

Histological study of eyelid hidrocystoma: A clinical case

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Abstract

Hidrocystomas are benign lesions with frequent localization on the eyelids and common appearance as isolated translucent cystic lesions. The multiple and bilateral localization has been associated with systemic diseases or epidermodysplasia. The histological and ultrastructural study contributes to the distinction in eccrine or apocrine. In this clinical case, multiple bilateral skin lesions of the eyelids were removed surgically from a 55-year-old male patient with free medical history. The lesions were fully removed surgically without rupture of the cysts. The obtained tissues were fixed and prepared for an anatomical pathology study as well as observation under the electron microscope. The histological study revealed cystic lesions with low cuboid epithelium and presence of cells and collagen fibrils, while the ultrastructural study also revealed the presence of apoptotic and secretory cells as well as vacuolization of the cytoplasm. No relapse of the lesions was obtained. In conclusion, the histological and ultrastructural study of multiple hidrocystomas can highlight special features that help to better classify and diagnose eccrine or apocrine hidrocystomas, while surgical removal without rupture of the cyst can prevent reappearance.

Keywords

Hidrocystomas, eyelid, benign lesions, ultrastructural study, histological study

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Introduction

The eyelid is quite complex in its structure despite its very small surface area and presents a large number of benign and pathological lesions. After chalazia, the most prevailing benign lesions of the eyelids are papillomas, seborrheic keratosis, melanocytic nevi, and cysts.^{1,2} The last encompasses epidermoid, hybrid and Meibomian keratinous cysts, as well as ectasias concerning the ducts of the eccrine and apocrine glands (hidrocystomas) and mildly proliferative ectasias of the follicular infundibulum (dilated pore of Winer).^{3–10}

Hidrocystomas are rare cystic lesions that form benign tumors of the sweat glands.^{11–14} They are found in the skin and commonly appeared in the eyelids.^{1,15–18} They can be either eccrine or apocrine and are often found on the head, neck, and the trunk regions. They have also been reported to occur on the ears, scalp, chest, and shoulders. Rarely, they occur on the penis, the vulva, in the axillae, and in the anal region.^{12,14,19,20} In the orbit, they are extremely rare with only a few cases reported, mainly occurring in children or in adults after trauma.¹² The multiple and bilateral

eyelids' hidrocystomas can signal a systemic disease or a genodermatosis.^{21–24} Unusual blepharoptosis, dysmotility, globe displacement, and/or proptosis can be variable manifestations of an orbital hidrocystoma.^{12,25}

According to Maeng et al.,¹³ hidrocystomas may be more common in female, African American, Caucasian, and Hispanic patients. They are also presented most commonly in adults in their mid-50s, while the recurrence of the lesions

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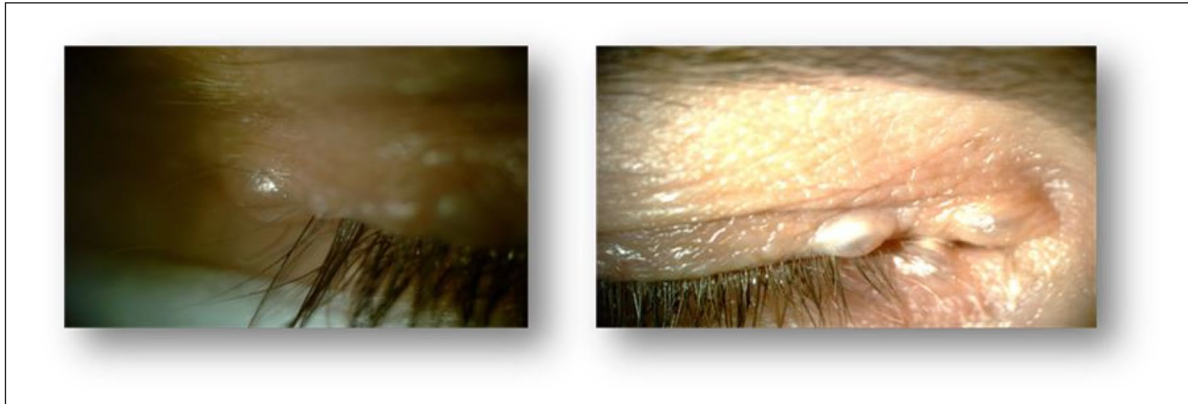


Figure 1. Multiple bilateral skin lesions of the eyelids before the surgical excision.

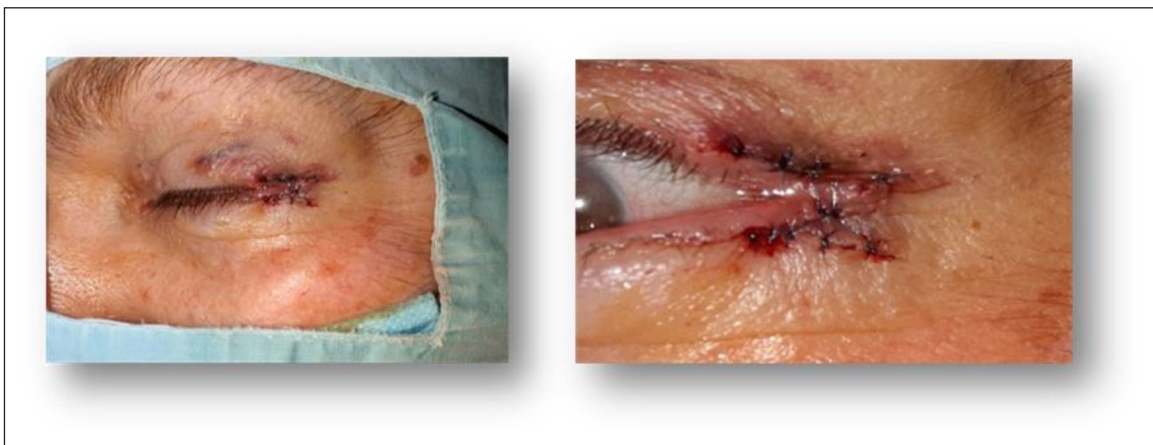


Figure 2. Multiple bilateral skin lesions of the eyelids after the surgical excision.

may be more common in the Hispanic and Caucasian population. However, the overall recurrence rate is low with current excisional techniques.

Eccrine hidrocystomas, also known as “sudoriferous or sweat gland cysts,” are thought to be ductal retention cysts and appear as multiple (the Robinson) or solitary (the Smith type, which is the most prevalent solitary type), small and tense thin-walled, dome-shaped nodules located on the eyelids, ranging from 1 to 6 mm in diameter. The overlying skin is shiny and smooth, and the cyst usually is translucent, fluid filled, and typically has an amber, brown, or bluish tint. They do not involve the eyelid margin which differentiates it from the apocrine type along with a lighter color presentation. They are predominantly found in adult females.^{9,11,12,14}

Apocrine hidrocystomas, also known as “cystadenoma,” usually appear as a solitary, translucent cyst on the face, head, and neck as well as sometimes at the eyelid margin, especially near the inner canthus but less likely than eccrine lesion to occur at periorbital regions. It arises from the proliferation of apocrine glands and is usually small (less than 1 cm in diameter) with shiny, smooth overlying skin and

filled with clear or milky fluid and with a variety of colors ranging from flesh-colored to blue-black with the bluish coloration attributed to the Tyndall effect. Unlike the eccrine variety, these lesions are thought to be proliferative in origin and do not increase in size in hot weather.^{9,11,12,14}

In this case report, we present a patient with multiple bilateral hidrocystomas of the eyelids that were successfully treated with simple surgical excision without rupture of the cysts while transmission electron microscopy established the final diagnosis determining the subsequent management of the patient.

Case report

This is a clinical case of a 55-year-old male patient with multiple bilateral skin lesions of the eyelids and free medical history (Figure 1). The lesions were fully removed surgically without rupture of the cysts (Figure 2). The obtained tissues were fixed and prepared for an anatomical pathology study as well as observation under the transmission electron microscope. Written informed consent was obtained retrospectively from the patient for anonymized patient information to

be published in this article. The study was conducted in accordance with the Declaration of Helsinki.

At presentation, a comprehensive ophthalmic assessment was performed. Ophthalmic examination was unremarkable.

Treatment was performed in the operating theater under topical (eyedrops of proxymetacaine hydrochloride 0.5%) and local anesthesia (injection of lidocaine in the sites of eyelids' lesions) as well as sterile conditions. Periocular region was cleaned with povidone-iodine and simple surgical excision of the lesions was performed. The cystic lesions were removed carefully in order to prevent any rupture of the cysts and leakage of its content. Prolene stitches 7-0 were applied for the restoration of the eyelids' anatomy. The patient received antibiotic ointment in the area of the surgical removal for the prevention of inflammation. Stitches were removed 10 days later.

Follow-ups were performed at 1, 6, and 12 months after the surgery. No recrudescence of the lesions was observed.

The obtained tissues were fixed and prepared for an anatomical pathology study as well as observation under a transmission electron microscope (TEM JEOL 1011 in 80 kV (JEOL-Tokyo, Tokyo, Japan)).

The anatomical pathology study revealed hidrocystomas in all specimens. The histological study with eosin–hematoxylin staining showed cells and fibrils within the cyst, the presence of simple cuboidal epithelium on the cyst's wall, stratified columnar conjunctival epithelium around the cyst's wall, and vacuolization in the intercellular space (Figures 3–5) indicating an eccrine hidrocystoma. For the establishment and accuracy of the diagnosis, ultrastructural study was performed. The ultrastructural study showed some cells in apoptosis with nucleus showing condensation of chromatin as well as some cells with secretory characteristics. The presence of vacuoles inside the cytoplasm, collagen fibrils, fine granular cytoplasmic material, and vacuolization in the intercellular space were also observed. The presence of bacteria was also detected (Figures 6–8).

Discussion

Eccrine and apocrine hidrocystomas of the eyelids, and those located elsewhere in the skin,^{3,19,20,26} have both been described as single (Smith–Chernosky type) or multiple (Robinson type, more consistently eccrine) papulonodules; unilocular and multilocular (same as unicameral and multicameral) or confluent; smooth, shiny, skin-colored, translucent, milky (sometimes with a whitish or yellow upper polar cap of lipidic material in presumed apocrine cysts), amber, brown or bluish; and poorly vascularized to nonvascularized, compressible, and exacerbated by heat exposure (allegedly eccrine).²⁷

The presence of multiple apocrine hidrocystomas may be a marker of two rare inherited disorders: the Schopf–Schulz–Passarge syndrome and the Goltz–Gorlin syndrome. The Schopf–Schulz–Passarge syndrome is a rare autosomal

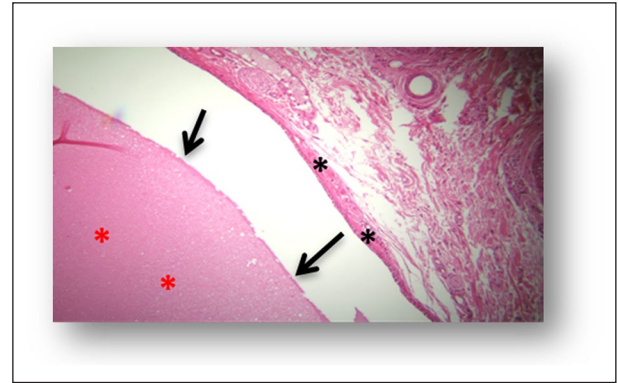


Figure 3. Cells inside the cyst (*). Simple cuboidal epithelium on the cyst's wall (↑). Stratified columnar conjunctival epithelium (*). EH×40.

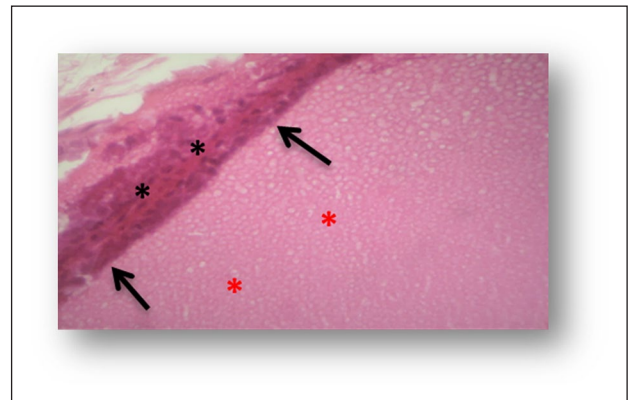


Figure 4. Cells and fibrils within the cyst (*). Simple cuboidal epithelium on the cyst's wall (↑). Stratified columnar conjunctival epithelium (*). EH×160.

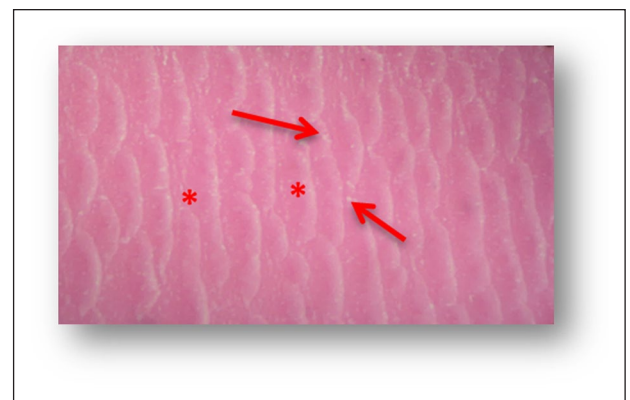


Figure 5. Cells and fibrils within the cyst (*). Vacuolization in the intercellular space (↑). EH×400.

recessive form characterized by multiple bilateral eyelid marginal apocrine hidrocystomas with concomitant ectodermal dysplasias causing anhidrosis, palmar hyperkeratosis, hypodontia, hypotrichosis, and hypoplastic nails.²³

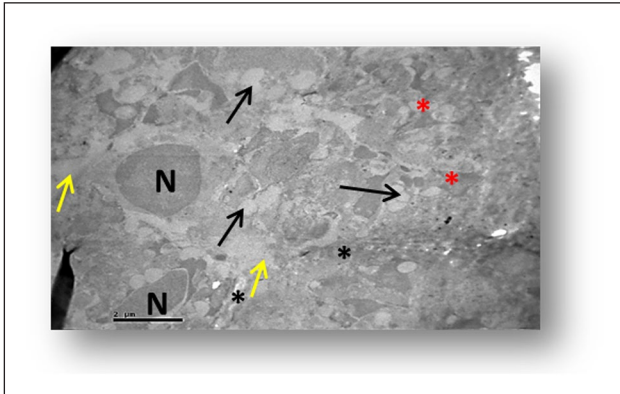


Figure 6. Cell nucleus showing condensation of chromatin (N). Vacuoles inside the cytoplasm (↑). Collagen fibrils (*). Fine granular cytoplasmic material (*). Vacuolization in the intercellular space (↑).

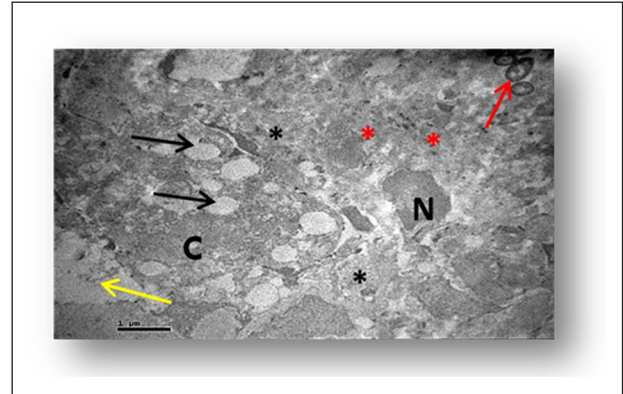


Figure 8. Cell with secretory characteristics (C). Cell nucleus in apoptosis (N). Vacuoles inside the cytoplasm (↑). Collagen fibrils (*). Fine granular cytoplasmic material (*). Vacuolization in the intercellular space (↑). Presence of bacteria (↑).

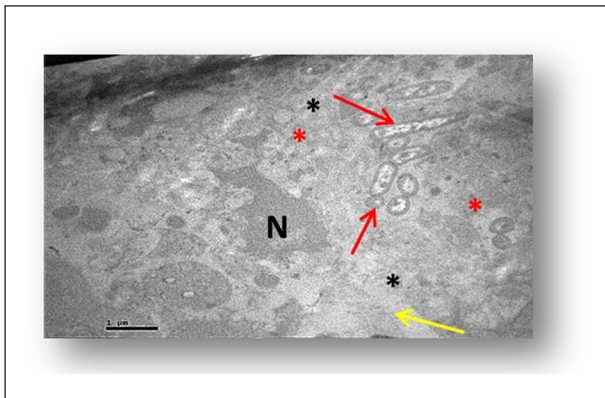


Figure 7. Cell nucleus in apoptosis (N). Collagen fibrils (*). Fine granular cytoplasmic material (*). Vacuolization in the intercellular space (↑). Presence of bacteria (↑).

The Goltz–Gorlin syndrome is sporadic in nature in most cases, but it is assumed to have an X-linked dominance mode of inheritance, as it occurs more commonly in women (male fetuses die in utero). The disorder, also called focal dermal hypoplasia, is characterized by its broad spectrum of mesoectodermal defects associated with the skin as well as with the eyes, skeletal system, and teeth. Reported cutaneous associations include linear or reticulated atrophic hypopigmented or hyperpigmented skin lesions, papillomas, lipomatous nodules, and periocular multiple hidrocystomas.²⁴ Graves' disease has also been associated with multiple eccrine hidrocystomas which disappear after treatment of hyperthyroidism. The cause is possibly the hyperhidrosis, which is present in hyperthyroid patients.^{28,29} Our patient did not have any features suggestive of either disorder.

In the largest reported series, 5504 cases of eyelid skin tumors were examined by Deprez et al. Tumors with eccrine and apocrine gland features were the most frequent adnexal tumors and accounted for 7%. The most common type was

hidrocystoma, 326 in number (5.9%) located equally in the upper and lower eyelids.² In another review comprising 867 biopsied eye lesions, 691 were benign.³⁰ Among them, 73 (10.5%) were hidrocystomas.³¹ In one more series of 2529 eyelid lesion excisions (1993–2002), the 91.5% were histopathologically benign.³² None of the above distinguished the eccrine and apocrine hidrocystomas.

The treatment of these lesions entails a variety of surgical techniques with the majority typically providing only temporary relief because the cysts usually refill with fluid within 2 to 6 weeks. Lesion puncture or incision followed by drainage, cauterization, and electrodesiccation of the cyst wall as well as surgical excision associated with electrodesiccation are only some of the recommended methods in order to prevent recurrence of the lesions. In addition, application of carbon dioxide laser vaporization, trichloroacetic acid, and hypertonic glucose has also been reported.³³

Histopathologically, hidrocystomas exhibiting a deeply eosinophilic low cuboidal to columnar cellular lining with oval to round basal nuclei and apical cytoplasmic decapitations have conventionally been designated as apocrine. In addition, it demonstrates a multilocular (multicameral) architecture, an artifactual splaying off into the lumen of the inner columnar cells that dehisced from the underlying basal layer, eosinophilic secretions in the lumen which can become organized into spheroidal bodies sometimes called Liesegang rings, a thick fibrous wall outside of the cyst's epithelial lining that is easily distinguishable from the surrounding reticular dermis with Masson's trichrome staining as well as nearby portions of an apocrine gland. In contrast, cysts lined by a low cuboidal or flattened epithelium with paler cytoplasm, no snouts, and a watery secretion without cytoplasmic contributions have been classified as eccrine. All hidrocystomas are bereft of goblet cells, which separates them from conjunctival cysts.^{2,11,15,25,27,32,34,35} Regarding our patient, the eosin–hematoxylin staining showed cells and

fibrils within the cyst, the presence of simple cuboidal epithelium on the cyst's wall, stratified columnar conjunctival epithelium around the cyst's wall as well as vacuolization in the intercellular space indicating an eccrine hidrocystoma. For the establishment and accuracy of the diagnosis, ultrastructural study was performed.

In the literature, the observation by electron microscopy of myoepithelial cells in rare variants of cutaneous sweat gland tumors has been a valuable aid in distinguishing between apocrine and eccrine solid lesions, but has enjoyed only a limited utility. Nonophthalmic eccrine hidrocystomas examined ultrastructurally have lacked this feature. While currently not routinely used, enzyme histochemistry has been applied to determine either eccrine or apocrine differentiation: identification of succinic dehydrogenase, amylophosphorylase, and leucine aminopeptidase favored the former, whereas acid phosphatase, -glucuronidase, and indoxyl esterase favored the latter. Alkaline phosphatase has been employed for the identification of myoepithelium. This methodology has been found to be less insightful when applied to hidrocystomas.^{11,34} The ultrastructural study of our clinical case showed some cells in apoptosis with nucleus showing condensation of chromatin as well as some cells with secretory characteristics. The presence of vacuoles inside the cytoplasm, collagen fibrils, fine granular cytoplasmic material, and vacuolization in the intercellular space were also observed. However, myoepithelial cells were not observed in any sample indicating the diagnosis of eccrine hidrocystomas. Conclusively, in addition to the absence of myoepithelial cells, our study highlighted the presence of apoptotic cells and vacuolization which has not been reported before, contributing to the existing literature and proving that ultrastructural study can reveal further characteristics. However, a series of cases certainly could provide more information regarding the importance and the variety of ultrastructural findings. So this could be a possible future extension of our study.

Conclusion

In hidrocystomas, the complete surgical resection to healthy limits with complete removal of the cyst's wall without its rupture can prevent its recurrence, which may explain the increased recurrence rate in cases of ruptured cysts. The ultrastructural study can be crucial for the establishment of the final diagnosis between eccrine and apocrine hidrocystomas, exhibiting further characteristics, in order to predict the possible clinical outcome of these patients and contribute to the existing literature, especially in cases where histopathological study cannot always lead to a certain diagnosis.

Author contributions

S.K., F.A., T.P., A.S., and E.F. contributed to conceptualization; S.K., G.F., P.A., T.P., A.S., and E.F. contributed to methodology; S.K., G.F., V.K., F.A., T.P., A.S., and E.F. contributed

to validation; S.K., G.F., V.K., T.P., A.S., and E.F. contributed to formal analysis; S.K., G.F., V.K., F.A., T.P., A.S., and E.F. contributed to investigation; S.K., G.F., and V.K. contributed to writing—original draft preparation; S.K., G.F., and V.K. contributed to writing—review & editing; S.K., G.F., F.A., T.P., A.S., and E.F. contributed to visualization; S.K., T.P., A.S., and E.F. contributed to supervision; S.K. contributed to project administration.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

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Informed consent

Written informed consent was obtained retrospectively from the patient for anonymized patient information to be published in this article.

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