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Maxillofacial Surgery

A Rare Case of Eccrine Hidrocystoma of The Eyelid: Case Report and Literature Review

Abdelghafour Jaifi^{1*}, Meriem El Fatihi¹, Alban Déo Christian OPANGO¹, Fadoua Fettal¹, Hamza El Yachkouri¹, Zakaria Aziz¹, Nadia Mansouri¹

¹Maxillofacial Surgery Department, University Hospital Mohamed VI of Marrakech, Morocco

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*Corresponding author: Abdelghafour Jaifi

Maxillofacial Surgery Department, University Hospital Mohamed VI of Marrakech, Morocco

Abstract Case Report

Eccrine hidrocystomas, is a rare cystic lesion of unknown origin linked to sweat retention, First described in 1893 and often present as small, translucent cysts, they derive from eccrine glands. While usually on the face, atypical locations are possible and can be single or multiple. Histology is crucial for diagnosis, revealing retention cysts involving intact eccrine glands. Differential diagnoses encompass various palpebral cystic tumors. Surgical treatment is recommended for significant lesions, while innovative approaches, including carbon dioxide laser and trichloroacetic acid injection, prove effective for smaller ones. Despite the potential for dyschromic sequelae, botulinum toxin type a injections offer lasting efficacy. No risk of malignant transformation is reported, but recurrence is possible.

Keywords: Hidrocystomas, Eccrine, Histology, Differential diagnoses, Surgery, Botulinum toxin.

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Introduction

Eccrine hidrocystomas, is a rare benign cystic lesion associated with unknown sweat retention origins, pose a dermatological challenge. First documented in 1893, these cysts, originating from eccrine or Moll glands, exhibit diverse forms: single or multiple, isolated or associated, primarily on the face. Clinically, they manifest as small, translucent cysts, prompting histological examination for precise diagnosis (Boukhlouf & Pr, n.d.; Fiani *et al.*, 2018). The primary aim of this article is to add more cases of this rare phenomenon to literature and provide a descriptive review.

CASE REPORT

A 54-year-old female patient with no particular pathological history, phototype III, consulted for a right

lower palpebral nodular lesion in the external paracanthal region, evolving for 3 years and progressively increasing in size up to 6 mm in diameter, translucent, non-inflammatory and non-painful and not bleeding on contact with a mobile implant base without intraorbital extension or oculomotor signs (Figure 1).

A spindle-shaped surgical excision, oriented in the crow's feet wrinkles, was performed to minimize scarring (Figure 2).

Anatomopathological examination of our patient's palpebral lesion revealed that it was an eccrine hidrocystoma with no detectable signs of malignancy (Figure 3).

Post-operative follow-up was straightforward, with a hidden scar and no sign of recurrence after 6 months' follow-up (Figure 4).



Figure 1: Right lower palpebral lesion (a) external paracanthal (b)

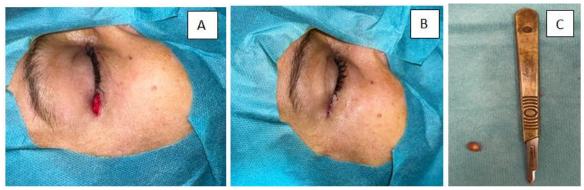


Figure 2: Spindle-shaped surgical excision (a) with separate prolene 6.0 sutures (b)

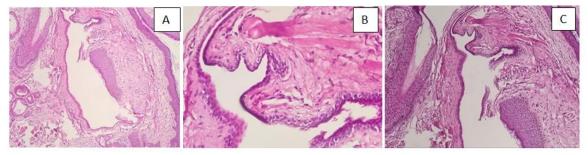


Figure 3: Eccrine hidrocystoma, showing the typical appearance of a cyst lined by a cubic bistratified coating reminiscent of a sweat duct: A: Unilocular cystic subepidermal formation (HEx20); B: Edge of cystic formation made up of a double layer of regular epithelial cells (HEx40); C: Focal squamous metaplasia of the epithelial margin of my cystic formation (HEx25)



Figure 4: Hidden scar in the wrinkles of the crow's feet

DISCUSSION

Eccrine hidrocystomas are rare benign cystic lesions of sweat retention of unknown etiology (Boukhlouf & Pr, n.d.) (Fiani *et al.*, 2018).

First described by Robinson *et al.*, in 1893 (Jakobiek, 1994), eccrine hydrocystomas, are rare cystic lesions, derive from the eccrine glands or the excretory segment of the Moll glands an.

A study of the histologic features of 5504 eyelid skin tumors diagnosed in Lausanne, Switzerland showed 84% of them to be benign, and 8% of those were hidrocystomas (eccrine and apocrine) (Deprez & Uffer, 2009).

They may be single (Smith type) or multiple (Robinson type), isolated or associated (syndromic forms) (Rodallec *et al.*, 2006) (Agharbi, 2019).

The average age at onset is 56 years, with extremes ranging from 22 to 85 with a female: male ratio of 2:1 (Maeng *et al.*, 2017).

Preferentially located on the face, especially the palpebral and zygomatic regions, but other atypical locations have been reported: thorax, shoulders and prepuce. In our case, the lesion was located on the lower eyelid (Agharbi, 2019; Fiani *et al.*, 2018).

Clinically, hidrocystomas often appear as small, translucent, shiny cysts, single or multiple, the histogenesis of which remains debatable.

Histology is the only way to avoid confusion by confirming the diagnosis, and to differentiate between eccrine and apocrine hidrocystomas (Boukhlouf & Pr, n.d.).

Histologically speaking, eccrine hidrocystomas are retention cysts often involving intact eccrine glands. Their walls are also formed by a double row of cells, but no myoepithelial cells, papillary digitations or characteristic decapitation secretions are found (Rodallec *et al.*, 2006).

There are two clinical forms: isolated and associated. Associated forms are rarer, and are described only for multiple hidrocystomas (Agharbi, 2019) (Rodallec *et al.*, 2006).

Cases of multiple eccrine hidrocystomas associated with Parkinson's disease and hyperthyroidism have been reported. (Nagai *et al.*, 1996) (Schröder & Goerdt, 1997).

More rarely, eccrine hidorcystomas are described with Shopf-Shultz Passarge syndrome, which

is associated with apocrine hidrocystomas (Nouri *et al.*, 2009).

Because of its clinical presentation and location, many differential diagnoses remain to be discussed with other palpebral cystic tumours, such as sebaceous cysts, Moll's gland adenoma, nodular basal cell carcinoma in the early stages, milium grains, syringomas and adenocarcinoma of the sweat glands. Hence the importance of systematic histological analysis of all lesions removed from the palpebral region (Rodallec *et al.*, 2006).

On the other hand, the main differential diagnosis is apocrine hydrocystoma, which are defined as cystic adenomas located in the dermis and bordered by a double cell layer, an outer layer composed of cubic myoepithelial cells and an inner layer composed of secretory columnar cells with eosinophilic cytoplasm and a characteristic apical secretory decapitation protrusion (Adenis *et al.*, 1998) (Rodallec *et al.*, 2006).

The latter can be either isolated or associated. Associated forms are rarer and described only for multiple hidrocystomas. Two hereditary syndromes have been described in the context of multiple apocrine hidrocystomas, the syndrome described by Zala *et al.*, and the Schôpf syndrome described in 1971 and reported since by several authors (Nouri *et al.*, 2009).

Hidrocystoma is generally treated surgically, essentially for single lesions that are large and/or cause functional or aesthetic damage. However, for multiple or small lesions, no more than a few millimeters in diameter and not causing deformity, carbon dioxide laser and trichloroacetic acid injection have proved effective (Fiani *et al.*, 2018).

Nevertheless, given the risk of dyschromic sequelae following the above-mentioned therapeutic modalities, botulinum toxin type A injections have already proven their lasting efficacy, while adding collateral positive effects on periorbital fine lines (Fiani *et al.*, 2018).

Based on our review of the literature, eccrine hidrocystomas, present no risk of malignant transformation, but a risk of recurrence is observed in 2.3% of cases related to incomplete excision or cyst rupture with residual cyst wall (Maeng *et al.*, 2017).

CONCLUSION

The exploration of eccrine hidrocystomas reveals a nuanced landscape of dermatological intricacies. From their historical documentation in 1893 to contemporary insights, these benign cystic lesions present as a diverse spectrum. The clinical and histological examinations underline the need for

precision in diagnosis and highlight the challenges in differentiation from apocrine hidrocystomas.

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