Int J Clin Exp Pathol 2013;6(12):3019-3022
www.ijcep.com /ISSN:1936-2625/IJCEP1309026

Case Report

Follicular hybrid cyst: a combination of bullous pilomatricoma and epidermoid cyst

Tutyana Sanusi[1], Xiaoying Qu[2], Yanqiu Li[3], Jing Zhang[4], Ming Wang[1], Yun Zhao[1], Zhen Yang[1], Xiangjie An[1], Yue Qian[1], Chunsen Wang[1], Hongxiang Chen[1], Siyuan Chen[1], Changzheng Huang[1]

[1]Department of Dermatology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan 430022, China; [2]Department of Dermatology, The 457th Airforce Hospital, Wuhan 430022, Hubei, China; [3]Department of Dermatology, Zhongshan Hospital, Wuhan 430030, China; [4]Department of Dermatology, Wuhan General Hospital of Guangzhou Command, Wuhan, 430070, China

Received September 8, 2013; Accepted October 3, 2013; Epub November 15, 2013; Published December 1, 2013

Abstract: The follicular hybrid is composed of more than two components of pilosebaceous unit. There are several studies of hybrid cyst, combination of trichilemmal and epidermoid cyst was the most frequently reported. In this paper, we reported one case of hybrid cyst composed of bullous pilomatricoma and epidermoid cyst. A 14-year-old girl was complaint of a solitary flesh-colored to erythematous nodule with flaccid appearance sized 3.2×1.8 cm in diameter on her right upper back for one year. The histologic findings showed there were edema and proliferation of capillaries in the superficial dermis, a cyst in the middle to deep dermis. There were laminated keratins in the cystic space. The cyst wall was composed of two different components, one was composed of epithelial cells containing of granular layer, and another consisted of basophilic cells, transient cells and shadow cells. The cyst not related with Gardner's syndrome. Hybrid cyst such as trichilemmal cyst, epidermoid and pilomatricoma cysts maybe have same clinical features or mimicking each others, but we can distinguish them from histopathology evaluation.

Keywords: Hybrid cyst, bullous pilomatricoma, epidermoid cyst

Introduction

The follicular hybrid cyst is combination of infundibular (epidermoid) and trichilemmal cysts which include cyst wall components with pilomatrical differentiation, vellus hair cysts and steatocystoma, any types of cysts arising from the various parts of the pilosebaceous unit and can combine with others to form a large series of follicular hybrid cysts [1].

Takeda et al reviewed of 15 cases of follicular hybrid cysts in Japan, the most frequent cases was infundibular and trichilemmal cyst (60%), only 2 cases showed combination of epidermoid cyst and pilomatrical differentiation [2]. Herein we reported one case of hybrid cyst with combination of bullous pilomatricoma and epidermoid cyst. To our knowledge, this is the first case of this combination in the literature.

Case report

A 14-year-old girl presented with subcutaneous nodule on the right side of her upper back. The lesion had been present for 1 year and had increased in size gradually with a flaccid appearance. It was asymptomatic. On physical examination there was a solitary flesh-colored to erythematous nodule with the size of 3.2×1.8 cm in diameter on the right upper back of the patient. The lesion showed a thick walled bulla with flaccid appearance on the surface and hard consistency in the subcutaneous nodule (**Figure 1**). There was no family history of cutaneous diseases. None of the patient had any signs of Gardner's Syndrome.

The biopsy was taken from the lesion. Histologically, the superficial dermis showed edema and proliferation of capillaries filled with red blood cells and dilated lymphatics. There was a cyst in the middle to deep dermis which contained laminated keratins in the center of the cyst. The cyst wall was composed of two different components, one showed the feature of epidermoid cyst composed of epithelial cells containing of granular layer, and another pilom-

Figure 1. A solitary flesh to pink colored thick-walled bulla with flaccid appearance and an underlying palpable nodule with hard consistency sized 3.2×1.8 cm in diameter on the right upper back.

atricoma changes consisted of basophilic cells, transient cells and shadow cells (**Figure 2**).

Discussion

Epidermoid cysts are the most common benign cutaneous cyst, also called infundibular cyst. Epidermoid cyst has a wall composed true epidermis that is squamous, granular, and horn cells. The cyst is filled with horny material arranged in laminated layers [3]. Pilomatricoma or pilomatrixoma is a uncommon types of benign skin tumor which associated with hair follicles. The cyst is often well circumscribed and composed of epithelial islands embedded in cellular stroma. Two types of cells comprise the islands: basophilic cells and shadow cells. The basophilic cells resemble hair matrix cells. The shadow cells show a central unstained shadow at the site of the lost nucleus in the center of the field, one can see transformation of the basophilic cells into shadow cells [4].

Hybrid cyst was a combination of infundibular and trichilemmal cyst, described by McGavran et al [5]. Brownstein in 1983 reported a mixed type of cutaneous cyst characterized by infundibular and trichilemmal keratinization [6]. Nevertheless hybrid cyst should not be restricted to those composed only of epidermoid and trichilemmal cyst. Requena et al, expanded the concept of hybrid cyst. Hybrid cyst is a combination of infundibular (epidermoid) and trichilemmal cysts to include cyst wall components with pilomatrical differentiation, vellus hair cysts and steatocystoma, any types of cysts arising from the various parts of the pilosebaceous unit and can combine with others to form a large series of follicular hybrid cysts [1]. The hybrid cysts also include a combination of infundibular cyst and apocrine hydrocystoma [7].

Takeda et al. reviewed of 15 cases of follicular hybrid cysts in Japan, the most frequent cases was infundibular and trichilemmal cyst (60%), only 2 cases showed combination of epidermoid cyst and pilomatrical differentiation [2]. There have been several studies on hybrid cyst, in this case we reported one case of pilomatricoma like change in the epidermal cyst, the patient did not have any feature of Gardner's Syndrome, even though Cooper et al, has hypothesized that pilomatricoma like change in the epidermal cyst may be feature of Gardner's Syndrome [8].

A true hybrid cyst from pilomatricoma and epidermoid cyst is a single cyst with both types of keratinization in its wall, epidermoid in the upper areas connected with epidermis surface and hair matrix-like in deeper zones of the cyst wall. The transition between both zones is also sharp. A combined pilomatricoma cyst and epidermoid should not be confused with the so-called perforating pilomatricoma, which consisted of masses of shadow and basophilic cells of pilomatricoma that are extruded from the upper dermis to the skin surface through a perforated epidermal channel. The pilomatricoma like changes in epidermoid cyst of Gardner's syndrome are not regarded as true pilomatricoma [1].

Bullous pilomatricoma is a special variant of pilomatricoma. Chen SY et al reviewed 16 cases of bullous pilomatricoma which mainly occurred on shoulders and upper arms at 10-20 years old people with a female predominance [9]. None of which has been found to have a combination with epidermoid cyst. To our knowledge, our case is the first hybrid cyst composed of bullous pilomatricoma and epidermoid cyst published in the literature.

A combination of Hybrid cyst between pilomatricoma cyst and epidermoid is still a rare condition, only few cases have been reported [2, 10]. The pathogenesis of their coexistence at the

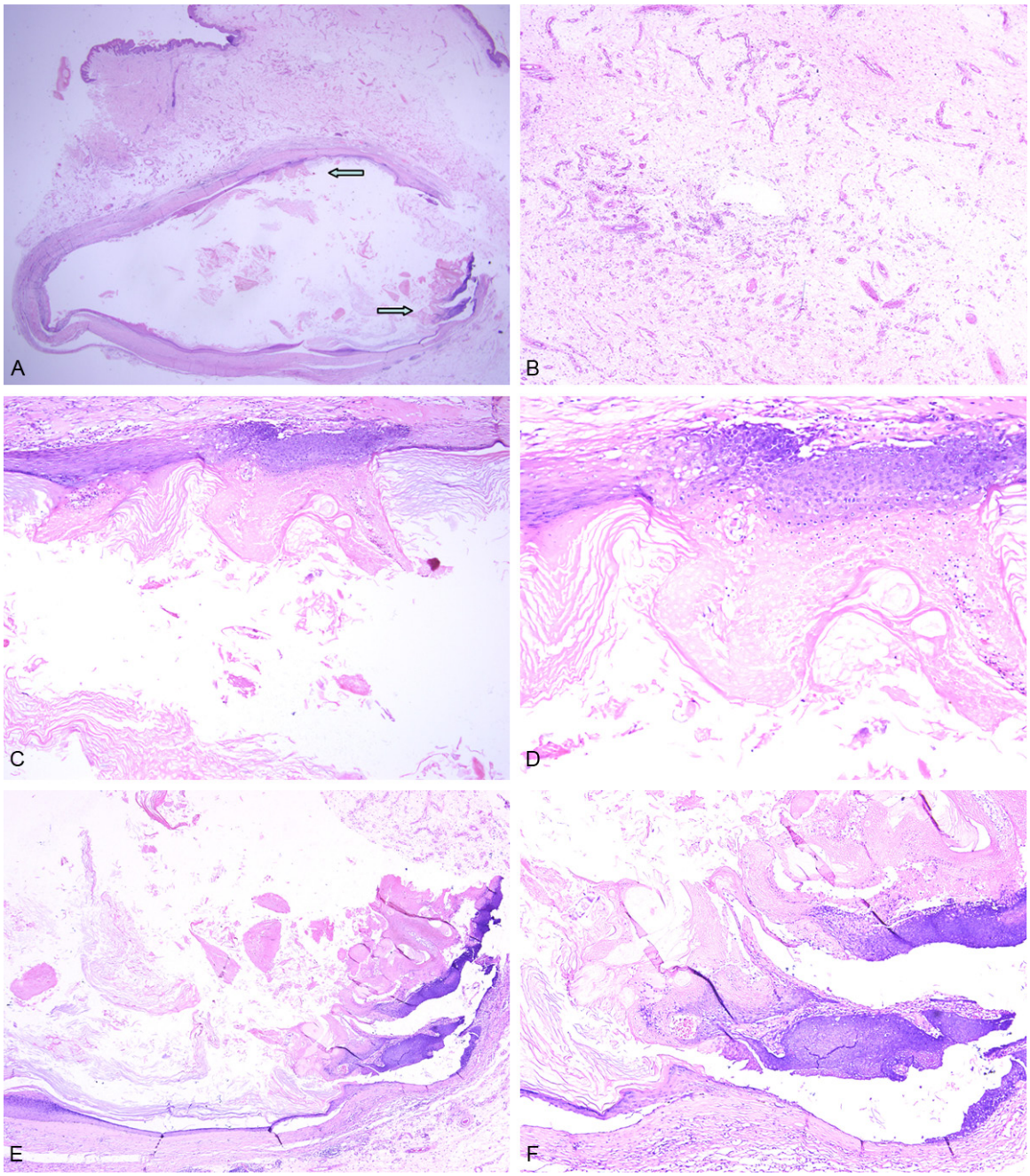

Figure 2. The superficial dermis showed edema, proliferation of capillaries, dilated lymphatics. There was a cyst in the middle to deep dermis (A and B). There were laminated keratins in the center of the cyst. The cyst wall showed both epidermoid epithelium containing granular cell layer and epidermoid keratinization and pilomatricoma like changes on the upper (arrowed and as shown in C and D) and lower parts (arrowed and as shown in E and F) (HE: A×2.5; B×40; C×100; D×200; E×40; F×100).

same site is still not fully understood. Hybrid cyst such as trichilemmal cyst, epidermoid and pilomatricoma cyst maybe have same clinical features or mimicking each other's, but we can distinguish it from histopathology evaluation. The cysts often require by surgical excision, and recurrent of hybrid cysts can be attributed to traumatic surgery [11].

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Siyuan Chen or Dr. Changzheng Huang, Department of Dermatology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan 430022, China. Tel: +86-27-85726195; E-mail: siyuanc_cn@126.com (SYC); hcz0501@126.com (CZH)

References

[1] Requena L, Sanchez Yus E. Follicular hybrid cysts. An expanded spectrum. Am J Dermatopathol 1991; 13: 228-233.

[2] Takeda H, Miura A, Katagata Y, Mitsuhashi Y, Kondo S. Hybrid cyst: case reports and review of 15 cases in Japan. J Eur Acas Dermatol Venereol 2003; 17: 83-86.

[3] Fieselman DW, Reed RJ, Ichinose H. Pigmented epidermal cyst. J Cutan Pathol 1974; 1: 256.

[4] Solanki P, Ramzy I, Durr N, Henkes D. Pilomatricoma. Cytologyc features with differential diagnostic considerations. Arch Pathol Lab Med 1987; 111: 294-297.

[5] McGavran MH, Binnington B. Keratinous cysts of the skin. Identification and differentiation of pilar cyst from epidermal cysts. Arch Dermatol 1966; 94: 499-508.

[6] Brownstein MH. Hybrid cyst: a combined epidermoid and trichilemmal cyst. J Am Acad Dermatol 1983; 9: 872-875.

[7] Anderson WK, Rao KR, Bhawan J. The hybrid epidermoid and apocrine cyst: a combination of apocrine hidrocytoma and epidermoid inclusion cyst. Am J Dermatopathol 1996; 18: 364-366.

[8] Cooper PH, Fechner RE. Pilomatricoa-like change in the epidermal cysts of Gardner's syndrome. J Am Acad Dermatol 1983; 8: 639-644.

[9] Chen SY, Wu F, Qian Y, Zhu L, Tu YT, Huang CZ. Pilomatricoma with bullous appearance: a case report and review of literature. Int J Dermatol 2011 May; 50: 615-618.

[10] Tadashi T. Cutaneous hybrid tumor composed of epidermal cyst and cystic pilomatricoma expressing p53 and high Ki-67 labeling. Int J Clin Exp Pathol 2013; 6: 1187-1189.

[11] Pujani M, Agarwal S, Goyal RK, Jyotsna PL, Tejwani N, Rautela A. Pilomatricoma coexistent with epidermal cyst. Int J Dermatol 2012; 51: 624-625.