A Case of a Follicular Hybrid Cyst

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Various cutaneous cysts such as epidermal cysts, trichilemmal cysts, vellus hair cysts, steatocystoma, or pilomatricoma can arise from a different part of the pilosebaceous unit, namely the infundibulum, isthmus, sebaceous ducts, and bulbar or inferior portion. Rarely, a hybrid cyst that includes two or more components of a cystic lesion arising from the pilosebaceous unit can develop. The pathogenesis of this unusual disease is not yet known. We report a case of a follicular hybrid cyst which showed combined histologic features of both an epidermal cyst and pilomatricoma, and discuss the possible pathomechanism of the development of this hybrid cyst with a review of the literature.
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Key Words: Epidermal cyst, Follicular hybrid cyst, Pilomatricoma

INTRODUCTION

An epidermal cyst is quite a common cystic disease of skin, which originates from the follicular infundibulm and contains stratified squamous epithelium with lamellar keratinized material. Pilomatricoma, a tumor with differentiation toward hair matrix cells, histologically shows basophilic cells and shadow cells with non-lamellar homogenous keratinized material. Rarely, pilomatricoma-like changes with sharply-bordered shadow cells and homogenous keratinized material are observed in an epidermal cyst, especially in Gardner's syndrome[1], but the pathomechanism of the change is uncertain. Different types of cutaneous cysts such as epidermal cysts, trichilemmal cysts, vellus hair cysts, steatocystoma, or pilomatricoma can arise from each of pilosebaceous units, namely the infundibulum, isthmus, sebaceous ducts, and bulbar or inferior portion. The lesion, which includes more than two components of the cystic lesion arising from the pilosebaceous unit is called a follicular hybrid cyst. We report a case of a follicular hybrid cyst on the thigh with histologic features of both an epidermal cyst and pilomatricoma and discuss the possible pathogenesis of this hybrid cyst.

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CASE REPORT

A 28-year-old Korean woman presented with an 8-month history of a slowly-enlarging and hardening subcutaneous tumor on the lateral side of her left thigh. She initially recognized the lesion as a small bean-sized nodule but it slowly enlarged and hardened. There were no signs or symptoms of Gardner's syndrome. Physical examination revealed a 1.8 × 1.2 cm sized, solitary, skin-colored and firm subcutaneous nodule on the lateral side of her left thigh, without tenderness (Fig. 1). For the evaluation of the characteristics of the mass, an ultrasonogram was performed. The ultrasonogram showed a 1.8 × 1.0 cm sized, well defined, ovoid heterogenous mass in the subcutaneous layer, and the mass consisted of a hypoechoic cystic portion and hyperechoic solid portion with no vascularity under Doppler (Fig. 2). The tumor was totally

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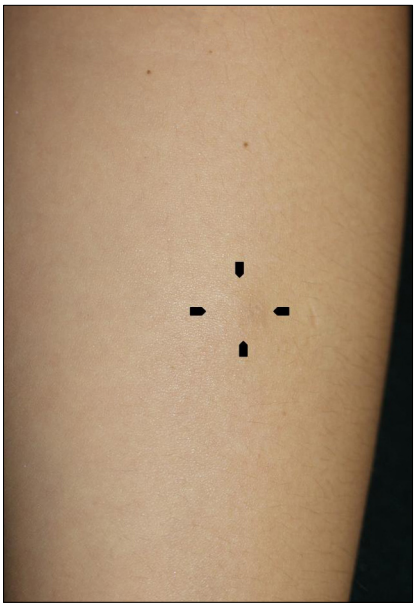

Fig. 1. A solitary, 1.8 × 1.2 cm in sized, skin-colored and firm subcutaneous nodule on the lateral side of the left thigh.

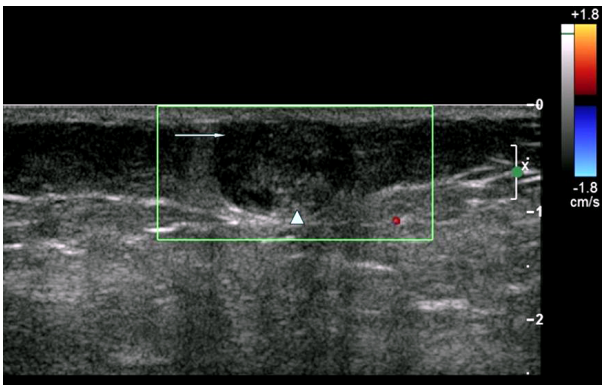

Fig. 2. Ultrasonography revealed a 1.8 × 1.0 cm sized, well defined, ovoid, heterogenous mass in the subcutaneous layer without vascularity, and the mass consisted of a hypoechoic cystic portion (arrow) and hyperechoic solid portion (arrowhead).

excised under local anesthesia for pathologic diagnosis and treatment.

Histopathologically, the mass in the lower dermis was well-encapsulated with a fibrous capsule and composed of an outer cellular portion and inner eosinophilic materials. Both the outer wall and inner material showed two different histologic patterns. One consisted of several layer of squamous cells, granular cells and horny materials arranged in laminated layers which is seen in epidermal cysts (Fig. 3A). The other showed basaloid cells, transitional cells, shadow cells, homogenous eosinophilic keratin materials and focal calcium deposition, which indicated pilomatricoma (Fig. 3B, 3C). We also observed a transitional zone of pilomatricoma to the epidermal cyst (Fig. 3D).

DISCUSSION

The follicular hybrid cyst was first described by McGavran and Binnington in 1966 as a cystic tumor that was a combination of an epidermal cyst and a trichilemmal cyst[2]. In the reports by Requena and Sanchez in 1991, the concept of a follicular hybrid cyst was extended to a cystic lesion that included more than two components of a cystic lesion arising from the pilosebaceous units such as an epidermal cyst, trichilemmal cyst, pilomatricoma, eruptive vellus hair cyst, steatocystoma, or apocrine hidrocystoma[3]. Some authors reported a few cases of eccrine sweat gland components with the pilocebaceous unit origin tumor, but they seemed to be an invaginated eccrine duct (not neoplastic) and embryogically the development of the eccrine sweat gland is different from that of the pilosebaceous unit[4].

Takeda et al.[5] reviewed 15 cases of follicular hybrid cysts in Japan. The scalp and face were most often involved, and the most frequent histological type was the combination of an epidermal cyst with a trichilemmal cyst (10 cases). In Korea, 5 cases of follicular hybrid cysts have been reported, including our case[6-9]. Three cases were similar to our case and the other one involved the combination of an eruptive vellus hair cyst and steatocytoma, and all of them except our case were located on the head or neck.

Cooper and Fechner reported that epidermal cysts developed in 50-60% of Gardner's syndrome and 63% of them showed pilomatricoma-like changes[1]. Squamous cells that projected from the epithelial lining of the cyst were hardly distinguishable from the shadow cells, and the cells at the base of the column were also hardly distinguishable from the basophilic cells of pilomatriocoma. They suggested that these finding could be a useful diagnostic marker of Gardner's syndrome. Leppard and Bussey

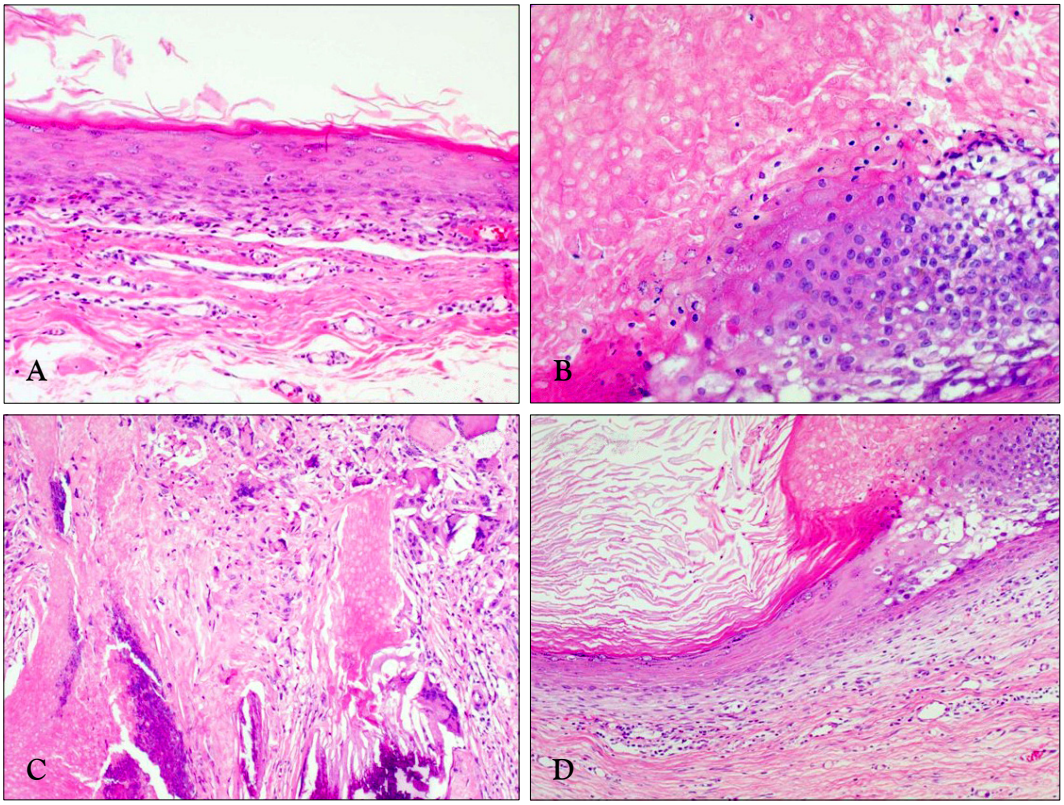

Fig. 3. Histologic features of the follicular hybrid cyst composed of an epidermal cyst and pilomatricoma. (A) Several layers of squamous cells, granular cells and horny material arranged in laminated layers. (B) Basaloid cells, transitional cells, and shadow cells. (C) Homogenous, eosinophilic, keratin material and focal calcium deposition. (D) Sharp transition zone between the epidermal cyst and pilomatricoma.

suggested that materials in the cyst cause a foreign body reaction which induces pilomatricoma-like change including shadow cells[10], and King suggested that it is just a transitional status from an epidermal cyst to pilmatricoma[11]. However, keratinized material of an epidermal cyst is from keratohyaline granules, which are not observed in pilomatricoma or the transitional zone of a follicular hybrid cyst[12]. Therefore the transition theory from an epidermal cyst to pilmatricoma is not likely and the pilomatricoma-like changes of epidermal cyst in Gardner's syndrome might not be true pilomatricoma. Just as part of the pilosebaceous unit and epidermis could originate from a bulge of the hair follicle, a follicular hybrid cyst might originate from multipotential stem cells.

Follicular hybrid cysts are very rare and interesting pathologic phenomenon, and studying this disease might be useful in understanding the pathogenesis of tumors from the pilosebaceous unit and multipotential stem cells of hair follicles.

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