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Case Report

Solitary infundibulocystic basal cell carcinoma of lid- A case report and review of literature

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ABSTRACT

Purpose: The infundibulocystic subtype of Basal cell Carcinoma (IBCC) is a rare entity. It was proposed as a new BCC variant and has been described as a sporadic lesion or as part of Multiple Hereditary Infundibulocystic BCC (MHIBCC). IBCC in the lids associated with MHIBCC or basal cell naevus syndrome has been reported. However an isolated IBCC of lids has seldom been reported in ophthalmic literature. We report a rare case of solitary IBCC of lids.

Materials and Methods: We report a rare histopathological variant of the commonest lid malignancy, ie BCC.

Results: Histopathology of excised lesion of a recurrent chalazion turned out to be IBCC, a rare variant to date reported only as part of MHIBCC.

Conclusions: The solitary IBCC, though yet to be reported in lids in ophthalmic literature does exist and ophthalmologists as well as pathologists should be aware of this entity and its less aggressive course.

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1. Introduction

Basal cell carcinoma (BCC) is the most common cancer in the world and constitutes 90% of malignant lid tumours. The infundibulocystic sub type of BCC (IBCC) is a rare entity first described in 1987 and proposed as a new BCC variant by Ackerman and Walsh in 1990.^{1,2} It has been described as a sporadic lesion or as part of Multiple Hereditary Infundibulocystic BCC (MHIBCC).³ There have been a few reports of IBCC in the lids associated with MHIBCC or basal cell naevus syndrome.^{3,4} However solitary IBCC of lids is a least documented entity in ophthalmic literature.

2. Case Summary

A 63 year old gentleman, a nondiabetic, presented to our outpatient department with complaints of a small painless

swelling in his upper lid since 3 weeks, which he felt was gradually increasing in size. He gave history of a similar swelling in the same lid 5 months before which was surgically removed elsewhere. He did not have any records of the previous intervention and was not sure whether the swelling was at the same site. There was no family history of similar eye lesions.

Ocular examination showed a lesion involving the upper lid just above the margin with fixity to skin and tarsal plate. There was no ulceration, vascularisation or discoloration of the overlying skin, or any regional lymph node enlargement. With the provisional diagnosis of a benign lesion, presumptively a chalazion or sebaceous cyst, he was taken up for Excision biopsy of the lesion with RF knife after the routine investigations. Intraoperatively it was a solid lesion involving the anterior lamella of the lid which was excised and sent for histopathological examination and the defect sutured.

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Histopathology grossly showed a brownish nodule measuring 0.5x 0.3x 0.2 cm, attached to the skin. Microscopy showed the tumour arising from the epithelium composed of basaloid cells with hyperchromatic pleomorphic nuclei, occasional mitoses and peripheral palisading. A few tiny cysts lined by infundibular epithelium were seen and stroma had foci of mucin pools suggesting a diagnosis of IBCC. (Figure 1)

Since the margins and base showed involvement, he underwent a wider excision of the base and margins and after pathologic confirmation that the margins and base were free secondary reconstruction of the defect was done. The patient has been asymptomatic subsequent to healing of the wound and suture removal and now has completed an uneventful follow-up of 6 months.

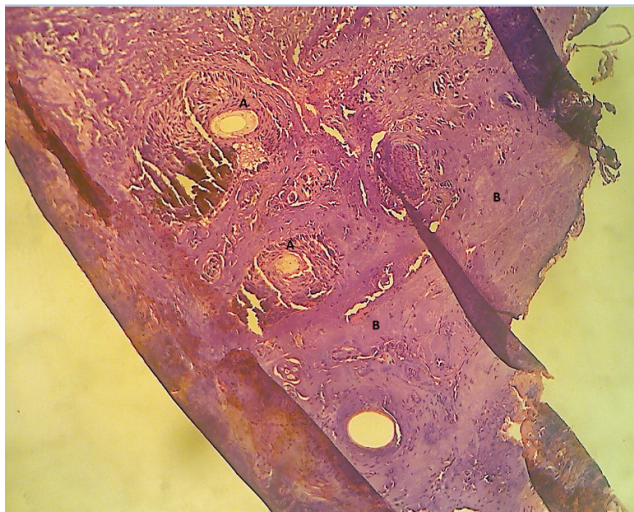


Fig. 1: Histopathology H&E stain Original magnification x 100; **A:** Tiny cysts lined by infundibular epithelium; **B:** Stroma showing mucin pools

3. Discussion

The first report of IBCC in ophthalmic literature is a retrospective series (2010) of 5 patients with basal cell naevus syndrome where 23 of the 26 eyelid lesions were BCC on histopathology and 57% were IBCC.⁴ IBCC which can be clinically indistinguishable from the more common BCC forms, are thought to be less aggressive than other types, tend to be indolent with little tendency to ulcerate and are a reassuring histopathologic diagnosis.^{3,5}

Histopathologically the tumour variant shows basaloid cells with hyperchromatic, pleomorphic nuclei, rare mitoses, multiple tiny cysts containing cornified cells lined by infundibular epithelium and abundant mucin in stroma.²

It is important for the ophthalmologist and pathologist to be aware of IBCC, as they are more common in patients

with basal cell nevus syndrome and may be a clue to the diagnosis of this autosomal dominant cancer-predisposition syndrome. Also, the rarity of the subtype can result in histopathologically incorrect classification as the solitary type of IBCC is yet to be reported in ophthalmic literature. Ophthalmologists in particular should be aware of this less recognised entity for which a non-surgical expectant management has been advocated in MHIBCC.^{5,6} though evidence for such less aggressive management is lacking for solitary IBCC, due to its rarity in literature.

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5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Ackermann TT, Ackerman AB. Basal Cell Carcinoma with follicular differentiation. *Am J Dermatopathol.* 1987;9(6):474–82. doi:10.1097/00000372-198712000-00002.
2. Walsh N, Ackerman A. Infundibulocystic basal cell carcinoma: a newly described variant. *Mod Pathol.* 1990;3(5):599–608.
3. Requena L, Farina M, Robledo M. Multiple hereditary infundibulocystic basal cell carcinoma: a genodermatosis different from nevoid basal cell carcinoma. *Arch Dermatol.* 1999;135(10):1227–35. doi:10.1001/archderm.135.10.1227.
4. Belliveau MJ, Coupal DJ, Brownstein S, Jordan DR, Prokopetz R. Infundibulocystic basal cell carcinoma of the eyelid in basal cell nevus syndrome. *Ophthal Plast Reconstr Surg.* 2010;26(3):147–52. doi:10.1097/IOP.0b013e3181b80b65.
5. Janjua R, Ali AP, Siddiqui AP. Peri-Ocular and Facial Multiple Hereditary Infundibulocystic Basal Cell Carcinoma. *Pak J Ophthalmol.* 2008;24(1):41–4.
6. Herman AR, Klaus JB, Greenberg RA. Multiple infundibulocystic basal cell carcinomas: Case report of unique unilateral presentation. *Dermatol Surg.* 2003;29(4):436–9. doi:10.1046/j.1524-4725.2003.29107.x.

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