

Heterotopic Lacrimal Gland as a Component of Ocular Choristoma

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= Abstract = Review of 8 cases of heterotopic lacrimal gland tissue in epibulbar conjunctival region disclosed frequent mixture of pilosebaceous structure, adipose tissue, smooth muscle, etc. The term complex choristoma appears appropriate to designate a mixture of this variety, although each element can be predominant in a single lesion.

Complex choristoma containing lacrimal gland was commonly seen in both childhood and adulthood but had no sex predilection. The occurrence was frequent in bulbar conjunctiva and limbus although orbit and cornea were also the sites of involvement in some. The most frequent element other than lacrimal gland was fibroadipose tissue. One case showed nevocellular nevus involving the ducts of heterotopic lacrimal gland.

Key words: *Ectopic lacrimal gland, Heterotopia, Choristoma, Eye*

INTRODUCTION

Heterotopic or aberrant lacrimal gland tissue has been observed in the eye and in various sites in the ocular adnexa (Mettier 1958; Dal-lachy 1961; Baldrige 1981; Conway *et al.* 1985). The lesion was often diagnosed as dermoid or dermolipoma clinically. They appear as small nodules in epibulbar region and are usually present at birth. The majority are unilateral and are located in the superior temporal or temporal quadrant of the eye (Green and Zimmerman 1967; Spencer 1985).

The term choristoma has been used to describe dermoids and dermolipoma of the eyeball (Spencer 1985) because they contain displaced epithelial and dermis-like elements that are not normally found in these areas. Although the lacrimal gland may be one of ectopic tissues that are found in complex epibulbar choristoma, variable combinations of cartilage, adipose tissue and smooth muscle are also seen. When acinar elements predominate, the lesions have been classified as ectopic lacrimal glands.

We describe histological features of 8 cases

of heterotopic lacrimal glands of the eyeball and orbit, trying to elucidate the pattern of mixture with other tissue element.

MATERIALS AND METHODS

Cases with heterotopic glands were collected from the file of Department of Pathology of Seoul National University Hospital (SNUH) during last 3 years from 1986 to 1988. Four were patients of SNUH and the remainder (Case 1, 2, 3) were consulted cases from outside clinics.

The specimens were submitted for histological diagnosis under various clinical diagnoses (Table 1). Samples were grossly examined and oriented. In each case, the entire specimen was submitted for microscopic examination. The slides were made through routine procedure.

RESULTS

1. Clinical findings (Table 1)

The common presenting sign was a painless growing mass in bulbar conjunctiva in childhood. The age of the patients ranged from 2 years to 19 years. There were 5 males and 3 females. The onset ranged from birth to older childhood.

The lesion involved the right eye in 4 cases, left eye in 2 cases and both in 2 cases. In all cases the bulbar conjunctiva was the site of in-

Table 1. Summary of cases showing heterotopic lacrimal gland

Case	Age/ Sex	Onset	Eye effected	Site of lesion	Size (cm)	Clinical Diagnosis
1	5/F	birth	right	lateral bulbar conjunctiva	1.5×1×1	Dermoid
2	15/F	childhood	left	superior nasal bulbar conjunctiva	2.5×2×1	Dermolipoma
3	10/F	early childhood	right	superior lateral bulbar conjunctiva	1.8×1×0.5	Dermoid
4	12/M	4 years ago	right	laterla bulbar conjunctiva	0.9×0.6×0.4	Dermoid
5	19/M	10 years ago	bilateral	nasal bulbar conjunctiva	0.3×0.3 & 0.3×0.2	Nevus
6	14/M	childhood	right	lateral bulbar conjunctiva	1.5×2×1	Dermoid
7	2/M	birth	left	lower lateral limbus	1.0×0.5×0.5	Dermoid
8	5/M	birth	bilateral	lateral bulbar conjunctiva	1.5×0.8×0.3	Dermoid

Table 2. Histological components of the lesion with lacrimal gland

Case	Squamous epithelium	Skin apendage	Adipose tissue	Fibrocollagenous tissue	Smooth muscle	Skeletal muscle	Nerve
1	+	—	+	+	+	+	—
2	+	—	+	+	+	+	+
3	+	pilosebaceous	+	+	+	+	—
4	+	pilosebaceous & sweat glands	+	+	+	+	—
5	+	—	—	—	—	—	—
6	+	pilosebaceous	+	+	+	—	+
7	—	—	—	+	—	—	—
8	—	—	+	+	+	+	—

volvement although a portion of cornea was also affected in one case (case 7). Six out of 8 cases showed lateral (temporal) conjunctival involvement. The lesion appeared as a mass in most cases and measured from 0.3 × 3 cm to 2.5 × 2 cm. All cases except one (case 5) had a preoperative diagnosis of dermoid or dermolipoma.

2. Pathological examination

The histological features are summarized in Table 2. The individual case showed the following microscopical findings.

Case 1: Irregular islands of tubuloalveolar glands (Fig. 1) are admixed with large lobules of mature fat. The glands are 100% serous type with abundant zymogen granules in the cyto-

plasm. Small amount of skeletal and smooth muscle fibers are seen next to the lobules of glands. The ductal structures are often dilated, sometimes cystically, and contained fuzzy eosinophilic material. Mild small round cell infiltration is seen in interstitium between the lobules. A fine nerve fiber is embedded in mature fat.

Case 2: In the background of lobules of mature fat there are islands of tubuloalveolar glands. Acini consist of cells having coarse eosinophilic zymogen granules. Ductular and interlobular ducts are also seen without dilatation. Small round cell infiltration is present in the interstitium. One focus shows dilated ducts with flattened myoepithelial cells. Skeletal muscle fibers are included in the specimen together with

smooth muscle bundles (Fig. 2).

Case 3: The epithelial surface is of the stratified squamous type with pilosebaceous unit. The stroma contains lobules of lacrimal glands, bundles of dense collagen, smooth muscle fibers and adipose tissue (Fig. 3). Small round cell infiltrate is marked subepithelially.

Case 4: Irregular skeletal and smooth muscles are seen, intermingled with mature fat lobules of tubuloalveolar glands consisted entirely of serous glands and ductal structures. Focally dilated ducts are seen, inside and outside of the lobules. Small round cell infiltration is fairly diffuse. Overlying conjunctival epithelium is also infiltrated by small round cells. In one area dense collagenous connective tissue is seen partly mixed with fat tissue. Other section shows features of dermolipoma.

Case 5: The specimen was a small round nodule of dark tan tissue with focal whitish mottlings. The outer surface is smooth and pinkish white. Microscopically, the tissue is a globular mass covered by nonkeratinizing stratified squamous epithelium. Subepithelial stroma is loosely textured fibrous connective tissue in which islands of tubuloalveolar glands and dilated ductal structures are present. There are scattered islands of polygonal nevus cells mainly in the subepithelial stroma, focally extending into the overlying squamous epithelium (Fig. 4). The epithelium contains scattered mucus secreting cells and associated with chronic inflammatory cell infiltrate focally. The tubulo-alveolar glands form a few lobules of pure serous glands (Fig. 5). The nevus cell clumps involve the ductal structure but not the lacrimal gland proper. Some of the dilated ducts are surrounded by lymphoid infiltrate with several focal follicle formation. Approximately half of the nevus cells are heavily pigmented.

Case 6: In the background of typical dermolipoma, consisting of stratified squamous and pilosebaceous epithelium together with scattered hair follicles (Fig. 6) are irregular islands of tubuloalveolar glands of pure serous type. A small amount of chronic inflammatory exudate is seen in the interstitium. There are ductular and ductal structures some of which are dilated. The glands are clustered forming small to large islands without definite fibrous capsule but embedded in dense collagenous tissue.

Case 7: In the vicinity of nonkeratinizing squamous epithelium of conjunctival type, there are several islands of tubulo-alveolar glands composed 100% of serous cells, and the ductules are surrounded by a single layer of myoepithelial cell and are dilated containing eosinophilic secretory material. Inflammatory cell infiltration is minimal. In some areas the acini are also dilated. No other element is found.

Case 8: The lacrimal glands take approximately one-tenth of the entire mass and consist of tubulo-alveolar glands entirely of the serous variety. The acini are compact with cells having abundant zymogen granules. Ductal structures are seen with slightly distended lumens. Small round cell infiltrate is mild in the interstitium. A few fat cells are seen inside lobules. The remainders of the mass consist largely of mature fat and dense collagenous connective tissue. In addition, irregular bundles of smooth muscle are scattered. Small nerve twigs and vessels are also contained in the specimen.

DISCUSSION

Normally the lacrimal gland is in the superolateral corner of the bony orbit. It is divided into a deep orbital and a superficial palpebral lobe. Small accessory tear glands, the glands of Krause, are scattered along both fornices. And still smaller glands are present in the caruncle. The glands are compound serous tubuloalveolar type (Ham and Cormack 1979).

The term choristoma has been applied to microscopically normal cells or tissues that are present in abnormal locations (Robbins *et al.* 1984). Generally a choristoma is a cohesive mass of aberrant or heterotopic tissue. When more than one tissue element is involved it is sometimes called complex choristoma (Spencer 1985). When the lacrimal gland tissue is the sole element of the choristoma, well formed acinar and ductal structures are seen with lobule formation. Varying degree of inflammatory reaction is commonly associated (Green and Zimmerman 1967; Kim and Ahn 1981; Lee *et al.* 1987), as seen in our cases.

In our series of 8 cases it was obvious that heterotopic lacrimal gland were often associated with other types of heterotopic tissue rather than existed as a single component. In seven out of 8 cases the lacrimal gland was associated

with skin appendage, adipose tissue or smooth muscle. The lacrimal gland tissue was histologically quite normal, consisting of tubuloalveolar gland of pure serous type. Zymogen granules were frequently found in cytoplasm of the acinar cell cytoplasm. The ductular and ductal structures were also well formed.

Frequent association of other choristomatous elements along with lacrimal gland tissue prompted us to review the ocular dermoids to find if there is any small focus of salivary gland in the specimen. It appears reasonable to say that any heterotopic tissue in epibulbar region should be designated as choristoma. Particularly, when the heterotopic tissue includes salivary gland tissue, there is a higher chance that adipose tissue, smooth muscle and collagen vascular tissue are frequently accompanied to the complex choristoma.

It is likely that all epibulbar choristomas share a common origin from ectopic pluripotential cells capable of developing either into complex growths composed of several elements or into lesions containing a predominance of a single tissue. Thus a choristoma containing a predominance of pilosebaceous structures and collagen would be designated as a dermoid; one primarily composed of adipose tissue would be called a dermolipoma, while one predominantly comprised of lacrimal gland would be called heterotopic lacrimal gland, respectively. And if these are mixed together the term complex choristoma would be appropriate. It is apparent from this study that one should look more carefully microscopically when lacrimal gland tissue is found in the mass, for associated choristomatous tissue, since most lesions containing lacrimal gland tissue are actually complex choristomas.

Reviewing the Korean literature there are 2 reports on the heterotopic lacrimal gland. The first report is by Kim and Ahn in 1981 describing a 10 year old girl who came in with a painless nodule in right limbal region of 3 years duration. It was 12×4.5 mm and yellow white, and consisted of 3-4 lobules of acinar tissue with serous cells. Mild inflammatory change was seen in the mass. The other report by Lee *et al.* (1987) describes 2 cases of ectopic lacrimal gland. These 2 cases were included in the present series (Case 4, 5).

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= 국문초록 =

안구 미입종의 성분으로서의 이소성 누선조직

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안구결막에 발생하는 종괴는 대개의 경우 선천성 기형이라고 취급되는 병변인 경우가 많고 이것은 더모이드(dermoid)란 이름으로 잘 알려져 있다. 그런데 아주 드물게 이 부위에 누선조직이 발견되어 이를 문헌상 이소성 누선이라고 기술되어 왔다. 그러나 이러한 이소성 누선이 어떻게 나타나게 되었는가에 대하여는 정설이 없다.

본 논문은 문헌상 그 보고례가 흔치않고 한국에서는 3예만이 보고되어 있는 본 질환을 8종례를 통하여 자세한 조직학적 검색을 시행한 결과를 기술한 것이다. 그 결과 누선조직은 단독으로 존재하는 경우 보다는 기타조직 특히 지방조직, 모발피지조직, 평활근조직 등과 혼재하는 경우가 더욱 흔하며 따라서 이것은 단순한 이소성 조직이 아니라 미입종(choristoma)의 일종으로 취급되어야 할 증거를 찾을 수 있었다. 한편 이소성 누선조직의 의미와 이와 수반된 임상적 소견을 아울러 집약하였다.

LEGENDS FOR FIGURES

- Fig. 1. Photomicrograph of heterotopic lacrimal gland forming lobules and draining duct structure (Case 1). H & E X40.
- Fig. 2. Microscopic feature showing bundles of smooth muscle and mature fat tissue that are mixed together (Case 2). H & E X100.
- Fig. 3. Photomicrograph of the mass in case 3 revealing smooth muscle bundles and fat tissue. H & E X100.
- Fig. 4. Microscopic picture of bulbar conjunctiva, showing nests of pigmented nevus cells residing predominantly in subepithelial stroma. However, a small focus of epithelial involvement is seen in the left upper corner (Case 5). H & E X100.
- Fig. 5. Photomicrograph of heterotopic lacrimal glands made of acini of serous gland cell forming lobules and a draining duct (Case 5). H & E X100.
- Fig. 6. Photomicrograph showing dense collagenous connective tissue with hair follicles, sebaceous glands, and ductal epithelium (Case 6). H & E X100.
- Fig. 7. Microscopic picture of the well-formed serous gland with rich zymozen granules and slightly dilated ducts (Case 8). H & E X200.



