Case Report DOI: 10.6003/jtad1263c4

Trichilemmal Cyst with Ossification and Marrow Formation: A Case Report

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Published:

J Turk Acad Dermatol 2012; **6 (3)**: 1263c4

This article is available from: http://www.jtad.org/2012/3/jtad1263c4.pdf

Key Words: Ossification, trichilemmal cyst, pilar cyst.

Abstract

Observation: Trichilemmal cysts, also known as sebaceous or pilar cysts, are found as solitary or multiple intradermal or subcutaneous nodules with a predilection for the scalp. They are derived from the isthmus of the hair follicle. They have keratinous material, which may contain cholesterol clefts and calcifications, in the lumen. Ossification with marrow formation is extremely rare. Here, we report an unusual case of ossifying trichilemmal cyst in a 40-year-old healthy woman. The possible mechanism is the formation of bone from osteogenic stromal elements secondary to cyst wall rupture.

Introduction

Trichilemmal cyst is a common cutaneous lesion, that arises from hair follicles. Their usual presentation is a hard nodule in the scalp. Although calcification is a common histopathological feature, ossification and marrow formation is extremely rare. There are a few case reports in the literature and here we report an other case in a 40-year-old woman that have mature bone with marrow inside and outside the cyst.

Case Report

A 40-year-old healthy woman presented with a nodular mass, measuring $2 \times 1 \times 1$ cm in the scalp. The lesion was locally excised for histological examination. The specimen was fixed in 10% formal-dehit. In the macroscopic examination, the material was a cystic lesion with keratin and a hard, bony material in the lumen. After decalcification, tissue was embedded in paraffin and stai-

ned with Hematoxylin-Eosin. Microscopic examination revealed a cyst, lined by pilar type epithelium, with keratinous material in the lumen. The cyst wall was ruptured and there were mature lamellar bone formation with hypocellular marrow inside and outside the cyst (**Figure 1**). No chondroid tissue was observed.

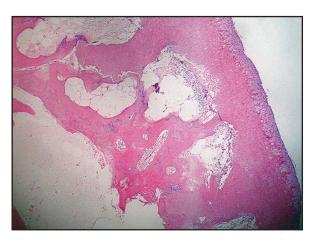


Figure 1. Mature bone and marrow in the lumen of the cyst (HE, original magnification is x 40).

Discussion

Trichilemmal cysts are smooth lesions with a cream to white wall and semi-solid, cheesy contents. The lining is stratified squamous epithelium showing tricholemmal keratinization in which the individual cells increase in bulk and vertical diameter towards the lumen. There is a sudden keratinization without the formation of a granular layer and an uneven interphase between the keratinized and nonkeratinized cells. The keratin inside the cyst is not lamellated, some of the nuclei are retained, and focal calcification is frequent [1].

In 1974, *Civatte* et al. reported a case of perforating ossified (trichilemmal) "sebaceous" cyst [2]. The osseous tissue was composed of haversian canals and medulla. Osteoblasts were present but there were no osteoclasts or cartilage. In this report, it is indicated that, he had previously observed two other cases of ossifying trichilemmal cysts and there are also three cases of ossifying keratinizing cysts previously described in literature [3, 4,5]. After this report, two isolated cases were reported in 2011 [6, 7]. *Pusiol* et al. reported a solitary cyst, while *Mommers* et al. reported multiple ossifying trichilemmal cysts in the scalp.

Calcification and ossification can be seen in cutaneous and subcutaneous tissues as a result of deposition of calcium salts and they are associated with some medical conditions. Cutaneous ossification is rarer than calcification and has traditionally been divided into two categories [8]. The primary form (osteoma cutis), where there is an absence of a preexisting or associated lesion, includes Albright's hereditary osteodystrophy, multiple miliary osteomas of the face, isolated osteoma, widespread osteoma and congenital plaque-like osteoma. In the secondary form (metaplastic ossification), which account for 85% of cutaneous ossifications, ossification develops in association with or secondary to a wide range of inflammatory (syphilis, pyogenic granuloma, folliculitis), traumatic or scarring (acne scars, injection sites, hematomas, surgical scars) and neoplastic process (metastatic bronchogenic carcinoma, basal cell carcinoma, Gardner syndrome, hemangioma). The most common cutaneous lesions showing ossification are nevi, basal cell carcinoma and pilomatricomas [8]. Although perforation of trichilemmal cyst wall may cause inflammation and foreign body type granulation tissue formation peri- and intracystically, they rarely show ossification [2, 6, 7].

The mechanism of cutaneous ossification is unclear, but the most accepted one is the metaplasia of the pluripotent mesenchymal cells to osteogenic cells (membraneous/mesenchymal ossification) [9]. Several bone-forming growth-regulating factors have been identified that may also participate in secondary ossification.

Here we report an ossifying trichilemmal cyst with marrow formation. The ossification was in both intra- and extracystic localisation. No chondroid tissue was observed.

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