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Review Article

Osteoma Cutis: Report of a Case and Literature Review

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Abstract

Osteoma Cutis is a rarely seen benign disease. Osteoma cutis (OC) is an abnormal development of bone within tegumentary tissue. OC is a benign and asymptomatic lesion, characterized by presence of ectopic osseous lamellae with osteoblastic cells in dermis and hypodermis.

The case described in this report regards a healthy 57-years-old man with a neoformation on left abdominal wall, without pathologic anamnestic history.

Keywords: Cutaneous ossification, Ectopic bone, Osteoma cutis

Introduction

Osteoma cutis is a cutaneous ectopic ossification caused by an anomalous growth of osteoblastic cells and osseous lamellae in dermis and hypodermis cutaneous layers. The first case of osteoma cutis was described in 1858 by Wilckets.[1] Cutaneous ossifications are divided into two categories[2]: primary and secondary. In the first category numerous hereditary pathologic conditions can be identified, such as Albright's ereditary osteodystrophy, Gardner syndrome, multiple widespread osteomas as in CREST syndrome and congenital progressive osseous heteroplasia (POH). Many of these sub-types of primary OC are related to genetic mutations, one of the most studied involving GNAS genetic complex locus.[3]

Secondary osteoma cutis is related to a pre-existent lesion: inflammatory skin disease, skin tumours, trauma and scars. On the previous skin pathology a process of ossification on a dystrophic basis is established.

Skin ossifications themselves are not symptomatic and do not affect the quality of life when not related to hereditary syndromes and in sporadic and solitary cases. Nevertheless, recent studies have provided novel insights into a common pathogenesis of acquired and genetic forms of heterotopic ossification, despite their different clinical presentation and biological course.[4]

OC represents a rare nosological entity with complex pathogenesis, but in most of cases it can be easily surgically removed.

Case Report

In the present article a rare case of osteoma cutis on abdominal wall is reported. A 1.5x1x2 cm cutaneous excision biopsy, obtained from the lesion, has been investigated in surgical pathology.

During the macroscopical description the specimen showed a calcified nodule with a maximum diameter of 1 cm localized in hypodermis. This nodule was a hard body difficultly at the sampled. First of all, the specimen was put in a decalcifying solution, than formalin fixed and paraffin embedded. Finally, so decalcified tissue was sectioned in micrometres slides and Hematoxylin and eosin stained.

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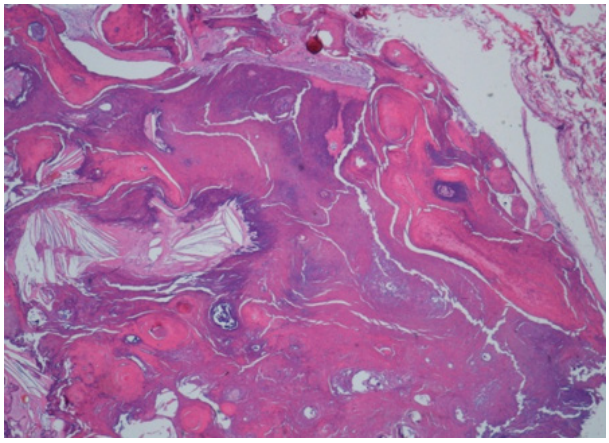

Figure 1.Hematoxylin and eosin slide lamellar bones (10x)

Histologically, a nodular well-defined lesion composed by lamellar bone was detected in hypodermis (Figure 1 and 2).

Higher magnification showed the presence of osteocytes in the centre and osteoclasts in the external area, as it can be detected in mature osseous tissue (Figure 3 and 4).

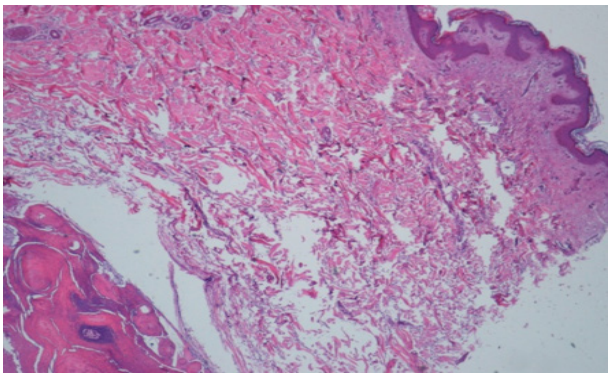

Figure 2.Hematoxylin and eosin show the growth of bone in hypodermis layers (10x)

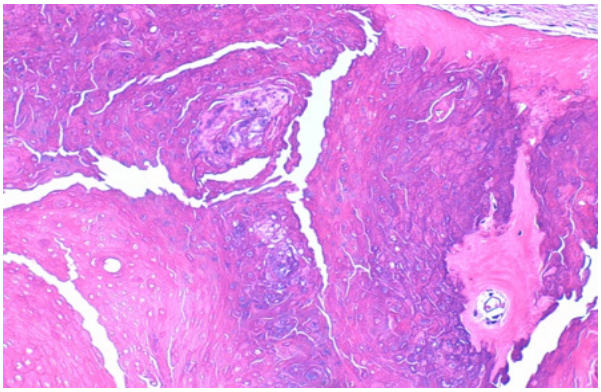

Figure 3.Hematoxylin and eosin slide with osteocyte in hypodermis (20x)

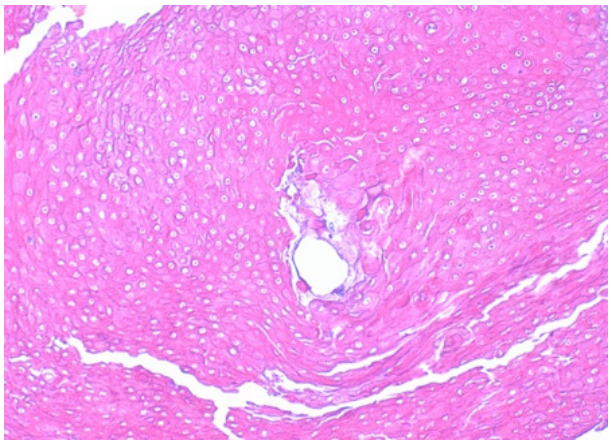

Figure 4.Lamellar bone at higher magnification (40x)

Discussion

The formation of bone through the deposition of calcium and phosphorus in a proteinaceous matrix is defined ossification. Osteoma cutis is a rare skin lesion linked to cutaneous ossification due to the differentiation of mesenchymal stem cells into bone tissue outside the context of the normal skeleton. OC is a rare disorder that generally arises during the middle age, even if many heterotopic cutaneous ossifications can be detected in children with congenital progressive osseous heteroplasia or Albright's ereditary osteodystrophy.

In literature most of cases of OC are reported in age range of 40-60 years. At the same time these cases manifest typically lesions in some cutaneous regions such as head, face, scalp, back, chest and, more rarely, fingers.[5-8]

Our case might be considered a rare localization, because the lesion has been removed from abdominal wall, a less described anatomic site of origin for heterotopic ossifications. The highest incidence of OC occurs among female patients.[9] These observations could be supported from the fact that in women hormonal alterations are more frequent and many cutaneous inflammatory diseases can be related with hormonal disfunction.

Previous primitive or secondary cutaneous disorders could be the cause of secondary OC. Acne vulgaris, scleroderma, pilomatricoma, dermatomyositis, basal cell carcinoma and scars from the removal of many cutaneous primitive lesions have been described in site of origin of osteoma cutis.[6-8]

Rare cases of primary osteoma cutis without previous diseases have been reported, some of them not related

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with calcium-phosphate abnormal metabolism or relevant clinical history.[10]

The pathogenesis of primitive and isolated OC is still unknown. Different theories have been supposed, the most reliable of which recognizes the probable cause in bone metaplasia of dermal fibroblasts.[11] In fact, fibroblasts might produce type I collagen and osteonectin, both of which are produced by normal osteoblasts. This pathogenetic theory agrees with the results of genetic analyses of congenital cutaneous ossification syndrome. In this last pathology, there have been detected mutations in the gene GNAS1, encoding the α-subunit of the stimulatory G protein that regulates adenyl cyclase activity.[10] These inactivating mutations reduce the levels of downstream proteins that are insufficient to maintain inhibitory control towards osteoblast differentiation in ectopic sites. More extensive molecular researches on solitary acquired osteoma cutis might revealed important in confirming etiopathogenetic correlation between primary and secondary cutaneous heterotopic ossification.

Conclusion

The case discussed in the present report has to be considered a primary and solitary osteoma cutis, because the patient hasn't a relevant clinical history or previous lesions. The best treatment choice was surgery through excision biopsy. No other treatments were considered in order of the fact that histopathological diagnosis of osteoma cutis was unexpected. The patient undergoes to clinical follow up and, in cases of other similar lesions, she can benefit from the use of retinoic acid with a satisfactory result. In this last case, the patient could be evaluated on genetic level to establish a molecular involvement of GNAS protein in formation of extra skeletal ectopic bone.

Conflict of Interest: None

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