A rare masquerade of chalazion: Adenoid cystic carcinoma

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Adenoid cystic carcinoma (ACC) is a rarely seen malignant epithelial tumor of the eyelids. We present a rare case of primary ACC arising from the tarsal plate of the lower lid that clinically resembled a chalazion. A 66-year-old female presented with a recurring nodule in her left lower lid. She gave history of surgery for chalazion removal from the same site twice earlier. An initial diagnosis of a recurrent chalazion was made, and incision and curettage was done. Light microscopy showed a solid tumor composed predominantly of sheet-like and nested pattern of basaloid to low-columnar cells with intervening fibrovascular septa and lacking an obvious cribriform or tubular architecture. The tumor cells were positive

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for pan-cytokeratin and CD117 and negative for adipophilin, HMB45, and BerEP4. A diagnosis of solid variant of ACC of the eyelid was made. Wide excision was performed and eyelid defect was reconstructed.

Key words: Adenoid cystic carcinoma, chalazion, eyelid malignancy

Adenoid cystic carcinoma (ACC) is a malignant epithelial tumor mostly arising from the major and minor salivary glands and the lacrimal gland. It has also been shown to arise from the esophagus, bronchi, skin, breast, lungs, and prostate.^[1,2] It is an extremely rare tumor of the eyelids, where it can arise from accessory lacrimal glands, palpebral lobe of the lacrimal gland, adnexal glands, or from ectopic lacrimal gland tissue.^[3]

We present a rare case of primary ACC arising from the tarsal plate of the lower lid that clinically resembled a chalazion.

Case Report

A 66-year-old female presented with a recurring nodule in her left lower lid. She gave history of surgery for chalazion removal from the same site, done elsewhere twice before. There was no other systemic positive history. On examination, her best-corrected visual acuity was 6/6 both eyes. There was a small nodule arising from tarsus measuring about 3 × 5 mm with an irregular surface, visible on the conjunctival side [Fig. 1]. No associated feeder vessels, lid margin deformity, or lash loss could be seen. The overlying skin was not adherent to the nodule. An initial diagnosis of a recurrent chalazion was made. A vertical incision was given from the conjunctival side and contents were curetted. Light microscopy of the curetted material showed a solid tumor composed predominantly of compact, sheet-like, and nested pattern of basaloid to low-columnar cells with intervening fibrovascular septa [Fig. 2a] and lacking an obvious cribriform or tubular architecture. The cells had moderate, pale eosinophilic cytoplasm and vesicular, round to elongated nuclei with conspicuous nucleolus in some [Fig. 2b]. No perineurial invasion was seen. The tumor cells were positive for



Figure 1: Small nodule arising from tarsus measuring about 3×5 mm with an irregular surface on the conjunctival side

pan-cytokeratin [Fig. 2c] and CD117 [Fig. 2d] and negative for adipophilin, HMB45, and BerEP4. A diagnosis of solid variant of ACC of the eyelid was made [Fig. 2]. Systemic examination was negative for any metastases.

To clear any microscopic residue, a wide excision under frozen section margin control was performed subsequently. Eyelid defect was reconstructed with a Hughes flap for posterior lamella and a temporal advancement flap for the anterior lamella of the eyelid. There was no recurrence after 3 years of follow-up.

Discussion

ACC is a slow-growing epithelial malignancy. ACC of the eyelid is a rare entity and has a tendency to recur locally. It is a subtype of sweat gland carcinoma, and metastasis to lymph nodes and distant organs is rare.^[1]

It may mimic a chalazion in that it presents as a slow-growing, painless nodule.^[4-6] However, firm nodules with associated loss or distortion of cilia or skin ulceration in a middle-aged or elderly patient give a clue about a possible underlying malignancy.^[2]

Goto *et al.* reported a case of primary ACC of the eyelid that clinically resembled a chalazion.^[7] In their case, although there was no skin ulceration, loss of cilia was observed corresponding to the indurated eyelid lesion. In our case, however, there was no skin ulceration, no rubor, neither was there any loss of cilia or feeder vessels noted. The only clue to a possible malignancy was the history of recurrence of the chalazion-like lesion at the same site.



Figure 2: Histopathologic features of solid adenoid cystic carcinoma. (a) Compact, sheet-like, and nested arrangement without any obvious cribriform or tubular pattern (hematoxylin and eosin, ×40). (b) Relatively uniform basaloid cells with pale cytoplasm and vesicular nuclei (hematoxylin and eosin, ×400). (c) Positive pan-cytokeratin in tumor cells (pan-CK, ×200). (d) Positive staining for CD117 in tumor cells (CD117, ×400)

On histology, ACC is classified into solid, tubular, and cribriform variants. Among these, tumors showing solid pattern on histology have a worse prognosis.^[10] Histopathologically, it is one of the differential diagnoses of adenoid basal cell carcinoma.^[3] They are differentiated from each other in that ACC lacks contiguity with the hair sheaths or epidermis, and there is lack of peripheral palisading of nuclei on histopathology. In the present case, there was no contiguity with the epidermis or hair sheath, and peripheral palisading of nuclei was not seen. Other differential diagnoses include basal cell carcinoma, sweat gland carcinoma, and sebaceous carcinoma. While negative staining with BerEP4 excludes a BCC and lack of adipophilin expression excludes a sebaceous carcinoma, CD117 expression helps ascertain the diagnosis of ACC.

ACC of the eyelid has a better prognosis when compared with ACC of the lacrimal gland, the other most common site of ACC in ocular adnexa.^[2] One of the possible causes could be that patients seek medical attention sooner in eyelid ACC that typically presents as nodules. Also, eyelid ACC has a more indolent course. The origin of ACC in this case is unclear. The possible sites could be glands of Moll, accessory lacrimal glands in conjunctiva, or ectopic lacrimal gland tissue. Since the overlying skin was free from the nodule, it is less likely to be of cutaneous origin. ACC being a relatively rare tumor of the eyelid, optimal treatment is yet unknown. We chose to do a wide excision with verification of margins in our case. In cases where total resection is not possible, such as in cases of perineural invasion, radiotherapy and chemotherapy also have an adjuvant role.^[3]

It cannot be commented upon whether the previous lesions that were excised for this patient were chalazion or ACC. Since the patient had a history of recurrence, long-term follow-up is needed. There are no signs of recurrence in this case after 3 years of follow-up. Our case highlights the importance of histopathological examination in a case of chalazion. Histopathological examination of the curetted material must be considered in recurrent chalazion-like lesion in elderly, or associated with destruction of meibomian gland architecture and loss of cilia or when intralesional or feeder vessels are seen.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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