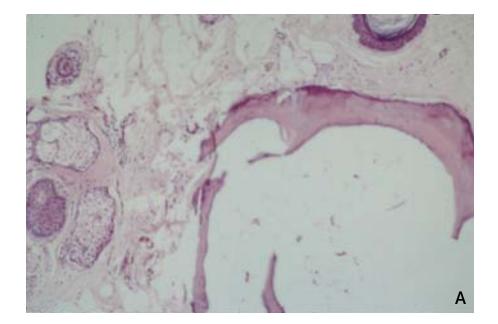


Figure 1. Multiple lesions of osteoma cutis.

blood urea nitrogen [BUN], creatinine) were within normal limits. Histopathology of a papule without decalcification showed sheets of compact bone in the dermis.

Patient 2—A 70-year-old white woman of Lebanese origin had lesions on her cheeks of unknown duration. On examination, there were innumerable small papules that were coalescing bilaterally on her cheeks, forming a stone-hard plaque with a pebbly surface (Figure 1). She denied having had any medical conditions, including acne. Laboratory results (eg, CBC, glucose, sodium, potas-



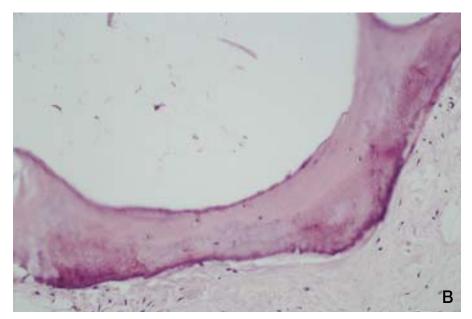


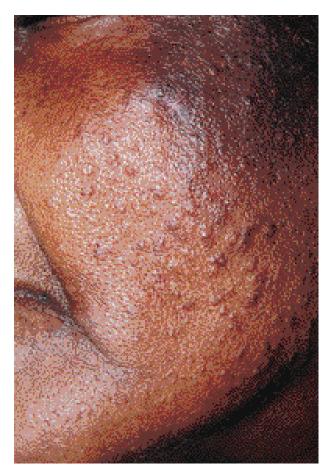
Figure 2. Histopathologic features of osteoma cutis. Sheets of compact osseous tissue in the dermis (A). Higher magnification shows osteocytes within the compact bone (B)(H&E, original magnifications ×100 and ×200).

sium, calcium, phosphate, BUN, creatinine) were within normal limits. Histopathology after decalcification showed sheets of compact bone in the dermis.

Patient 3—A 52-year-old white Brazilian woman had a 6-year history of facial lesions. Clinical examination results showed multiple (more than 40) pinhead-sized, skin-colored, firm papules on her cheeks. She had a history of severe scarring acne during puberty. She was otherwise in good health. Laboratory results (eg, CBC, glucose, cholesterol, sodium, potassium, calcium, phosphate, BUN, creatinine) were within normal limits. Histopathology after decalcification showed

sheets of compact bone with some osteocytes within them (Figure 2).

Patient 4—A 63-year-old black Brazilian woman with a history of acne vulgaris in her adolescence presented with multiple skin-colored tumoral papules on her cheeks that were hard on palpation (Figure 3). No other medical condition was detected. Laboratory results were within normal limits (eg, CBC, glucose, calcium, phosphate, BUN, creatinine). Facial x-ray showed small round opacities, mostly seen in the projection of her right mandibular angle (Figure 4). Histopathology of a papule showed sheets of compact bone in the dermis.





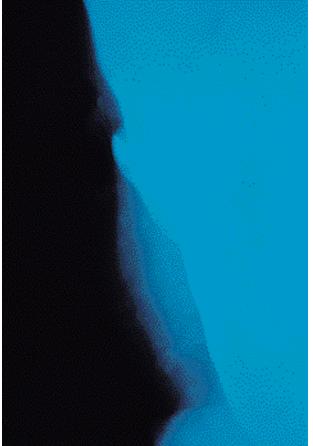


Figure 4. Small round opacities near the mandibular angle.

Comment

OC is characterized by calcium and phosphate deposition similar to osseous tissue in the dermis and subcutis.⁵ Current classifications into primary and secondary forms are based on the absence or presence of a preceding cutaneous lesion.^{2,6,7,10} Primary OC is divided into generalized and localized forms. In generalized OC, there are multiple widespread lesions that can be related to hereditary osteodystrophy (Albright's disease) or can be idiopathic. Localized OC may present in several ways: as a single large plaque present at birth or beginning in the first 2 years of life, as an isolated nodule in an older child or adult, or as MOF.¹¹ MOF is mostly seen in light-skinned, middle-aged to older adult women. It was first described by Hopkins in 1928.⁹

Secondary OC is more common than primary OC.⁷ The underlying cutaneous disorders mostly found in secondary OC are either tumors (pilomatricomas, chondroid syringomas, basal cell carcinomas, pilar cysts, nevi, sebaceous adenomas)^{1,6-9} or inflammatory disorders (scars, stasis dermatitis, dermatomyositis, scleroderma, folliculitis).^{1,6-9}

We present the findings of 4 new patients with MOF. Patients 1, 2, and 3 are typical and very similar to the cases previously reported in the literature. Patient 4 is probably the first described in a black patient, indicating that MOF is not exclusive to light-skinned persons. MOF in black women might be an underreported phenomenon.

There is debate as to whether MOF represents a primary form of osteoma or results from acne vulgaris scars.^{7,8} Some authors believe that acne is a precipitating factor of MOF rather than a cause.¹⁰ Because many patients reported in the literature (as well as 3 of our patients) recall having previous acne, it is probably an important predecessor of MOF. Because MOF is diagnosed much later in life, we also speculate that some patients might have forgotten they had acne. There are reported cases in which the lesions of MOF assumed a bluish tone because of tetracycline and minocycline deposition.^{12,13} Minocycline is known to deposit on acne scars.¹⁴ This fact supports theories relating MOF to acne scars.

Diagnosis of OC may be suspected clinically or radiographically, but can be confirmed only by

histopathology. Microscopic findings consist of wellformed and calcified bone arranged in spicules of spongy bone or sheets of compact bone. There are always identifiable osteocytes, and occasionally haversian system is seen. Howship's lacunes and osteoclasts are seen only rarely.² Decalcification of biopsied tissue can be performed for a better visualization of bone tissue.

Several theories have been proposed to explain the pathogenesis of OC; however, 2 are currently more accepted. The first theory involves a disordered embryological process in which primitive mesenchymal cells differentiate normally into osteoblasts but in a wrong location in the body (eg, the skin). The second theory concerns metaplasia—undifferentiated or already mature mesenchymal cells stimulated by unknown forces to assume an osteoblastlike role.^{6,8}

Treatment of MOF is difficult. Several modalities have been described, such as surgical excision, dermabrasion, topical tretinoin, ^{1,4,10} and more recently, destruction of lesions by the erbium:YAG laser. ¹⁵ Some lesions of patient 1 and 2 were excised with a 2-mm punch followed by 6-0 suture closure; patients 3 and 4 refused treatment.

Acknowledgment—We are grateful to Jaime Piquero, MD, for his help in studying patient 4.

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