Cyst of Accessory Lacrimal Gland

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When a patient is presented with a subconjunctival cyst, it is not only hard to reveal its true nature clinically but also easy to rupture during excision. We experienced cases with cysts of the accessory lacrimal gland in two patients with subconjunctival cysts. They had lid swelling at initial presentation and underwent surgical excision of subconjunctival cysts located in superior portion of the upper tarsal plate. The lining of these cysts composed of ductal epithelia. Biochemical analyses for serum and cystic fluid were performed in one case, in which was found high Ig A titer in the cystic fluid. These cysts seemed to originate from the duct of Wolfring's accessory lacrimal gland, considering their anatomic locations and pathologic findings. Complete removal of the cyst is important, because recurrences have been reported in cases of incomplete removal or simple aspiration.

Key words: accessory lacrimal gland, subconjunctival cyst, Wolfring’s gland

INTRODUCTION

Cysts in the duct of accessory lacrimal gland are infrequently reported. Clinically it is diagnosed as a conjunctival cyst, and only histopathologic examination can reveal its true nature. Typical pathologic findings and anatomic location help to make a definite diagnosis of the subconjunctival cyst. Therefore, through the two cases we experienced, we present clinical descriptions of cysts of accessory lacrimal gland and techniques of surgical management.

CASE REPORTS

Case 1

A 23-year-old female patient experienced painless swelling on the right upper lid which had gradually developed for 3 years. There was no history of trauma or surgery. The right upper lid showed a 2 mm ptosis with the function of levator muscle decreased to 4 mm (Fig. 1A). On evertting the upper lid, a large, tense, cystic conjunctival mass measuring 2 × 1 cm was seen above the upper border of upper tarsal plate (Fig. 2). The upper tarsal conjunctiva showed scar change. There was no evidence of proptosis or limitation of eye movement, and other ocular examinations revealed normal findings. Computed tomographic scan showed ovoid mass measuring 2.1 × 1.5 cm in centromedial portion of upper lid and showed no orbital extension or bony changes (Fig. 3).

Under local anesthesia, the cyst was excised using conjunctival approach. During excision, the cyst was ruptured and thin, watery, clear, and slightly yellow fluid gushed out. Careful excision was done after confirming the extent of the cyst by injecting gentian violet into the cystic space (Fig. 4). The upper lid was still ptotic in spite of cyst removal, so levator aponeurotic resection was performed. The conjunctiva was not sutured because
**Fig. 1.** A: The right eyelid shows swelling and ptosis preoperatively. B: It shows good cosmesis after 3 months of follow-up postoperatively.

**Fig. 2.** A large subconjunctival cyst is seen above the upper border of tarsal plate on lid eversion. Tarsal conjunctiva shows scar change.

**Fig. 3.** Computed tomographic scan shows ovoid mass in centromedial portion of anterior orbit.

**Fig. 4.** The cyst is excised using conjunctival approach.

**Fig. 5.** A cyst lined by two layered epithelium with apical changes of apocrine secretion is observed (arrow). Fibrosis is found in the cystic wall. (H & E, × 200)
of the preexistent shortening of conjunctiva in the superior fornix. Therefore, Gore-Tex® surgical membrane of 0.1 mm thickness was sutured to bulbar conjunctiva with 6-0 polyglactin suture material in order to prevent symblepharon formation.

Histopathological examination showed a cyst lined by two layers of epithelium inner columnar or cuboidal with apical changes of apocrine secretion, and slightly flattened outer cellular layer. Fibrosis was found in cystic wall. Pathological findings were consistent with those of the distension cyst of the duct of accessory lacrimal gland (Fig. 5). The Gore-Tex® surgical membrane was removed 4 weeks after operation and there was no recurrence throughout the follow-up period of 8 months (Fig. 1B).

<table>
<thead>
<tr>
<th>Table 1. Cystic fluid analysis from case 2</th>
<th>serum</th>
<th>cystic fluid</th>
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<tbody>
<tr>
<td>Sodium (135-145 mmol/l)*</td>
<td>140</td>
<td>136</td>
</tr>
<tr>
<td>Potassium (3.5-4.5 mmol/l)*</td>
<td>4.5</td>
<td>3.5</td>
</tr>
<tr>
<td>Chloride (98-110 mmol/l)*</td>
<td>109</td>
<td>108</td>
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<tr>
<td>Total protein (6.0-8.0 g/dl)*</td>
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<tr>
<td>Ig A (68-378 mg/dl)*</td>
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<tr>
<td>Ig G (694-1618 mg/dl)*</td>
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<td>2,100</td>
</tr>
<tr>
<td>Ig M (60-263 mg/dl)*</td>
<td>111</td>
<td>62</td>
</tr>
</tbody>
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*normal range in serum level

Case 2

A 39-year-old female patient noted a mass in the left upper lid about 2 years previously. She was diagnosed as having Stevens-Johnson’s syndrome 10 years ago after taking some medication which is believed to be the cause of this syndrome. There is no evidence of active inflammatory response at the

![Fig. 6. A: A round, movable mass at center of left upper lid. B: A subconjunctival cyst is seen on lid eversion. (arrowheads)](image)

![Fig. 7. A: Two layered epithelium with apocrine differentiation. (H & E, × 400) B: Dilated ductal structure with goblet cells and subepithelial lymphoid cell aggregation. (H & E, × 100)](image)
present time. The soft and movable subconjunctival mass measuring $1.2 \times 1.5$ cm was noticed at center of left upper lid (Figs. 6A,B). The conjunctiva of both eyes showed symblepharon formation in medial side and scar changes in tarsal conjunctiva. In left eye, punctal occlusion was noticed but other ocular examinations showed no abnormality.

Lid incision was made along the lid crease line, and we found the cyst located beneath levator aponeurosis and above the upper border of tarsal plate. The cyst measured $0.9 \times 1.2 \times 1.5$ cm with clear yellow cystic fluid. The biochemical analysis of the cystic fluid showed similar appearance of that of serum. Immunoglobulin analysis revealed high Ig A titer (Table 1).

Histopathologic examination showed two layered epithelium with goblet cells and mononuclear cell infiltration in the subepithelial area surrounding the cyst (Figs. 7A,B). A follow-up for 7 months showed no recurrence.

Both of the presented cases had cysts of two-layered epithelium, consistent with the findings of the cyst originating from the ductal structure. In both cases, the location of subconjunctival cyst was on the border of upper tarsal plate. Therefore, presented cysts were considered as ductal cysts of Wolfring’s accessory lacrimal gland.

**DISCUSSION**

The preceding cause of ductal cyst of accessory lacrimal gland is regarded as cicatricial disease of the conjunctiva.\textsuperscript{2,5} Presented cases also showed conjunctival scar change in varying degrees. When the duct of lacrimal gland was occluded experimentally, atrophic change was only noticed without cyst formation.\textsuperscript{5-6} Therefore, additional mechanism should be needed to explain the formation of the cyst. Weatherhead\textsuperscript{5} suggests that the driving force of cyst formation is produced by osmotic gradient produced by concentrated Ig A in cystic fluid which is secreted by plasma cells located in subepithelial area. Our second case also showed high Ig A concentration in cystic fluid, but further studies on pathogenetic mechanism is necessary.

The cystic change can be developed wherever lacrimal gland tissues are present: orbital or palpebral lobe of main lacrimal gland, accessory lacrimal gland, ectopic lacrimal gland, choristoma, and so on.\textsuperscript{4} Therefore, the origin of cyst can be considered as the site of the cyst. The cyst of main lacrimal gland is located on superolateral side of the lid.\textsuperscript{4,6-7} The cyst of Wolfring’s lacrimal gland is located on the superior border of upper tarsal plate or the inferior border of lower one.\textsuperscript{5} The cyst of Krause’s lacrimal gland is situated in the fornix of conjunctiva and cysts of ectopic gland or choristoma are found in various sites.\textsuperscript{2,4}

Implantation conjunctival cyst should be regarded as a differential diagnosis.\textsuperscript{2,6} True conjunctival cysts are characterized by the presence of non-keratinizing stratified squamous epithelium containing goblet cells which may or may not be present. Also, there might be a history of surgery or trauma. Conjunctival dermoid usually develops in the superomedial portion of the lid and consists of non-keratinizing stratified squamous epithelium and goblet cells, showing conjunctival characteristics and dermal appendages.\textsuperscript{8-9} Congenital orbital cysts associated with the common sheath of superior rectus and levator muscle should be considered for differential diagnosis. They are confirmed by the location of the cyst in operative findings and stratified squamous epithelium.\textsuperscript{10} Lymphatic cyst, lined by one layer of epithelium used to be reported as a subconjunctival cyst.\textsuperscript{2}

Management is done primarily by surgical intervention. It is important to remove the cyst completely because incomplete excision or simple aspiration can cause recurrence.\textsuperscript{5,7} In order to avoid cystic rupture, careful dissection is needed and it has been reported that a thin strip of 1-2 mm of tarsal plate upper margin should be excised.\textsuperscript{5} There are two approaches in cyst excision. Conjunctival approach may be superior because it neither incises the skin nor disturbs the levator aponeurosis. However, in cases where conjunctival shortening is found after cyst removal, the preventive measures for symblepharon formation are needed.\textsuperscript{5} Moreover, if levator dysfunction is found, surgical procedures for levator aponeurosis are necessary through conjunctival approach. Although skin approach needs skin incision, anatomic changes of levator aponeurosis can be observed readily and corrective measures are performed easily. Therefore,
appropriate method of approach should be selected for each case.

REFERENCES
