

# Accessory lacrimal gland duct cyst: 23 years of experience in the Saudi population

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**BACKGROUND AND OBJECTIVES:** Accessory lacrimal gland ductal cyst is a rare clinical entity that has been reported after trauma, infection, or conjunctival inflammation. Trachoma has been postulated as an etiologic factor for this dacryops in Saudis. We studied the prevalence, demographics, clinicopathological features and surgical approach for these lesions.

**DESIGN AND SETTING:** Retrospective study of 23 consecutive ductal cysts diagnosed clinically and proved histopathologically at King Khaled Eye Specialist Hospital (KKESH) over 23 years (1991-2014).

**PATIENTS AND METHODS:** Data on patient demographics, clinical features, surgery, and outcome were collected by chart review. The histopathologic slides were reviewed by a single pathologist.

**RESULTS:** Of 23 cases of accessory lacrimal gland ductal cysts confirmed histopathologically, 14 were males and 9 females with a median age of 38.8 years. Cysts were located in the upper eyelid in 73.9%. The commonest presentation was a painless eyelid mass in 91.3%. Excision by conjunctival incision was performed in 14 and intra-operative perforation occurred in 9. Trachomatous scarring was evident in 39.1% but did not have significance in relation to this rupture. No recurrences have been observed with a mean follow up of 34.6 months.

**CONCLUSION:** The approximate prevalence of accessory lacrimal gland dacryops in the Saudi population is 1/6800. Trachoma does not seem to be a major predisposing factor. They are more frequent in males. Their histopathological appearance is identical regardless of origin. The presence of conjunctival scarring, dacryops size, and the surgical incision type did not seem to have significant correlation with the iatrogenic rupture of the cyst. We recommend careful dissection for complete cyst excision through conjunctival approach with no expected recurrence.

Accessory lacrimal gland ductal cyst is a rare clinical entity that has been reported with chronic onset after trauma, infection, or inflammation of the conjunctiva.<sup>1</sup> In the absence of the previous conditions, the etiology may be a congenital anomaly of the excretory duct or an alteration in the composition of the secretory product.<sup>2</sup>

We report a case series of 23 patients with accessory lacrimal gland ductal cysts. To our knowledge this is the largest case series reported in the English peer-reviewed literature.

## PATIENTS AND METHODS

This is a retrospective study evaluating 23 consecu-

tive accessory lacrimal gland ductal cysts diagnosed clinically and histopathologically at King Khaled Eye Specialist Hospital, (KKESH) Riyadh, Saudi Arabia over a 23-year period. All patients underwent surgical removal and were confirmed by histopathologic features. Data on patient demographics, the underlying disease, type of surgery, contents, and outcome were collected through chart review of all patients with the clinical diagnosis of accessory lacrimal gland duct between January 1991 to December 2014. Postoperative and long-term complications were also noted. The histopathologic slides of all cases were reviewed by a single pathologist for confirmation of the diagnosis.

Demographic variables were summarized as fre-

quency (percentage) or median (interquartile range) according to the type and distribution of variables. The proportions of all categorical variables along with 95% confidence interval (CI) were presented. The Pearson coefficient and chi square (or Fisher's Exact) test were used to check for association between any two variables. Odds ratio along with 95% CI were calculated. The level of statistical significance was 0.05. Statistical analysis has been performed using software SPSS version 22.0 (IBM Corp., Armonk, New York, USA).

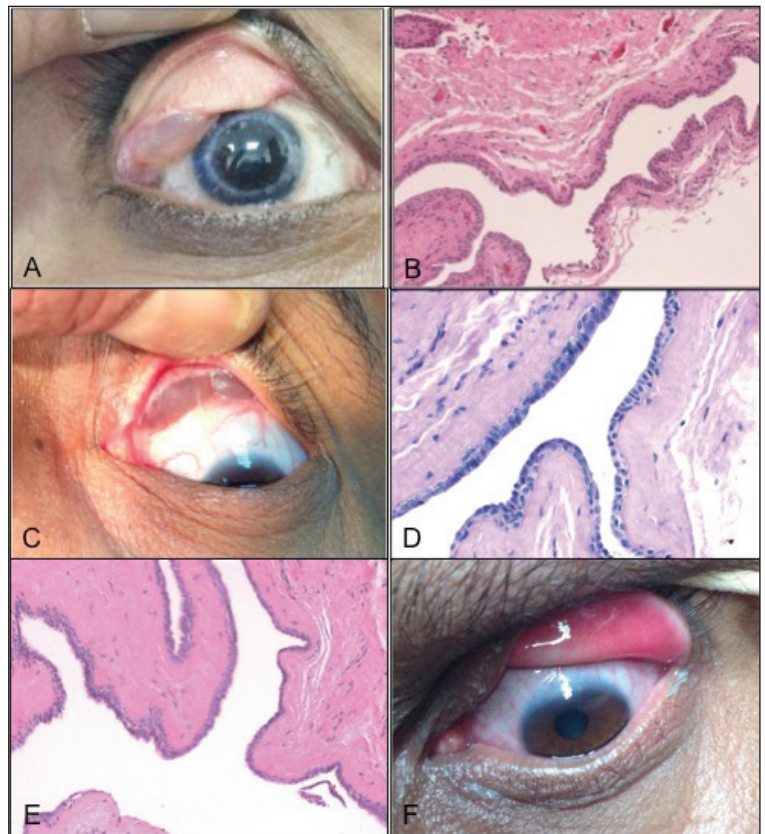
## RESULTS

A total of 23 cases of accessory lacrimal gland ductal cysts were confirmed by histopathologic examination. There were 14 (60.9%) males and 9 (39.1%) females with median age of 38.8 years (range 6.0 years to 81.8 years). The right side was involved in 15/23 (65.2%) and 8 cases only (34.8%) involved the left. The most common complaint at presentation was an eyelid mass in 21 patients (91.3%) followed by epiphora in 2 (8.7%). Cysts were located in the upper eyelid in 17 (73.9%) cases and in the lower eyelid in the rest (26.1%). The majority of cases (20; 87%) revealed clear fluid contents, while 3 (13%) had jelly-like contents within the cyst. The median largest dimension was 7 mm for cysts with clear content and 14 mm for the ones with jelly-like material. No significant association was found between the size and the contents of the cysts. The size of the cyst did not correlate with the presence of cyst rupture. In the ruptured group the median size was 13.71 mm (range, 7 mm to 25 mm), and in the non-ruptured group it was 7.77 mm (range, 2 mm to 16 mm).

Surgical excision with a conjunctival approach was performed in 14 (60.9%) cases and 9 (39.1%) cases were excised through the eyelid. Intra-operative perforation occurred in 9 (39.1%) cysts while the remaining 14 (60.9%) cysts were removed with an intact wall. The type of surgical approach did not seem to directly influence the iatrogenic rupture of the cyst during surgical excision. The mean follow up was 34.6 months (range, 0 months to 180 months) with no evidence of recurrence.

Histopathologically, the ductal cysts presented as irregular cystic cavities with a narrow lumen, lined by cuboidal to columnar epithelium, which correlated with the clinical appearance (Figure 1a-f). Apical decapitations were seen in focal areas (Figure 2a) as well as occasional goblet cells (Figure 2b). Some dacryops showed an adjacent lobule of the accessory lacrimal gland acinar cells (Figure 2c and d).

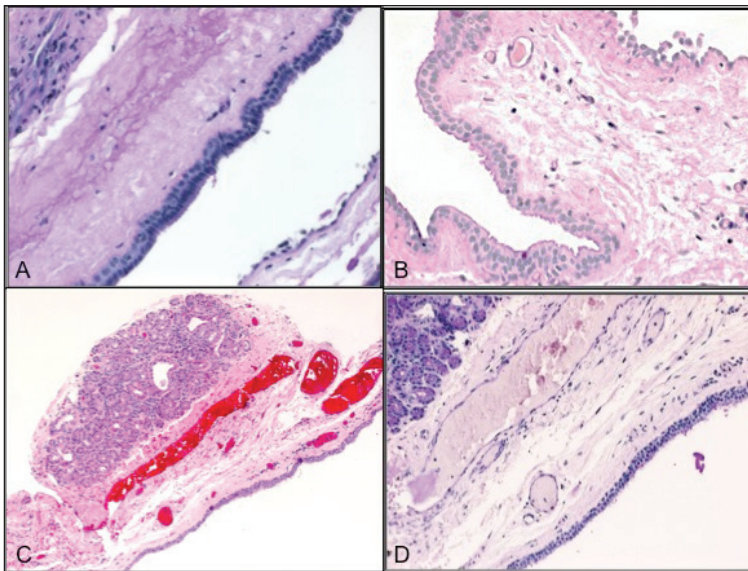
Imaging studies were ordered only in cases where the cyst size at the time of diagnosis was big and/or the



**Figure 1.** The clinical appearance of the upper lid accessory lacrimal gland ductal cyst in two patients (A & C). (B) The corresponding histopathological appearance of the dacryops in the first case, which is mostly lined by double cuboidal epithelium (original magnification  $\times 200$  hematoxylin & eosin). (D & E) Similar histopathological appearance of the epithelial lining of the dacryops in the second case (original magnification  $\times 400$  periodic acid-Schiff in D & original magnification  $\times 200$  hematoxylin & eosin in E). (F) The post-operative appearance of the second patient after excision of his upper lid dacryops with excellent result.

cyst outline was not well defined. In these cases, the imaging studies confirmed the clinical impression of a ductal cyst; however, we did not study the radiologic features in depth in our series.

As to presumed etiology, 39.1% of the cases were associated with trachomatous scarring while the remaining cases were not. No specific diagnostic test was performed in these cases since the trachoma was inactive and the clinical diagnosis of trachoma had been clearly documented in the charts and was also evident clinically. There was no association between the presence of trachomatous scarring and the cyst contents or the iatrogenic rupture. The patient age in the two groups (patients with trachoma scarring and patients without) did not correlate with the etiology of the dacryops. The surgical outcome was satisfactory in all patients with no evidence of recurrence (Figure 1f).



**Figure 2.** Gland of Wolfring dacryops with apical decapitations (original magnification  $\times 400$  periodic acid-Schiff) (A). (B) Dacryops with occasional goblet cells (original magnification  $\times 400$  periodic acid-Schiff). (C) Dacryops with adjacent glandular acini (original magnification  $\times 100$  hematoxylin & eosin). (D) The higher power appearance of the same ductal cyst with adjacent glandular acini (original magnification  $\times 200$  periodic acid-Schiff).

## DISCUSSION

The term dacryops was proposed by Schmidt in 1803 and refers to lacrimal ductal cysts of the primary or accessory lacrimal glands.<sup>3-5</sup> Lacrimal gland cysts develop insidiously in the supero-temporal cul-de-sac and were classified by Bullock as: 1) palpebral lobe cysts (simple dacryops); 2) orbital lobe cysts; 3) accessory lacrimal gland cysts of Krause and Wolfring or 4) ectopic lacrimal gland cysts.<sup>1,6,7</sup> The incidence of accessory lacrimal gland dacryops is unclear, as they have been reported only rarely.<sup>8,10</sup>

In the Saudi population, Weatherhead described 13 patients from the same institution (1984-1990) and concluded that the prevalence of Krause and Wolfring dacryops is 1/7000 (0.00014286%).<sup>3</sup> Our evaluation in a similar population following that period (1991-2014) revealed 23 cases in a total of 1562672 new patients seen in the oculoplastic out-patient clinic with a prevalence of 0.001471838% which is approximately 1/6800 in our study. This value is slightly higher and we believe that it is more representative because our patient population is larger over a longer period.<sup>3</sup>

Duran in 1883 was the first to report a subconjunctival cyst associated with accessory lacrimal glands, followed by the review of Weatherhead in 1992, then several sporadic cases were published in the reviewed literature (from 1983 to 2015) with a total of 23 cases, which is equivalent to our total number of patients.<sup>3,8-15</sup>

If we compare the demographic results in our series, accessory lacrimal gland dacryops is more frequent in males with a ratio of 14:9. This ratio is reversed in the reviewed published cases (9 males: 14 females) as summarized in **Table 1**.

Clinically, patients usually present with a painless mass at the base of the superior conjunctival cul-de-sac and the dacryops appears as a conjunctival translucent cyst. These cases are often associated with mechanical ptosis and no other ocular symptoms.<sup>11</sup> In our series, almost all our patients presented with a painless mass and none complained of ptosis.

The accessory lacrimal glands of Wolfring and Krause are responsible for basal tear secretion and differ from the major gland with respect to their location. Wolfring glands are located in and around the upper tarsal border and, to a lesser extent, in the lower tarsal border. Krause glands are located within the conjunctiva of both upper and lower fornices.<sup>14</sup> The sole duct of each accessory gland empties directly onto the adjacent conjunctival surface. The majority of the dacryops in our series (73.9%) were found in the upper lid, which is similar to what has been reported (75%). This might be explained by the fact that there are approximately 20 accessory glands of Krause in the superior fornix, and half in the inferior fornix in addition to the presence of the accessory glands of Wolfring above the superior tarsus as described above. The histopathologic appearance of the ductal cyst arising from both Wolfring and Krause glands is identical. Therefore, we were unable to differentiate between the two in terms of their origin.

As to the etiology, lacrimal gland ductal cyst is a rare clinical entity that has been reported in association with trauma, infection, or chronic conjunctival inflammation.<sup>1</sup> It has been proposed that peri-ductal inflammation or trauma would weaken the neuromuscular contractility of the lacrimal gland duct resulting in the passive dilatation of the duct and cyst formation. Conditions such as trachoma, which cause conjunctival scarring, are considered antecedents of dacryops formation.<sup>13</sup> Based on our current literature review, 15 reported cases (61%) were associated with trachoma, 2 cases were linked to other conjunctival scarring diseases and the remaining 3 had normal conjunctiva with no other pathology (**Table 1**).<sup>3,8,11-14</sup> In total, 85% of these reported cases were associated with conjunctival scarring disease. In our series, trachoma was associated in a lower percentage of patients (39.1%). This might be related to the recent decline in the incidence of trachoma in the Saudi population since the disease has been eradicated. Therefore, cysts can still occur even without conjunctival scarring from trachoma, chemical injuries or pre-

vious surgeries. Weatherhead has previously proposed this possibility.<sup>3</sup> In such cases where a history of trauma or inflammation is absent, a congenital anomaly of an excretory duct and/or alteration in the composition of the secretory products might be the cause.<sup>2,4,10,16-20</sup>

A double layer of non-ciliated columnar or cuboidal cells lines the excretory ducts of the accessory lacrimal glands of Wolfring. Occasionally goblet cells are pres-

ent within this epithelial lining. Small aggregates of lymphoid tissue might be present. Ductal cysts of the accessory lacrimal gland of Wolfring are uncommon. The anatomic clue is the location of the cyst near the superior tarsal border. The histologic clue is the double-layered epithelial lining with occasional goblet cells, apical apocrine changes and foci of lymphoid tissue.<sup>3</sup> Typical histopathologic findings would confirm these

**Table 1.** Summary of previously reported cases and our case series.

Study	Total # of cases	Case #	Gender	Age (y)	Side	Location	Size (mm)	Surgical approach	Associated disease	Fluid		
Sen DK 1967 <sup>5</sup>	1	1	f	31	LT	S	75×50	Posterior	-	Clear		
Duran JA 1983 <sup>11</sup>	1	1	m	29	LT	S	20	Posterior	Trachoma	Serum		
		1	m	8	LT	SN	25×10	Posterior		Clear		
		2	f	19	RT	IC	10×5	Posterior		Clear		
		3	f	22	LT	SC	10×5	Posterior		Clear		
		4	m	8	RT	SC	12×8	Posterior		Clear		
		5	m	15	LT	SC	12×5	Anterior		Clear		
		6	f	20	RT	IC	8×3	Posterior		Clear		
		Weatherhead RG 1992 <sup>3</sup>	13	7	m	14	LT	SN	10×4	Posterior		Clear
				8	f	65	RT	SN	15×8	Anterior		Jelly
				9	m	10	LT	SN	20×13	Anterior		Clear
				10	f	40	LT	SN	9×5	Posterior		Clear
				11	m	18	RT	SN	20×10	Anterior		Clear
				12	f	22	RT	SN	26×8	Posterior		Clear
13	f			8	RT	IC	20×10	Posterior		Clear		
Woo KI 1995 <sup>12</sup>	2	1	f	23	RT	SC	21×15	Posterior	Trachoma	Clear		
		2	f	39	LT	SC	12×15	Anterior	Stevens Jhonson	Clear		
Remulla HD 1995 <sup>13</sup>	1	1	m	63	LT	SC	34×18	Anterior	Ocular Cicatricial Pemphigoid			
O'Duffy D 1997 <sup>10</sup>	1	1	m	51	RT	S		Posterior	-	Clear		
Khoury NJ 1999 <sup>14</sup>	2	1	m	4	RT	IC	10×8	Posterior				
		2	f	37	RT	SN	25×20	Posterior				
Nakauchi K 2009 <sup>15</sup>	1	1	f	56	LT	S	18×7×13	Anterior + Cryoprobe	-	Clear		
Jastrzebski A 2012 <sup>8</sup>	1	1	f	2	LT	IC		Posterior				
		1	f	2	LT	IC		Posterior				
Lam K 2013 <sup>9</sup>	23	1	m	29	LT	ST	10×5	Posterior				
		2	m	47	RT	SN	20×25	Posterior		Jelly		

Abbreviations: m: male, f: female, LT: left, RT: right, SN: supero nasal, SC: supero central, ST: superotemporal, IN: infero nasal, IC: inferocentral, IT: inferotemporal, Anterior approach: skin incision, Posterior approach: conjunctival incision

ductal cysts. In all our cases, the histopathologic features were similar regardless of the original location, with narrow irregular lumina lined by a typical double cuboidal epithelium, areas of elongated inner epithelial cells and apical decapitations. Some cases had adjacent lobules of the accessory lacrimal gland, but none of our cases showed lymphocytic aggregates.

The differential diagnosis of upper and lower eyelid mass lesions includes tumors, foreign body granulomas, sarcoidosis, tuberculosis, and more importantly, dermoid or epidermoid cysts.<sup>14</sup> Other cystic lesions such as inclusion cyst and dermoid cyst of conjunctival origin should be easily differentiated histopathologically from ductal cysts.<sup>12</sup> Conjunctival epithelial inclusion cyst is typically lined by non-keratinizing stratified squamous epithelium possibly containing goblet cells, often with history of surgery or trauma. Dermoid cysts of conjunctival origin will also be lined by the same type of

epithelium but would show dermal appendages. Finally, lymphatic cysts would be differentiated by the appearance of endothelial-lined spaces.

Management of the accessory lacrimal ductal cysts is primarily surgical.<sup>3,5</sup> It is important to completely remove the cyst since incomplete excision or simple aspiration can result in recurrence.<sup>12</sup> To avoid iatrogenic cyst rupture, careful dissection is needed. There are two surgical approaches for the cyst removal: skin or conjunctival. The conjunctival approach might be superior because it does not pierce the skin or the levator. Simple excision of the intact cyst through a conjunctival approach should be the goal of therapy.<sup>3</sup> Failure to achieve this might result in fistula formation and recurrence. To avoid cyst rupture during surgery, it has been recommended to perform an excision of a small (1-2 mm strip) of the tarsal plate.<sup>3</sup> In our series, tarsal strip removal was not performed in any of the 23 cases.

**Table 1.** (cont.) Summary of previously reported cases and our case series.

Study	Total # of cases	Case #	Gender	Age (y)	Side	Location	Size (mm)	Surgical approach	Associated disease	Fluid
Present study		3	f	44	LT	SN	3×2	Anterior		
		4	f	81	RT	IN	3×2	Anterior		
		5	F	67	RT	SN	8×2	Anterior		
		6	m	11	RT	SN	12×5	Anterior		Jelly
		7	m	39	RT	ST	4×2	Posterior		Clear
		8	f	28	RT	SC	14×8	Posterior		Jelly
		9	m	26	RT	SN	10×4	Posterior		
		10	m	68	RT	SC		Posterior		Jelly
		11	f	6	LT	ST	9×8	Anterior		
		12	f	22	LT	SN		Posterior		
		13	m	15	LT	SN	16	Anterior		Clear
		14	m	47	LT	S		Posterior	Trachoma	Jelly
		15	m	11	RT	SN	7	Posterior		Clear
		16	f	25	RT	ST	2×1	Posterior	Trachoma	Clear
		17	m	58	RT	IC	14×6	Posterior	Trachoma	Clear
		18	m	44	LT	IC	5×3	Posterior	Trachoma	
		19	m	44	RT	SN	25×15×5	Anterior	Trachoma	
		20	m	61	RT	IC	5	Posterior	Trachoma	
		21	f	32	RT	IN	7×7	Anterior	Trachoma	
		22	m	62	RT	SN	10	Anterior	Trachoma	
		23	f	16	LT	IC	8×5	Posterior	Trachoma	

Abbreviations: m: male, ffemale, LT:left, RT:right, SN: supero nasal, SC: supero central, ST: superotemporal, IN: infero nasal, IC:infero central, IT:inferotemporal, Anterior approach:skin incision, Posterior approach:conjunctival incision

We believe that this step is not necessary, because with careful dissection, successful removal of the ductal cyst has been achieved through that approach. However, if levator surgery is needed or if there is expected shortening of the conjunctiva, the skin approach can be very useful.<sup>3</sup> Therefore, appropriate surgical methods should be tailored for each case.

In conclusion, accessory lacrimal gland ductal cysts are rare with an approximate prevalence of 1/6800 in the Saudi population. Their etiology is unclear and in our community has been associated with trachoma in only 39% of the cases. They are more frequent in males with a ratio of 14:9. They mostly involve the upper lid in 73.9% of the cases, and the majority (87%) has clear

fluid content. Their histopathologic appearance is identical regardless of origin and location. The presence of conjunctival scarring, size of the dacryops and the type of surgical incision (skin versus conjunctival) did not seem to have direct etiologic relation to the iatrogenic rupture of the cyst. Careful dissection aiming at excision of the intact cyst through a conjunctival approach is the preferred method of treatment with no expected recurrence. The small sample size of our series limits our ability to draw conclusions related to the statistical test results. However, these tests were performed to rule out a specific association or a trend. Studies with larger sample sizes and meta-analyses are recommended to support our conclusion.

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