CASE REPORT

Apocrine Hidrocystoma on the Penis: Report of a Case and Review of the Previous Cases

Jeong Won Jo, Jung Wook Yang¹, Do Seon Jeong*Departments of Dermatology and ¹Pathology, Gyeongsang National University School of Medicine, Jinju, South Korea*

Apocrine hidrocystoma, also called apocrine cystadenoma, is a benign cystic tumor-like lesion that arises from the proliferation of apocrine glands. Clinically, it usually occurs singly as a unilocular or multilocular, dome-shaped translucent cyst. Histologically, it appears as unilocular or multilocular cysts composed of an inner layer of single or double layer of secretory columnar epithelium with decapitation secretion lying above an outer myoepithelial cell layer. Apocrine hidrocystomas mostly occur within the head and neck region and involvement of genitalia is extremely rare. This paper emphasizes the importance of considering the differential diagnosis of a genital cystic lesion. Herein, we report a case of apocrine hidrocystoma occurring in the penis and compare the clinicopathological characteristics of apocrine hidrocystoma in genitalia with the previous cases. (*Ann Dermatol* 31(4) 442~445, 2019)

-Keywords-

Apocrine glands, Hidrocystoma, Penis

INTRODUCTION

Hidrocystomas are rare, benign, cystic lesions of sweat gland resulting from proliferation of the apocrine secretory coil or eccrine duct¹. Apocrine hidrocystoma, also called apocrine cystadenoma, is a benign cystic tumor-like lesion usually presenting as a solitary translucent nodule of cystic consistency². The apocrine glands are most frequent in the axilla, external auditory canal, eyelids, and on the nipple, and it is not surprising that most apocrine hidrocystomas occur within the head and neck region³. However, involvement of genitalia is extremely rare and a few cases have been reported³⁻¹³.

CASE REPORT

A 39-year-old male patient presented with a mass on the penis that had been growing slowly for 1 year (Fig. 1). On clinical examination, about 4.0×0.3 cm, serpiginous cyst was found. It was not tender, the skin over the cyst was normal and it was not attached to the penile shaft. There were no signs of pain, tenderness, pruritus, or other symptoms. His past medical history was irrelevant. He denied traumatism and risky or vigorous sexual activities. Patient was taken up for surgery and excision of the mass was performed under local anaesthesia. Histological evaluation revealed the presence of a cyst in the dermis (Fig. 2). The inner surface of the cyst and the papillary projections are lined by a row of columnar secretory cells of variable height showing decapitation secretion indicative of apocrine secretion. Peripheral to the layer of secretory cells are elongated myoepithelial cells. Immunohistochemically, the inner cells show cytoplasmic stains for cytokeratin (CK) 7 and outer cells show nuclear stains for p63, respectively. CK20 and carcinoembryonic antigen (CEA) are negative in both cells. A diagnosis of apocrine hidrocystoma of the

Received January 5, 2018, Revised June 18, 2018, Accepted for publication June 20, 2018

Corresponding author: Do Seon Jeong, Department of Dermatology, Gyeongsang National University Hospital, 79 Gangnam-ro, Jinju 52727, Korea. Tel: 82-55-750-8187, Fax: 82-55-758-8106, E-mail: dr7123@hanmail.net
ORCID: <https://orcid.org/0000-0002-5857-5670>

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Copyright © The Korean Dermatological Association and The Korean Society for Investigative Dermatology

penis was established.

DISCUSSION

Apocrine hidrocystoma is a benign cyst tumor that arises from the proliferation of apocrine glands in adults be-

Fig. 1. Clinical photograph of serpinginous cyst measuring about 4.0×0.3 cm on the penile shaft.

tween 30 and 70 years of age¹¹. The pathophysiology of hidrocystomas is unclear, though many theories exist. The occlusion or blockage of the sweat duct apparatus, which results in the retention of sweat, and a dilated cystic structure, are considered to be plausible causes¹⁴. The apocrine glands are most frequent in the axilla, external auditory canal, eyelids, and on the nipple, and it is not surprising that most apocrine hidrocystomas occur within the head and neck region³. However, involvement of genitalia is extremely rare, and to date twelve cases have been reported (Table 1)³⁻¹³.

Clinically, Apocrine hidrocystoma usually occurs singly as a unilocular or multilocular, dome-shaped translucent cyst, that is either flesh colored or bluish black and ranges from a few millimeters to more than 1 cm in diameter³. Histopathologically, apocrine hidrocystoma appears as unilocular or multilocular cysts composed of an inner layer of single or double layer of secretory columnar epithelium with decapitation secretion lying above an outer myoepithelial cell layer¹⁵. Immunohistochemically, apocrine hidrocystoma is positive for CK7, CK18 and gross cystic disease fluid protein 15 in the inner layer of epithelium, and alpha-smooth muscle actin and p63 in the outer myoepithelial cells^{1,16}.

For cystic lesions in genitalia, the differential diagnosis also includes congenital cysts of the median rapheae, eccrine

Fig. 2. Microscopic feature of apocrine hidrocystoma. (A) A cystic lesion in dermis is lined by (B) double layers of epithelial cells: an inner layer of cuboidal cells (black arrows) with apical snout (black arrowheads) and an outer layer of flattened cells (white arrows) with relatively small dark nucleus. The inner cells show cytoplasmic stains for (C) cytokeratin (CK) 7 and outer cells show nuclear stains for (D) p63, respectively. (E) CK20 and (F) carcinoembryonic antigen (CEA) are negative in both cells. (A) H&E, ×100, (B) H&E, ×400, (C) CK7, ×400, (D) p63, ×400, (E) CK20, ×400, (F) CEA, ×400.

Table 1. Summary of clinicopathological characteristics of previous cases of apocrine hidrocystomas involving the genitalia

Author	Age (yr)	Sex	Location	Clinical feature	Number/size	Immunohistochemistry finding
Ahmed et al. ⁴	29	Male	Prepuce	A small cyst	1/1 cm	PAS(+), D-PAS(+), PTAH(–), Heidenhain's iron haematoxylin(–)
	56	Male	Prepuce	A cyst	1/–	PAS(+), D-PAS(+), PTAH(–), Heidenhain's iron haematoxylin(–)
Powell et al. ⁵	23	Male	Penile shaft	Flesh colored with a slight bluish hue	1/0.4×0.5 cm	PAS(+), D-PAS(+)
de Dulanto et al. ⁶	49	Male	Prepuce	Hard tumor covered with normal skin	1/3 cm	PAS(+), D-PAS(+), Red with alcian blue staining
Flessati et al. ⁷	8	Male	Scrotum	Numerous brownish cyst formation along the midline	4/0.3~1.0 cm	PAS(+)
Glusac et al. ⁸	34	Female	Labium major	Freely mobile, fluctuant, dermal nodule	1/3.0 cm	D-PAS(+), GCDPF-15(+), CEA(–), factor XIIIa(–), Blue granules with Prussian blue staining
Mataix et al. ⁹	35	Male	Penile shaft	Translucent nodular lesion	1/1.5 cm	NA
Samplaski et al. ³	6	Male	Glans penis	Painless, glanular cystic mass	1/3×3 mm	NA
Liu et al. ¹⁰	25	Male	Penile shaft	Dome-shaped translucent nodule	1/1.2 cm	P63(+)
López et al. ¹¹	40	Male	Penile shaft	Painless, well-defined, serpiginous mass	1/3×0.5 cm	NA
Park et al. ¹²	4	Male	Scrotum	Multiple linear arranged skin-coloured papules	NA	PAS(+)
Taylor et al. ¹³	16	Male	Ventral urethral orifice	Wide bluish translucent cyst	1/0.8 cm	D-PAS(+), GCDPF-15(+), p63(+), S-100(–)
Our case	39	Male	Penile shaft	Serpiginous cyst	1/4×0.3 cm	CK7(+), p63(+), CK20(–), CEA(–)

PAS: periodic acid-Schiff stain, D-PAS: diastase resistant-periodic acid-Schiff, PTAH: phosphotungstic acid haematoxylin, GCDPF-15: gross cystic disease fluid protein 15, CEA: carcinoembryonic antigen, CK: cytokeratin, NA: not assessed.

hidrocystoma, sclerosing lymphangitis, and acquired lymphangioma⁹. Especially, differentiating apocrine hidrocystoma from median raphe cyst (MRC) could be difficult. In the past, some reports of apocrine hidrocystoma on the penis represented MRC¹⁷. However, according to histopathological finding, the lining entirely composed of pseudostratified columnar epithelium with no signs of decapitation secretion^{3,18}. Immunohistochemically, CEA is expressed in columnar cells presenting in the superficial layer of pseudostratified epithelium¹⁹. The presence of myoepithelial cells and decapitation secretion in apocrine hidrocystoma are important in differentiating those two diseases¹⁷. In our case, we conclude that apocrine hidrocystoma is more appropriate than MRC because CEA was negative and p63, myoepithelial marker, was positive. On the other hand, discrimination from eccrine hidrocystoma is also important. Eccrine hidrocystomas are true retention cysts that histologically are covered by two layers of cuboidal or flattened cells without myoepithelial cells and decapitation secretion. The immunohistochemical markers that are positive in apocrine hidrocystoma are all negative. Clinically, eccrine hidrocystoma can be seen as multiple lesions

of the face area and have seasonal variation^{1,3}. Sclerosing lymphangitis and acquired lymphangioma are also distinct histologically. Sclerosing lymphangitis shows hypertrophy of lymphatic channels with or without perilymphatic cell infiltration or thrombi. In case of acquired lymphangioma, numerous dilated lymphatic vessels are observed in upper dermis⁹.

The treatment for apocrine hidrocystoma is excision with narrow margins because of the benign nature of the lesion³. Other methods that have shown success include carbon dioxide laser vaporization and laser treatment¹⁵. In conclusion, although genital apocrine hidrocystoma is rare, we should be considered in the differential diagnosis of a genital cystic lesion. This paper emphasizes the importance of considering this diagnosis when evaluating a cystic lesion in genitalia.

ACKNOWLEDGMENT

We thank the patient for granting permission to publish this information.

CONFLICTS OF INTEREST

The authors have nothing to disclose.

ORCID

Jeong Won Jo, <https://orcid.org/0000-0002-4324-940X>

Jung Wook Yang, <https://orcid.org/0000-0002-9698-3667>

Do Seon Jeong, <https://orcid.org/0000-0002-5857-5670>

REFERENCES

1. Kikuchi K, Fukunaga S, Inoue H, Miyazaki Y, Ide F, Kusama K. Apocrine hidrocystoma of the lower lip: a case report and literature review. *Head Neck Pathol* 2014;8:117-121.
2. Mehregan AH. Apocrine cystadenoma; a clinicopathologic study with special reference to the pigmented variety. *Arch Dermatol* 1964;90:274-279.
3. Samplaski MK, Somani N, Palmer JS. Apocrine hidrocystoma on glans penis of a child. *Urology* 2009;73:800-801.
4. Ahmed A, Jones AW. Apocrine cystadenoma. A report of two cases occurring on the prepuce. *Br J Dermatol* 1969;81:899-901.
5. Powell RF, Palmer CH, Smith EB. Apocrine cystadenoma of the penile shaft. *Arch Dermatol* 1977;113:1250-1251.
6. de Dulanto F, Armijo-Moreno M, Camacho Martinez F. [Nodular hidradenoma (apocrine cystadenoma) of the penis]. *Ann Dermatol Syphiligr (Paris)* 1973;100:417-422. French.
7. Flessati P, Camoglio FN, Bianchi S, Fasoli L, Menghi A. [An apocrine hidrocystoma of the scrotum. A case report]. *Minerva Chir* 1999;54:87-89. Italian.
8. Glusac EJ, Hendrickson MS, Smoller BR. Apocrine cystadenoma of the vulva. *J Am Acad Dermatol* 1994;31(3 Pt 1):498-499.
9. Mataix J, Bañuls J, Blanes M, Pastor N, Betlloch I. Translucent nodular lesion of the penis. Apocrine hidrocystoma of the penis. *Arch Dermatol* 2006;142:1221-1226.
10. Liu YS, Wang JS, Li TS. A 25-year-old man with a dome-shaped translucent nodule on the glans penis. *Dermatol Sin* 2010;28:177-178.
11. López V, Alonso V, Jordá E, Santonja N. Apocrine hidrocystoma on the penis of a 40-year-old man. *Int J Dermatol* 2013;52:502-504.
12. Park J, Kim I, Jang HC, Chae IS, Park K, Kim Y, et al. Linear skin-coloured papules on scrotum: a quiz. Apocrine hidrocystoma. *Acta Derm Venereol* 2015;95:762-763.
13. Taylor D, Juhl ME, Krunic AL, Sidiropoulos M, Gerami P. Apocrine hidrocystoma of the urethral meatus: a case report. *Acta Derm Venereol* 2015;95:376-377.
14. Vani D, T R D, H B S, M B, Kumar HR, Ravikumar V. Multiple apocrine hidrocystomas: a case report. *J Clin Diagn Res* 2013;7:171-172.
15. Sarabi K, Khachemoune A. Hidrocystomas—a brief review. *MedGenMed* 2006;8:57.
16. Ohnishi T, Watanabe S. Immunohistochemical analysis of cytokeratin expression in apocrine cystadenoma or hidrocystoma. *J Cutan Pathol* 1999;26:295-300.
17. Persec Z, Persec J, Sovic T, Rako D, Bacalja J, Hrgovic Z, et al. Median raphe cyst—clinical report and immunohistochemical analysis. *J Clin Exp Dermatol Res* 2013;S6:011.
18. Deliktas H, Sahin H, Celik OI, Erdogan O. Median raphe cyst of the penis. *Urol J* 2015;12:2287-2288.
19. Dini M, Baroni G, Colafranceschi M. Median raphe cyst of the penis: a report of two cases with immunohistochemical investigation. *Am J Dermatopathol* 2001;23:320-324.