RESEARCH LETTER

Congenital Megacaruncle: A Unique and Innocent Ocular Adnexal Anomaly

Normal caruncular tissue contains a goblet cell–rich nonkeratinizing squamous epithelium, pilosebaceous units, eccrine and/or apocrine sweat glands, acini of lacrimal gland tissue, lobules of fat, and striated orbicularis muscle. Acquired lesions of the caruncle have been well categorized. The rarest lesions are congenital, including ectopias, dysgeneses (dysplasias), and duplications (supernumerary caruncles). We report a case of a congenitally well-formed massive caruncle that we have designated as a “megacaruncle.”

Report of a Case | A 55-year-old man had a lesion near the medial commissure of his right eye noted since birth (Figure 1A). It had never displayed a growth spurt. He had no ocular discomfort, visual decline, epiphora, or diplopia. Findings on a review of systems were negative. The lesion was firm, reddish, nontender, and attached to the right lower eyelid margin, which did not have a punctum. Visual acuity was 20/20 OU and the fundus was normal. The lesion was excised for cosmetic reasons.

The excised formalin-fixed tissue was white and firm and measured $7 \times 4 \times 3 \text{ mm}$. It was a symmetrically dome-shaped structure on both its external and cut fibrous surfaces. Histopathologically, it was covered by a goblet cell–rich nonkeratinizing squamous epithelium that was continuous with the lower eyelid margin. There was hyperkeratinization of the surface epithelium with focal invaginations of the epithelium into the stroma. The underlying stroma contained abundant goblet cells (Figure 1B). The lesion was composed of a combination of glandular structures and had a connective tissue stroma. The lesion was well-demarcated from the surrounding normal tissue.

Figure 1. Clinical and Histopathologic Features of Megacaruncle

A, An enlarged, reddish, right caruncle was present since birth with a glistening and opalescent apex. It was 8.5 mm in its greatest diameter compared with a 3-mm left caruncle. B, The megacaruncle is symmetrically globular with proliferation of the surface epithelium at the apex. The surgical margin is present below. Arrow indicates the point of transition between nonkeratinizing squamous epithelium that covers most of the structure and keratinizing epithelium on the left, which is where the mass was fused with the medial lower eyelid margin (hematoxylin-eosin, original magnification ×20). C, Higher-power photomicrograph of the mucocutaneous junction (crossed arrow) depicts the transition to keratinizing epithelium on the left. Arrows indicate sebaceous glands and lanugo hairs (hematoxylin-eosin, original magnification ×40). D, Cytokeratin 7 expressed in nonkeratinizing conjunctival epithelium is identified at the apex and on the right of the lesion. The epidermis on the left is nonstaining due to its keratinizing character (arrow) (immunoperoxidase reaction, diaminobenzidine chromogen, hematoxylin counterstain, original magnification ×20). E, Mild acanthosis with epithelial invaginations into the stroma typify the surface epithelium (hematoxylin-eosin, original magnification ×40). F, The invaginations (arrows) have a tubular structure and contain numerous goblet cells (hematoxylin-eosin, original magnification ×100).
tinizing squamous epithelium with a mild subepithelial infiltrate of chronic inflammatory cells (Figure 1B). There was a transition to keratinizing epidermis on its inferior aspect (Figure 1C). Cytokeratin 7 was found in the conjunctival epithelium but not the epidermis (Figure 1D). Epithelial tubular invaginations with goblet cells extended into the stroma, suggesting pseudoglands of Henle (Figure 1E and F and Figure 2A and B). Scattered lanugo hairs with their associated sebaceous glands were enveloped by thickly textured collagen fibers in the deep connective tissue (Figure 2C and D). The superficial stroma had the delicate collagen fibers of a substantia propria (Figure 2E). In the depths of the lesion were small lobules of adipose tissue and a few striated fibers of orbicularis muscle (Figure 2F). Eccrine, apocrine, and lacrimal gland tissues were not observed.

Under the jurisdiction of Massachusetts Eye and Ear Infirmary’s institutional review board, this study was considered exempt from institutional review board review. Informed consent was waived.

Discussion | The current lesion displayed many attributes of a normal caruncle except for its disproportionately large size and lack of sweat and lacrimal glands. Only 1 previous case seems to be related and was reported as an ectopic caruncle. This “ectopia,” however, was continuous with the normal caruncle and thus was most likely the placoid extension of one large caruncle.

Dysplastic caruncles, which can be bilobed (encantho-schisis), are usually small nubbins of tissue that are in situ or displaced onto the medial aspect of the inferior palpebral conjunctiva. Bilaterally dysplastic caruncles can be a stigma of Goldenhar syndrome. By contrast, supernumerary caruncles are always unilateral and unassociated with other ocular abnormalities or Goldenhar syndrome. In such cases, there is a normally formed caruncle in its usual anatomical locale and a second or, exceptionally, third structure located in the medial palpebral conjunctiva. To our knowledge, a supernumerary caruncle has never been encountered in the superior palpebral conjunctiva. The predilection for lower
medial palpebral conjunctiva probably stems from the embryologic origin of the normal caruncle from this topographic site.\textsuperscript{4,5} The major differential diagnostic consideration is a solid caruncular dermoid, which is also present at birth. There is only 1 persuasive caruncular dermoid in the ophthalmic literature,\textsuperscript{6} and it adhered to the superomedial eyelid margin (where colobomas may also occur with dermoids).\textsuperscript{7,8} In contrast, abnormal caruncular lesions always involve the lower eyelid. Microscopically, the caruncular dermoid possessed a keratinizing epidermis-like surface and dense, thick collagen in place of a substantia propria.\textsuperscript{6} In contrast, the present lesion exhibited a goblet cell-rich nonkeratinizing squamous epithelium with pseudoglands of Henle and subepithelial, thin collagen strands typical of the caruncular substantia propria.

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