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Case report

Dacryops of the lacrimal gland in an elderly woman: A case report

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ABSTRACT

Introduction: Dacryops is a rare benign cystic lesion of the lacrimal gland often developing in the palpebral gland. The exact etiology of dacryops remains unclear. Diagnosis could be suspected clinically and established histopathologically. Treatment is commonly surgical.

Presentation of case: We report the case of a 75-year-old woman with past history of total conservative parotidectomy for a benign tumor 10 years ago. She presented to the Ophtalmology Department at Farhat Hached University Hospital of Sousse with a painful swelling of the left supero-external orbital angle. The patient underwent complete excision of the cyst using the conjunctival approach. Histopathological examination showed characteristic features of dacryops with foci of chronic inflammatory cell infiltrates.

Discussion: Dacryops is commonly seen in young adults or middle-aged individuals with a slight female preponderance. In some cases, cystic enlargement of dacryops can lead to mechanical ptosis, dystopia, limitation of extraocular movements, diplopia, and, in rare cases, blepharoptosis. However in this case, the patient was an elderly woman presenting a painless mass without ptosis. As in our case, excision using the conjunctival approach under microscope has excellent results without complications. Histopathological examination confirms the diagnosis and helps to rule out differential diagnoses.

Conclusion: As reported in the present case, histopathological examination confirms clinically recognized or suspected dacryops. If untreated, dacryops may lead to ptosis, proptosis and/or dystopia. Complete excision of the cyst is often curative.

1. Introduction

Dacryops is a rare benign cystic lesion of the lacrimal gland often developing in the palpebral gland. Intra-orbital location is exceptional [1]. The exact etiology of dacryops remains unclear [2,3]. Cyst formation may be due to periductal inflammation or trauma affecting the lacrimal ducts. Diagnosis could be suspected clinically and established histopathologically. Treatment is commonly surgical [1,2]. Herein, we report a rare case of dacryops, arising from the palpebral lacrimal gland in an elderly woman. This case report has been reported in line with the SCARE Criteria [4].

2. Presentation of case

A 75-year-old woman presented to the Ophtalmology Department at Farhat Hached University Hospital of Sousse with a painful swelling of the left supero-external orbital angle. Her history included total conservative parotidectomy for a benign tumor 10 years ago. On physical examination, the tumefaction was renitent and mobile. The patient underwent complete excision of the cystusing the conjunctival approach under general anesthesia. The surgical specimen was submitted to the Pathology Department for examination. Gross examination revealed a cystic mass measuring 20 x15x8 mm with a grayish white smooth outer surface. On the cut surface, the excised cyst showed a multilocular, smooth, and thin-walled cavity. Histopathological examination showed a multiloculated cyst with infoldings (Fig. 1). The cyst was lined by a non-keratinized layer of columnar to cuboidal epithelial cells showing apical cytoplasmic snouts and having an underlying layer of myoepithelial cells (Fig. 2). The cyst wall consisted of fibro-collagenous tissue, including many dilated and congested vessels, areas of hemorrhage, and foci of chronic inflammatory cell infiltrates including plasma cells, lymphocytes, and mast cells. The cyst was surrounded by foci of

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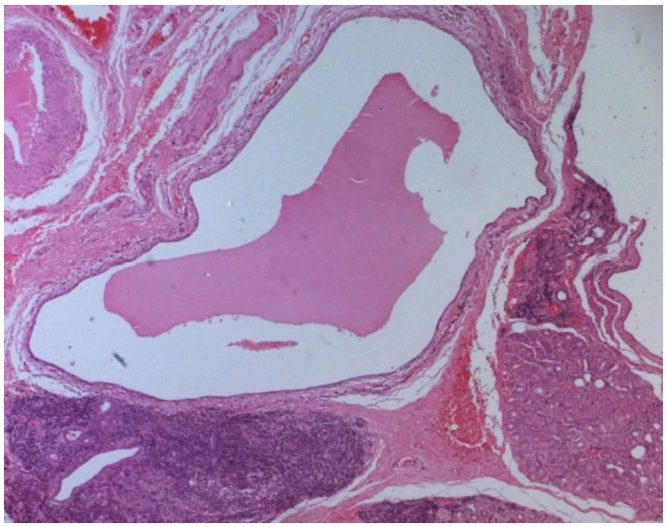

Fig. 1. Cyst lumen filled with eosinophilic secretory material and surrounded by chronic inflammatory infiltrate (hematoxylin–eosin [H&E], original magnification ×40).

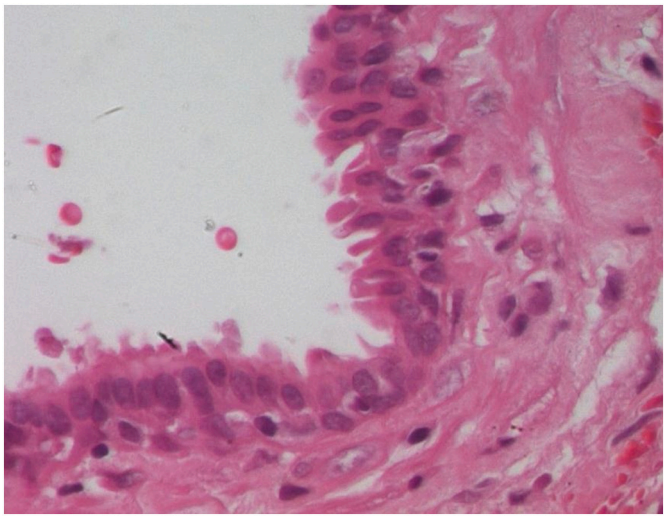

Fig. 2. Cyst lined by a double layer of columnar to cuboidal epithelium with apical cytoplasmic projections (snouts) and apocrine decapitation secretion (H&E, original magnification ×400).

inflammatory lobules of lacrimal gland. These histopathological features allowed confirming the diagnosis of dacryops arising from the accessory lacrimal gland. At six months follow-up later, no complications neither recurrence were observed.

3. Discussion

Many cystic lesions may occur in the orbital and periorbital areas. This heterogeneous group of cystic lesions is often referred to as orbital cysts [5]. Duke-Elder [6] proposed, in 1974, a classification of the lacrimal duct cyst, that was modified by Bullock et al. [7] in 1986, based on the locations of the lacrimal gland tissue: (1) palpebral lobe cysts (simple dacryops), (2) orbital lobe cysts, (3) cysts of the accessory lacrimal glands of Krause (along the conjunctival fornices) and Wolfring (along the superior tarsal), and (4) cysts of ectopic (choristomatous) lacrimal glands. Rush and Leone [6] reported an intra-orbital ectopic lacrimal gland cyst.

Dacryops is a rare benign cystic lesion, first described by Schmidt in 1803 as a ductal cyst of the primary or accessory lacrimal glands [8].

The pathogenesis of dacryops is not well established. It is suggested that a multifactorial mechanism leads to a mechanical dilation of excretory ductules. It includes chronic inflammation, infection, and IgA hypersecretion revealed by immunohistochemical studies with an osmotic effect and traumatism of the lacrimal canaliculi [9,10]. In countries where trachoma is prevalent, the incidence of accessory ductal cyst formation is likely to be significantly higher because of conjunctival inflammation and scarring [5,8].

The real incidence of dacryops is unknown. A literature review revealed that dacryops has been reported in several case reports and series. Jakobiec et al. [10] reported a series of 12 dacryops out of 128 adnexal cystic lesions, with an incidence of 3.9%. Dacryops is commonly seen in young adults or middle-aged individuals with a slight female preponderance [1,8]. The clinico-epidemiological features of patients with dacryops reported in the literature are summarized in the Table 1.

Clinically, patients usually present with unilateral, smooth, mobile, whitish and blue painless swelling in the lateral portion of the upper eyelid. Thus, diagnosis can be obvious [8,11,12]. Bilateral dacryops is found in fewer than 10% of cases [5,10]. On physical examination, dacryops appears as a conjunctival and translucent cyst which is positive to transillumination. In some cases, cystic enlargement of dacryops can lead to mechanical ptosis, dystopia, limitation of extraocular movements, diplopia, and, in rare cases, blepharoptosis [1]. In our case report, the patient presented with a painless mass without complaints of ptosis.

At ultrasonography, dacryops is characterized by a single-walled cyst with a homogeneous hypoechoic content [13]. CT scan of the orbit with injection usually shows a cystic hypodense, surrounded by a thin border referring to the cyst wall [5,9,11].

Histologically, the cyst is lined by double-layered and focally non-ciliated pseudostratified columnar epithelium. Occasional goblet cells are present in the inner cuboidal layer, surrounded by an outer myoepithelial layer [9,14]. The cyst wall consists of fibrous tissue with chronic inflammation, occasionally accompanied with areas of hemorrhage, increased vascularization, and dystrophic calcification. Lymphoid tissue is found in the subepithelial connective tissue of the cyst wall [8,11]. Many authors have identified evidence of apocrine secretion, such as apical snouts projecting in the lumen, either in the cystic component of the proliferation or in the contiguous lacrimal ducts in all cases [3,10,15]. Dacryops associated with apocrine secretion may closely resemble apocrine hidrocystoma [3]. The material inside the cyst stained negatively for mucin using the mucicarmine stain and positively using the periodic acid-Schiff stain, indicating the presence of a glycoprotein material [10,16]. The histopathological features of dacryops are characteristic and can help to rule out differential diagnoses, including dermoid cysts, mucoceles, hidrocystoma, colobomatous cysts, and parasitic cysts [2,10].

Treatment management is primarily surgical [1]. Surgery is indicated in symptomatic patients for cosmetic concerns and to prevent dystopia and amblyopia due to mechanical ptosis [5,15]. Excision using the conjunctival approach under microscope is recommended. The results of this approach are excellent and free of complications [8]. Marsupialization, puncture, and aspiration of the cyst are other less performed surgical procedures. Incomplete surgical resection can cause recurrence, fistula formation, and secondary scar defects due to the destruction of the lacrimal gland tissue [11,12].

4. Conclusion

Dacryops is an uncommon benign cyst of the lacrimal gland with unclear pathogenesis. As reported in the present case, histopathological examination confirms clinically recognized or suspected dacryops. If untreated, dacryops may lead toptosis, proptosis and/or dystopia. Complete excision of the cyst is often curative.

Table 1
Summary of the clinicoepidemiological features of patients with dacryops from a literature review.

Authors	Number of dacryops	Median age years [range]	Gender M/F	Laterality (right/left)	Median duration of symptoms (months)	Median size (mm)
Kacerovska et al. [3]	6	48 [61–81]	5/1	0/6	NA	6.5
Weatherhead et al. [8]	13	18 [8–65]	6/13	6/7	22	14
Jakobiec et al. [9]	15	55.5 [4–68]	9/6	10/7	2	NA
Galindo Ferreiro et al. [11]	23	39 [6–81.8]	14/9	11/13	NA	10.5
Lam et al. [12]	5	57 [2–70]	4/1	2/3	19.2	NA
Ozgonul et al. [13]	14	48 [21–65]	1/1	NA	NA	NA

NA = not available.

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Ethical approval

Given the nature of the article, a case report, no ethical approval was required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Guarantor

Dr. Oussama Belkacem

State

The work has been reported in line with the SCARE 2020 criteria [4].

Registration of research studies

No registration.

CRediT authorship contribution statement

Oussama Belkacem: Writing original draft and data collection.
Atef Ben Abdelkader: Revision of article and final approval of the version to be submitted.
Leila Knani: Management of case and supervision.
Dorra Chiba: Reviewing.
Moncef Mokni: Study concept and supervision.
Nihed Abdessayed: Reviewing and data analysis.

Declaration of competing interest

The authors have no conflict of interest to declare.

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