Follicular Hybrid Cyst with Rare Juxtaposition of Epidermal Cyst and Steatocystoma

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ABSTRACT

Any cutaneous cyst differentiating toward two or more pilosebaceous components is known as follicular hybrid cyst (FHC). A combination of epidermal and trichilemmal cyst is its most frequent example. Other combinations of pilosebaceous derivatives occur uncommonly as well. The histogenesis of this condition has been controversial. In this latest report, we describe an unusual FHC from the earlobe of a 19-year-old male, which expressed the cohabitation of epidermal cyst and steatocystoma. A sharp transition was noted between the two kinds of epithelial components.

Key words: Earlobe, epidermal cyst, follicular hybrid cyst, pilosebaceous follicle, steatocystoma

INTRODUCTION

Cutaneous cysts usually develop as retention cysts from any of the three segments of a hair follicle, i.e., infundibulum, isthmus, and inferior portion. Occasionally, it exhibits two or more different epithelial derivatives from the pilosebaceous unit. Such cysts are known as follicular hybrid cysts (FHCs). They commonly express conjugal epidermal/infundibular and trichilemmal differentiation. However, other less frequent combinations exist as well. In this report, we describe a FHC located over the earlobe with features of epidermal cyst and steatocystoma.

CASE REPORT

At the surgical outpatient department, a 19-year-old boy presented with an insidious-onset, skin-colored firm subcutaneous nodule on the right earlobe for the past 2 years. It measured around 2 cm in diameter. A tiny surface punctum was also identified at its tip [Figure 1]. The lesion was enucleated under local anesthesia. Grossly, it appeared as well-encapsulated unilocular cyst beneath the epidermis. Its cavity contained oily cheesy substances. Under microscope, the cyst wall comprised two distinct patterns of pilosebaceous epithelium with an abrupt transition between them. Superficially, toward the epidermis, there was a stratified keratinizing squamous epithelium, reminiscent of the follicular infundibulum. It showed normal physiological maturation through stratum basale-spinosum-granulosum up to stratum corneum. Immediately juxtaposed to it, there was a multilayered epithelium characteristic of steatocystoma. This epithelium comprised three-to-four-cell layer thick strata of swollen sebaceous cells, which bore abundant foamy cytoplasm resembling as a delicate network. Toward the surface, these sebaceous cells were occasionally replaced by clear fat vacuoles, i.e., reminiscent of its disintegrated remnant in the process of holocrine secretion. Luminal to these cells, there was a discontinuous layer of stratified squamous epithelium without any intermediate granular layer. Focally, some multinucleate large polygonal cells with abundant...
dense granular cytoplasm were also present [Figure 2]. The cyst cavity contained keratin debris only. The ultimate diagnosis was rendered as “FHC with the components of epidermal cyst and steatocystoma.”

DISCUSSION

Pilosebaceous cysts include epidermal cysts, originating from the follicular infundibulum; trichilemmal cysts, from the isthmus; vellus hair cysts, from the developmental defect in the vellus hair follicles; and steatocystoma, from the sebaceous duct. Some authors also encompass pilomatrixoma and apocrine hidrocystoma within this spectrum. The term “FHC” is coined for lesions featuring two or more of these cystic components.\[2\] The pathogenesis of FHC has been debated as collision of multiple pilosebaceous cysts, or multidimensional differentiation of pluripotent follicular stem cells, or else metaplastic conversion of one component to the other.\[3\] The discussed FHC comprised epidermal cyst and steatocystoma, which originate from two contiguous pilosebaceous segments. Hence, the pathogenesis of this cyst is explainable by all those three pathomechanisms. Consequently, recognition of the appropriate histogenetic pathway involved in FHC requires further case studies including their scrutinizing molecular and genetic evaluation.

The maiden case of FHC was demonstrated by McGavran and Binnington,\[4\] which exhibited infundibular and trichilemmal differentiation. In subsequent years, the same combination turned into most frequent composition of FHCs.\[3\] Contradictorily, Kim et al.\[5\] reported seven FHCs of epidermal cysts combining with pilomatrixomas. Rarely, cysts featuring trichilemmal with pilomatrical, pilomatrical with steatocystoma, epidermal with apocrine, and vellus hair cyst with steatocystoma or epidermal or trichilemmal derivatives have also been described.\[3\] The coexistence of epidermal cyst with steatocystoma, like the present case, has been reported only once before. In that presentation, both the examined FHCs contained vellus hair cyst as the third component.\[6\] However, the currently described cyst exclusively consisted of epidermal cyst and steatocystoma. FHCs mostly arise in the face and scalp.\[2,3\] The present case only slightly differs for being located on the pinna. Histologically, there is usually a sharp transition between the epithelial components of FHCs.\[2,3\] However, few exceptional cases of combined epidermal cyst and pilomatrixoma featured a transformation zone at the interface.\[3,7\] Microscopically, the steatocystoma comprises 2–4 layers of swollen sebaceous cells with unapparent intercellular bridges. Keratinized squamous cells without interposing stratum granulosum cover its surface. Large polygonal cells with ample amount of
granular cytoplasm, of macrophage/monocyte lineage, occasionally form part of the cyst lining.\(^2\) Similarly, in the discussed case, there was a sharp demarcation between its epithelial components. The signature histomorphology of both compositions was maintained. At places, there was a relative predominance of sebaceous cells as well as the presence of multinucleate giant cells within the steatocystoma element. However, it did not pose any diagnostic difficulty at all.

**CONCLUSION**

FHCs are rare cutaneous appendageal pathology. Therein, conjugal juxtaposition of epidermal cyst and steatocystoma is furthermore rarer. With the detection of newer epithelial combinations, the pathomechanism of FHCs remains complicated. More implicit studies on the topic might be useful to amend a common summed pathogenesis for all the cutaneous cysts and tumors.

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**Conflicts of interest**

There are no conflicts of interest.

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