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CASE REPORT

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Idiopathic acquired dacryocystocele in an adult: case report of a rare entity

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Abstract

Background: Idiopathic acquired dacryocystocele in adults is a rare disorder of the lacrimal drainage pathway. Only 20 cases have been reported in the literature. It presents with a triad of epiphora, medial canthal swelling, and recurrent dacryocystitis. Patients presenting without recurrent dacryocystitis is quite uncommon. This presentation can be confused with secondary nasolacrimal duct obstruction and other non-inflammatory pathologies. Dacryocystorhinostomy with stenting is the treatment employed in most reported cases.

Case presentation: A 24-year-old male presented with epiphora and medial canthal swelling for 3 years. Examination revealed features of left nasolacrimal duct obstruction. Imaging was suggestive of a cystic, homogenous, non-enhancing lesion in the left lacrimal sac region without bone destruction. Findings were suggestive of dacryocystocele. Patient underwent endoscopic dacryocystorhinostomy without stenting. Postoperatively, the swelling and epiphora resolved. Patient is symptom-free on follow-up.

Conclusion: Idiopathic dacryocystocele must be considered in evaluation of a medial canthal mass with epiphora without dacryocystitis. Conservative treatment is ineffective. Endoscopic dacryocystorhinostomy is safe and effective.

Keywords: Dacryocystocele, Adults, Idiopathic, Epiphora, Dacryocystorhinostomy, Endoscopic

Introduction

Dacryocystocele is common in children and a rare entity in adults [1]. It occurs due to obstruction in the lacrimal drainage pathway. Adult-onset dacryocystocele is often secondary to trauma, CRS, post-surgery, tumor, and granulomatous diseases [2]. Primary dacryocystocele in adults is a very rare occurrence. Most cases present with epiphora and an antecedent positive history. There can be episodes of infection that result in medial canthal swelling and less commonly, lacrimal fistula with drainage of pus. Very rarely patients may present without a history of dacryocystitis. We present a rare case of acquired idiopathic dacryocystocele in an adult patient without any identifiable secondary causes successfully treated by endoscopic dacryocystorhinostomy without stenting.

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Case presentation

A 24-year-old male presented with a history of epiphora on and off for 3 years associated with occasional swelling below the medial canthus region. There was no history of pain, trauma, pus discharge, skin discoloration, or nasal complaints. No history of similar complaints during childhood, previous surgery, or associated comorbidities.

Examination revealed swelling in the left medial canthal region (Fig. 1). There was no tenderness, erythema, skin loss, or skin discoloration. Nasal endoscopy revealed DNS on the left and no other positive findings. The punctual examination was normal. No extravasation was observed from the puncta after pressing the swelling. Probing was done and there was a hard stop suggesting obstruction of the nasolacrimal duct. The lacrimal syringe was suggestive of NLDO (nasolacrimal duct obstruction). Computed tomography revealed a dilated left nasolacrimal duct with homogeneous, well defined, thin-walled mass with hyperdense attenuation involving the medial canthus of the left orbit and left

Fig. 1 Swelling in the medial canthal region (left)

nasal cavity with resulting thinning of the overlying bone and an enlarged nasolacrimal duct (Fig. 2). The rest of the nasal cavity, the paranasal sinuses, and the orbit were normal.

The patient did not improve with lacrimal massaging and conservative management. Endoscopic dacryocystorhinostomy was performed. Once the mucosal flap was raised, the bony cover of the NLD was found to be thinned out and bulging medially. A thin-walled cyst with a bluish hue was observed. A large dacryocystocele measuring 2×1.5 cm filled with straw-colored fluid was drained. Marsupialization of the cyst wall achieved (Fig. 3). Free flow was observed in irrigation. Rarefaction of the frontonasal process of the maxilla and widening of the nasolacrimal duct was observed. The culture from the fluid was sterile. Microscopic examination of the fluid did not reveal any neoplastic cells, organisms, or fungal hyphae. Histopathological examination showed the presence of epithelial and goblet cells. Post-surgery, patient received a short course of antibiotic, analgesic, nasal decongestant, and saline nasal spray. The swelling and epiphora disappeared. The patient is asymptomatic on follow-up after 6 months.

Discussion

Congenital dacryocystocele is more common than acquired dacryocystocele. Acquired dacryocystocele in adults is a rare occurrence and is usually secondary. Common conditions that can result in secondary dacryocystocele include chronic rhinosinusitis, trauma, tumors, radiation, granulomatous diseases, surgery, and acute

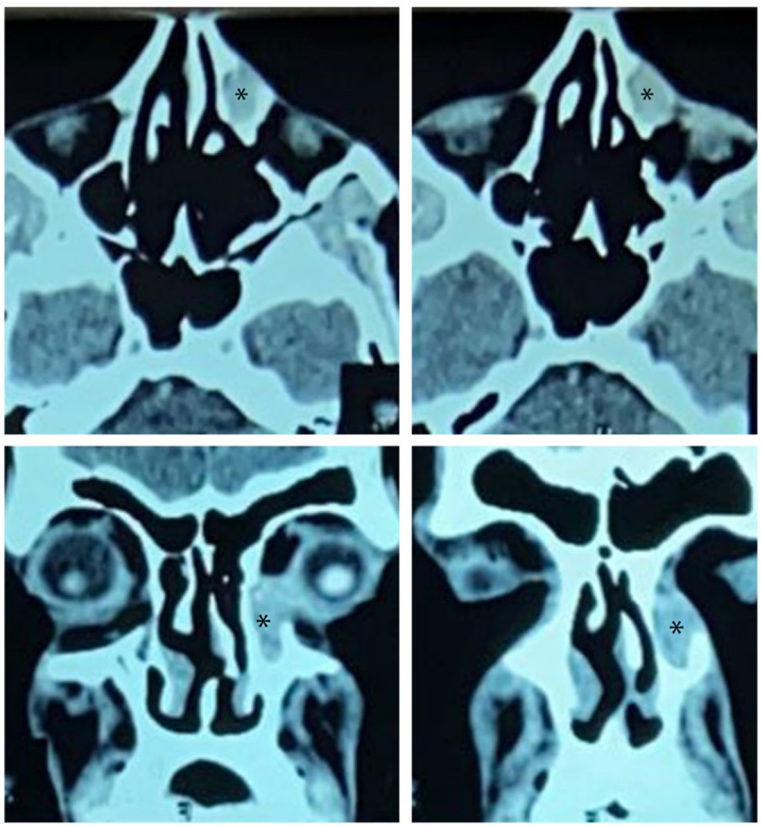

Fig. 2 CT scan showing a homogeneous lesion in the lacrimal region on the left side (asterisk)

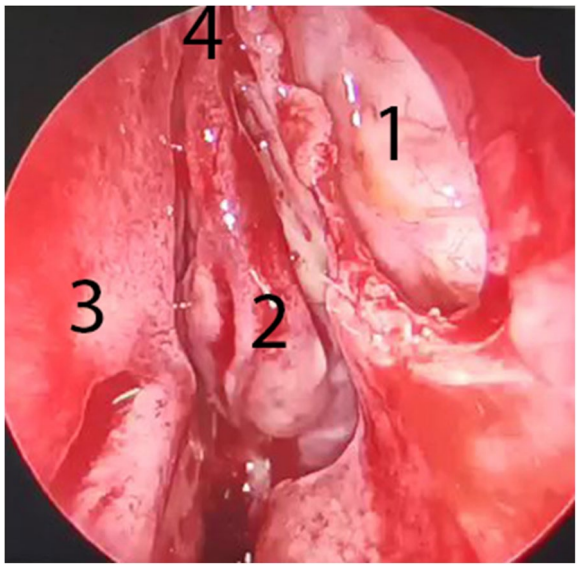

Fig. 3 Marsupialized lacrimal sac with a wide opening into the nasal cavity. 1 marsupialised sac, 2 middle turbinate, 3 nasal septum, 4 axilla of middle turbinate

invasive fungal sinusitis [3, 4]. All these conditions must be ruled out before labelling it as idiopathic [2]. The present case did not have any history or features suggestive of any such causes, and hence, it is most likely idiopathic. The pathophysiology of idiopathic acquired dacryocystocele is considered to be a long standing obstruction of nasolacrimal duct and functional blockade at the junction where common canaliculus opens into lacrimal sac resulting in dacryocystocele formation [1, 2].

Imaging is mandatory to aid in diagnosis. Both CT and magnetic resonance imaging are useful for studying the bony anatomy as well as the characteristics of the lesion. Imaging reveals a homogenous fluid-filled cyst that shows enhancement at its rim. Dacryocystoceles do not have any solid component within them [5, 6]. A forgotten foreign body that results in granuloma in the nasal cavity and lacrimal system can cause similar symptoms. Lacrimal syringing and probing help identify the site of obstruction. A hard stop while probing is due to NLD and a soft stop is due to canalicular obstruction. A soft stop is felt when the mucosal fold is encountered during examination and can be mistaken as canalicular obstruction [7]. Regurgitation of the turbid fluid during irrigation indicates NLDO and clear fluid indicates canalicular obstruction.

Dacryocystocele presents with recurrent epiphora, medial canthal swelling, and recurrent episodes of dacryocystitis [5]. A presentation without a history of dacryocystitis in acquired dacryocystocele is extremely rare, and only a few cases have been reported. Unlike its congenital counterpart, patients usually do not respond to conservative treatment. Surgical intervention is the treatment of choice [1, 2, 7–9]. Surgical drainage can be achieved via external or endoscopic approaches [10]. Success rates are similar, but with the advent of endoscopic DCR, most surgeons prefer endoscopic management, as it has major advantages over the external approach. Surgery is scarless and bleeding is less. Endoscopic DCR can be performed safely as a daycare procedure and does not injure or affect the action of the orbicularis oculi muscle, which helps in the pumping of tears [11].

Adequate opening of NLD, appropriate bone removal, prevention of postop synechiae and adhesions, proper postoperative follow-up, addressing anatomical factors such as concha bullosa, high DNS, large agger nasi, and proper placement of mucosal flaps are important to prevent recurrence. A large ethmoidal mucocele may sometimes mimic a dacryocystocele. Stenting may not be necessary in all cases. If a lacrimal stent is used, it is usually removed after 4–14 weeks [12]. The application of mitomycin C application is used in some cases [9]. Stenting was not done in our case.

On follow-up, the patient had no complaints.

Conclusion

Idiopathic acquired dacryocystocele should be considered as a differential in an adult patient presenting with epiphora, although it is rare. Patients may present without episodes of dacryocystitis. Endoscopic dacryocystorhinostomy is safe and effective and represents the most appropriate treatment.

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Authors' contributions

KK was involved in the collection of the data, literature search, writing the original draft, surgical assistance, and patient follow-up. RG was involved in the patient management, operating surgeon, follow-up, review of the final draft, and the corresponding author. Both authors have read and approved the manuscript.

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Availability of data and materials

The data collected are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written informed consent for publication of the case report was obtained from the patient.

Competing interests

The authors declare that they have no competing interests.

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