CASE REPORT

Giant apocrine hidrocystoma of the scrotum: An uncommon benign tumor at an exceptional site

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Introduction: Apocrine hidrocystoma (AH) is a rare, benign cystic tumour of apocrine glands, typically occurring in adults between 30 and 70 years of age without gender predilection. It most frequently affects the head and neck region, while genital localization, particularly in the scrotum, is exceedingly uncommon. To the best of our knowledge, this is the first reported case of giant scrotal apocrine hidrocystoma in an adult patient.

Case presentation: A 65-year-old male presented with a painless, translucent cystic nodule in the scrotal region gradually enlarging over several months. The lesion measured 45x30x25 mm and was surgically excised. Histopathological examination revealed a well-circumscribed, unencapsulated cystic lesion within the dermis, lined by two cell layers: an inner layer of apocrine cells exhibiting decapitation-type secretion and an outer myoepithelial layer. Immunohistochemical staining demonstrated AE1/AE3 positivity in luminal cells, and p63 expression in myoepithelial cells, confirming the diagnosis of AH.

Conclusion: Apocrine hydrocystoma of the scrotum is an extremely rare benign lesion and therefore represents a diagnostic challenge. Awareness of this entity is essential to avoid misdiagnosis with other cystic or adnexal lesions.

Keywords: apocrine hidrocystoma, scrotum, giantm cystic lesion

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Introduction

Apocrine hidrocystoma (AH) is a rare, benign cystic tumor of apocrine glands, typically affecting adults between 30 and 70 years of age, with no gender predilection [1,2].

Although tumor shows apocrine differentiation [1], it is very rare to be found in sites rich in apocrine glands, such as groin, axilla and anogenital region [3]. The most common location is head and neck area, with predilection for eyelids [4]. Usually, these tumors measure up to 15 mm in diameter [1], but giant cases, larger than 2 cm in diameter, have also been reported [5, 6].

Genital involvement of AH is exceptionally rare, with only a small number of cases reported in adult and pediatric patients [3, 7-9].

Scrotum is an exceedingly rare site of AH origin, reported for the first time in 1999 [8]. Later, few cases have been reported, mainly in pediatric population [9-11].

According to our review of the available literature, giant apocrine hidrocystomas in the anogenital region have been reported only rarely, and to date, no case strictly confined to the scrotal skin in an adult has been documented.

Case presentation

A 65-year-old male presented to the urology clinic with a progressively enlarging, painless, translucent cystic nodule in the scrotal region, which had developed over the past 3-4 months (Figure 1.). The patient's medical history revealed no prior trauma or tumors of the anogenital region. The urologist recommended surgical excision.

On gross examination, the specimen measured 45x30x25 mm. It consisted of a cystic lesion filled with clear yellowish fluid. The inner surface appeared smooth, while the wall was grayish, of variable thickness, and contained multiple small cystic spaces within the dermal zone.

On histopathologic examination, the epidermis showed no significant abnormalities. Within the dermis, a well circumscribed but unencapsulated cystic lesion was identified, without connection to the overlying epidermis. The lesion consisted of one large cystic cavity accompanied by numerous smaller ones (Figure 2A). The cyst wall was lined by two distinct cell layers: an outer myoepithelial layer and an inner layer of mature apocrine epithelial cells exhibiting

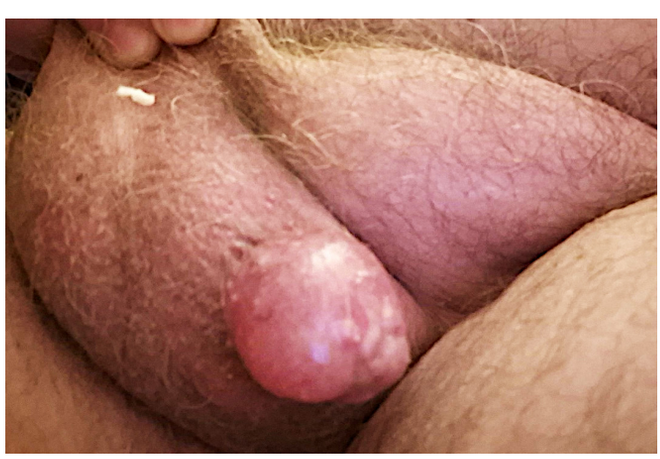

Fig. 1. Painless, translucent cystic nodule in the scrotal region.

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decapitation-type secretion and occasional papillary projections (Figure 2B). The cyst wall stroma showed areas of hyalinization, and the cyst lumen contained homogeneous eosinophilic secretory material.

Immunohistochemically, the luminal epithelial cells demonstrated CK7 (Figure 2C), AE1/AE3 (Figure 2D) and CEA (Figure 2E) positivity, while myoepithelial cells were p63 positive (Figure 2F).

Discussion

Apocrine hidrocystoma is now regarded as a proliferative tumor arising from the secretory part of the apocrine sweat glands, rather than a simple retention cyst, as previously believed [12].

Although sun exposure has been suggested in the literature as a potential risk factor for apocrine hidrocystoma, contemporary authors emphasize that the exact stimulus for AH proliferation remains unknown [1].

Since apocrine glands are also present in the anogenital region, the occurrence of hidrocystoma on scrotal skin, although extremely rare, is patophysiologically possible, even in the absence of UV exposure. Although the majority of AH develops on sun-exposed skin, our case further supports the notion that sun exposure is a possible but not

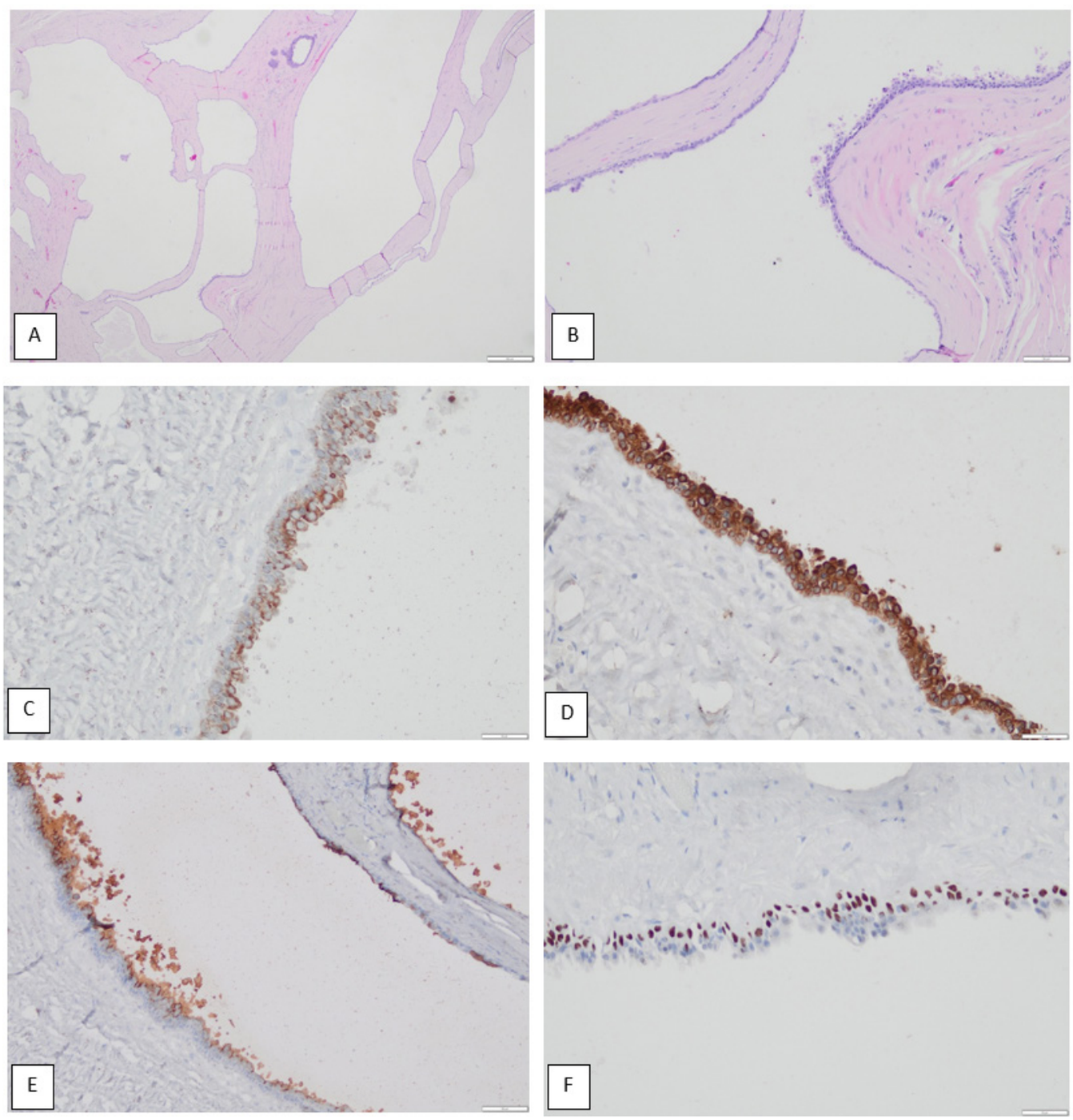

Fig. 2. A. Multilocular cystic cavity (HE, x 20); B. lined by two distinct cell layers: an outer myoepithelial layer and an inner layer of mature apocrine epithelial cells exhibiting decapitation-type secretion and occasional papillary projections (HE, x100); C. CK7 positivity of luminal cells (x200); D. AE1/AE3 positivity of luminal cells (x200); E. CEA positivity of luminal cells (x100); F. p63 positive myoepithelial layer (x200).

obligatory risk factor, and that intrinsic ductal or glandular factors, rather than UV exposure alone, may be responsible for lesion formation. Other proposed mechanisms include occlusion of the apocrine sweat gland duct; however, the more recent concept suggests that the lesion represents a dysregulated proliferative process of the apocrine secretory unit, resulting in cystic growth [1, 3].

Despite being rich in apocrine glands, scrotal skin is rarely affected. Although no definitive explanation exists in the literature, this may be related to differences in local glandular activity, microenvironment, or a lower tendency for ductal obstruction compared with other anatomical sites.

These tumors usually present as single, painless intradermal, moderately firm, dome-shaped, translucent, purple, or bluish cystic nodules. Multiple lesions may occur and are sometimes associated with different syndromes [1, 7]. They typically affect adults, with the most common localization being the head and neck region, especially the periocular area [3].

Several cases of scrotal AHs have been reported in the pediatric population. Flessati et al. for the first time described a scrotal apocrine hidrocystoma in a a child [8], while Park et al. reported multiple linear papules on the scrotum of a four-year-old boy, subsequently diagnosed as apocrine hidrocystoma [11]. More recently, Cuvelier et al. documented transient apocrine hidrocystomas of the scrotum in two infants [11], and Noviello et al. included a 15-year-old boy with a scrotal cystic lesion in their pediatric case series [10]. These reports highlight that, although uncommon, AH may arise in the anogenital region across a wide age range.

A summary of all previously published cases involving the genital area is provided in Table 1.

A review of the available literature, performed using PubMed and Google scholar, identified a total of 18 previously published articles related to apocrine hidrocystomas in genital region [3, 7-11, 13-24], encompassing 23 individual cases. Among these, 12 cases involved adult patients, while 11 cases involved children and adolescents, ranging from 4 months to 17 years. The reported lesions included those on the prepuce, penile shaft, glans penis, vulva, urethral meatus, and less commonly, the scrotum.

In the majority of published cases for which treatment information was available, complete surgical excision was performed, with no recurrence reported during follow-up periods ranging from six months to one year. An exception is the series by Cuvelier et al., who described two infants with multiple transient apocrine hidrocystomas of the scrotum, in whom lesions regressed spontaneously without the need for surgical intervention [9].

Clinical differential diagnosis of AH in genital region is broad, and includes various benign and malignant entities, depending on gross appearance of the lesion. Main differential diagnoses include median raphe cysts, eccrine hidrocystomas, epidermal inclusion cyst, sclerosing lymphangitis and acquired lymphangiomas. Less common differential diagnoses, especially when grossly presented as bluish or pigmented lesion, include basal cell carcinoma, blue nevi, or even cutaneous melanoma [1, 3].

Because the scrotal location is very rare, it poses a diagnostic challenge, therefore histopathological evaluation remains essential for definitive diagnosis. Surgical excision

Table 1. Summary of previously published cases involving the genital area

Author	No of cases	Age (years)	Sex	Location	Size (cm)
Ahmed et al.[13]	2	29	Male	Prepuce	1.0
		56	Male	Prepuce	NA
De Dulanto et al.[14]	1	49	Male	Prepuce	3.0
Powell et al.[15]	1	23	Male	Penile shaft	1.3x0.3x0.2
Glusac et al. [16]	1	34	Female	Labium major	3.0
Flessati et al. [8]	1	8	Male	Scrotum	0.3-1.0
Mataix et al. [17]	1	35	Male	Penile shaft	1.5
Samplaski et al. [18]	1	6	Male	Glans Penis	0.3x0.3
Liu et al. [19]	1	25	Male	Penile shaft	1.2
López et al. [20]	1	40	Male	Penile shaft	1.3x0.5
Park et al. [11]	1	4	Male	Scrotum	NA
Taylor et al. [21]	1	16	Male	Urethral orrifice	0.8
Jo et al.[3]	1	39	Male	Penis	4.0x0.3
Cuvelier et al. [9]	2	4 months	Male	Scrotum	<0.1
		6 months	Male	Scrotum	
Noviello et al. [10]					
	4	15	Male	Glans penis	2.0
		9	Male	Glans penis	0.4
		15	Male	Scrotum	3.0
		17	Male	Penis	0.8
Chang et al. [22]	1	4	Female	Vulva	NA
Val et al. [23]	1	52	Male	Foreskin	2.0x1.5
Dubuc et al. [24]	1	73	Male	Prepuce	NA
Bharti et al. [7]	1	29	Male	Penile shaft	2.0x2.0
Our case	1	65	Male	Scrotum	4.5x3.0x2.5

NA- not assessed

remains the treatment of choice, with a low risk of recurrence when completely excised [3, 8].

Histopathologically, apocrine hidrocystomas are characterized by unilocular or multilocular cystic spaces lined by an inner secretory epithelium showing apical decapitation-type secretion, overlying an outer myoepithelial layer [3]. Immunohistochemistry is usually not required for routine diagnosis, but luminal epithelial cells typically express CK7, AE1/AE3, CK18, CEA and EMA positivity, while outer myoepithelial cells are positive for SMA and p63 [7, 17]. We confirmed the diagnosis of AH by immunohistochemistry and found CK7, AE1/AE3 and CEA positive luminal cells, and p63 positive myoepithelial cell layer. Eccrine hidrocystoma is histologically most similar lesion, but differs from AH by the absence of decapitation secretions, and absence of p63 and SMA positivity. Also, eccrine hidrocystomas are predominantly unilocular lesions, rarely larger than 5 mm in diameter [4]. Another important differential diagnosis in genital region is median raphe cyst, but it lacks apocrine features and is lined by pseudostratified columnar epithelium [3]. The other mentioned lesions, although they may cause clinical confusion, are histologically clearly distinct from AH and do not require immunohistochemical analysis for accurate diagnosis.

Conclusion

Apocrine hidrocystoma of the scrotum is an exceptionally rare benign cystic lesion. Accurate diagnosis relies on detailed histological examination with immunohistochemical confirmation, when necessary. Complete surgical excision is curative, with an excellent prognosis and minimal risk of recurrence. Reporting such rare cases helps broaden awareness and understanding of apocrine gland tumors in unusual locations.

Author's contribution

NČ (Writing – original draft, Methodology, Conceptualization, Visualization); MD (Data curation, Investigation, Validation, Writing – review & editing, Supervision).

Conflict of interest

None to declare.

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Institutional review board statement

Ethical approval was not required for this descriptive case report; however, written informed consent was obtained from the patient.

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