A Case of Apocrine Hidrocystoma

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Apocrine hidrocystoma is an uncommon benign tumor derived from the secretory segment of apocrine sweat glands and another name for it is apocrine cystadenoma.

A 48-year-old female patient had a solitary, translucent red-brownish cystic nodule, 10×6×7mm in size, on the periorbital area of her left eye. The lesion had enlarged very slowly for 25 years. We excised the lesion and identified it as apocrine hidrocystoma. This is a case of apocrine hidrocystoma with typical clinical and histopathological findings.

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Key Words : Apocrine hidrocystoma, Apocrine cystadenoma

Apocrine hidrocystoma, also called apocrine cystadenoma, is a benign tumor of the apocrine sweat glands. Clinically this tumor usually occurs singly as a smooth, dome-shaped translucent nodule that possesses a flesh colored or red-brown, or bluish hue. This tumor is usually found on the face, especially the periorbital area and measures less than 15mm in size.

It was first described by Mehregan in 1964[1] and two cases of apocrine hidrocystoma have also been reported in the Korean literature[4,5]. We add here a typical case of apocrine hidrocystoma in a 48-year-old female.

CASE REPORT

A 48-year-old female came to our clinic with a cystic nodule on her left periorbital area which had been present for about 25 years. The tumor was a solitary, translucent red-brownish, raised, 10×6×7mm sized nodule with well-defined margins(Fig. 1). The lesion was mobile and had a cystic consistency and a smooth surface. The lesion was growing slowly in size and she decided to try and do something about the lesion. She herself tied the base of the nodule tightly with her scalp hair 7 days prior to visit and complained of mild tenderness. However, there were no other symptoms and the tumor was surgically excised under local anesthesia.

A histopathological examination revealed several well-circumscribed, cystic structures in the dermis and the cyst wall was lined by columnar to cuboidal cells with a decapitation secretion(Fig. 2). The stroma surrounding the cystic space was composed of fibrous tissue. The secretory cells contained a large amount of periodic acid-Schiff (PAS)-positive, diastase-resistant granules(Fig. 3). Immunohistochemical staining was done for carcinoembryonic antigen (CEA), epithelial membrane antigen (EMA) and S-100 protein. The inner surface of the cyst wall was positively stained with CEA(Fig. 4) and EMA, but not with S-100 protein.

After excision there was no recurrence during one year of follow up.

DISCUSSION

Apocrine hidrocystoma is an uncommon benign tumor derived from the secretory segment of apocrine sweat glands[1,2,3]. Some reserve the term apocrine hidrocystoma for fluid-filled cystic structures lined with a mature apocrine glandular epithelium with few if any papillary infoldings, and refer to neo-

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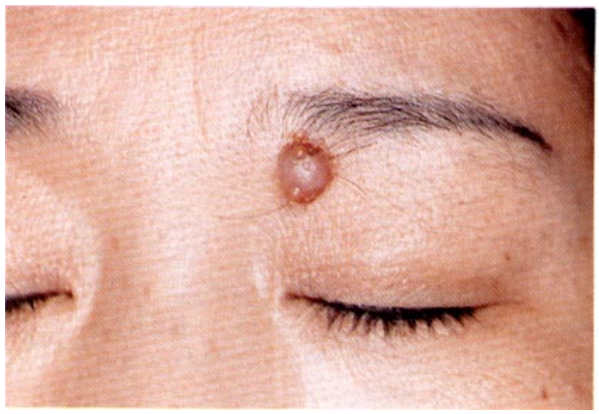

Fig. 1. 10× 6× 7mm sized, translucent red-brownish cystic nodule on the left periorbital area.

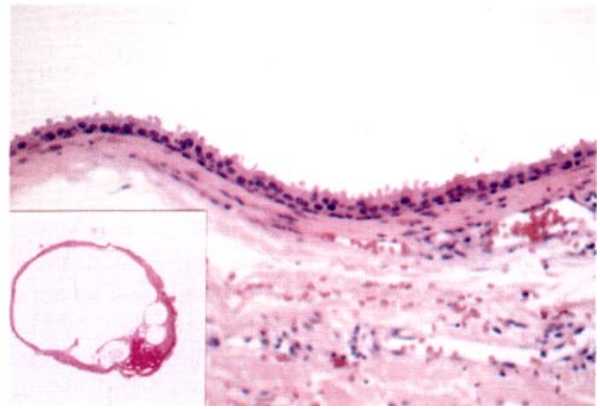

Fig. 2. The cyst wall is lined by secretory cells showing decapitation secretion(H&E stain, ×200). Inset: Low power photograph of the lesion shows several cystic structures in the dermis.

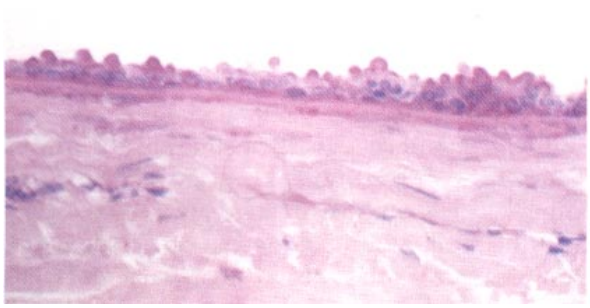

Fig. 3. PAS-positive, diastase-resistant granules are demonstrated within the cytoplasm of the secretory cells (dPAS stain, ×400).

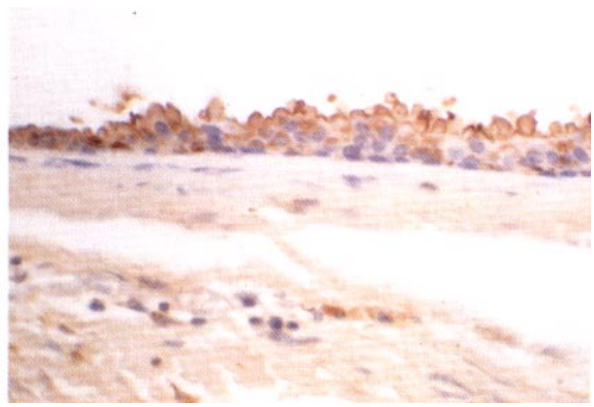

Fig. 4. Immunohistochemical staining for CEA shows a positive reaction in the cytoplasm and the luminal side of the epithelium (CEA, ×400).

plasms comprising both cystic and solid structures of apocrine glandular epithelium as apocrine cystadenomas[3]. However, clinically they do not differ, and histologically there is a broad spectrum of tumors between them[3] and we view apocrine hidrocystoma and cystadenoma as variants of the same pathological entity.

Histopathologically, the cyst wall of this tumor consists of columnar to cuboidal cells with a decapitation secretion indicative of apocrine secretion[1,2,3]. In addition, PAS-positive, diastase-resistant granules are identified in the cytoplasm of secretory cells[2,3,6,7]. Recently positive staining with antibodies to the breast/apocrine marker gross cystic disease fluid protein (GCDFP)-15 and lysozyme have been found useful in the diagnosis of tumors specific to apocrine sweat glands[8,9]. Immunohistochemical staining for S-100 protein is negative in normal apocrine glands as well as in apocrine hidrocystoma[10]. When positive staining for S-100 protein has been found in lesions histologically consistent with apocrine hidrocystoma, this appearance has been attributed to differentiation toward eccrine secretory cells, rather than toward cells of apocrine origin[10,11]. In our case, immunohistochemical stainings for GCDFP-15 and lysozyme were not done. Also, histological findings of our case showed that some part of the cyst wall was flattened and there were a few papillary projections into the cystic space. These findings may be due to increased intracystic pressure.

Apocrine hidrocystoma shows an equal sex inci-

dence and usually arises in the middle-aged6,7. The diameter varies between 3 and 15 mm[12] and the usual location is on the face, especially the periorbital area, but it is occasionally seen on the ears, scalp, chest, shoulders, or genitalia[4,5,6,7,13,14,15]. When localized on the male genitalia, it must be differentiated from the median raphe cyst of the penis, which has been mistakenly reported as apocrine hidrocystoma[16].

The color of the tumor can vary from flesh-colored to blue, or red-brown like our case and approximately 50% are pigmented[6,7]. Although the origin of the dark color in the apocrine hidrocystoma has not yet been established, the color seems to be due to the Tyndall effect or the lipofuscin-rich fluid content of the cyst, because there are no melanophages or melanocytes within the surrounding stroma[7,17]. On the other hand, multiple apocrine hidrocystomas are only rarely encountered[18]. Alessi et al[18] suggested that multiple apocrine hidrocystoma may be a marker of two rare inherited disorders, the Sch pf-Schultz-Passarge syndrome and a peculiar form of focal dermal hypoplasia.

Although the surgical excision is a usual treatment of apocrine hidrocystoma, carbon dioxide (CO2) laser vaporization is an effective treatment of multiple apocrine hidrocystomas[19].

Apocrine hidrocystoma was first described by Mehregan in 19641 and then many cases were reported. According to the literature more than one-hundred cases have been reported so far[7]. Apocrine hidrocystoma has rarely been reported in our country[4,5] but we suggest that there are probably numerous unreported cases and our case had typical clinical and histopathological findings. Therefore apocrine hidrocystoma should be considered in the differential diagnosis of solitary pigmented tumors of the face, particularly the periorbital area.

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