CASE REPORT

Caruncular Oncocytoma in an Elderly Pakistani Female: A Case Report

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ABSTRACT

Caruncular Oncocytoma (CO) is a benign epithelial tumor, frequent in elderly females. Caruncular lesions are rare compared to conjunctival lesions, with a reported incidence of 0.3 to 1.1 %, and histologically proven oncocytoma are 0.3 million/year. The mitochondrial and somatic gene mutations, resulting in defective oxidative phosphorylation, are responsible for **o**ncocytoma. This reduces ATP, resulting in a decrease in energy production, which, in compensation, increases mitochondrial content. The common complaint is epiphora, which is excessive lacrimation. CO is composed of oncocytes with fine, eosinophilic cytoplasm containing numerous mitochondria. A 60-year-old lady presented in an Ophthalmology outdoor clinic with a small, nodular, painless, reddish-colored swelling at the inner canthus of the left eye for three years. No clinical complaints and visual disability were noted. On physical examination, a mass with overlying, reddish-colored mucosa was located at the medial canthus of the left eye. Excision in Toto was done. Grossly, there was a 10 mm × 5 mm × 5 mm smooth surfaced, red-colored mass with round contours. Microscopy showed a well-encapsulated mass with overlying thinned-out stratified squamous epithelium and sebaceous glands. The underlying lesion is composed of tall, columnar, uniform cells with round to oval nuclei, fine chromatin, and abundant granular cytoplasm.

Key Words: *Caruncle, Oncocytoma, Mitochondria.*

Introduction

Caruncular Oncocytoma (CO) is a benign glandular epithelial/subepithelial tumor composed of granular abundant eosinophilic cytoplasm with para-central nuclei. Oncocytomas are also noted in other organs, i.e., thyroid gland, parathyroid gland, adrenal gland, kidneys, and gastrointestinal tract.[1] The most commonly reported are renal oncocytomas. Regarding head and neck region, OC are reported in lacrimal gland, salivary glands, caruncle, bulbar conjunctiva, and plica semilunaris. Caruncle is a bilateral triangular structure present distal to the medial canthus. Caruncular lesions are rare as compared to conjunctival lesions, with a reported incidence of 0.3 to 1.1 %, and histologically proven OC are 0.3 million/year.[2] CO, or oxyphil cell adenoma, is a benign tumor that presents unilaterally. Ninety five percent of CO is benign. Apart from caruncle, it is also noted in the lacrimal gland, sac, conjunctiva, and eyelid margins. The mitochondrial and somatic gene mutations, resulting in defective oxidative phosphorylation, are responsible for oncocytoma. This reduces ATP, resulting in a decrease in energy production, which, in compensation, increases mitochondrial content.[3] The common complaint is epiphora, which is excessive lacrimation. Slit-lamp examination is the initial investigation of choice. Complete surgical resection is the treatment of choice. There is minimal chance of recurrence. CO is an uncommon tumor but should be included in the list of differential diagnosis. CO is composed of oncocytes with fine, eosinophilic cytoplasm containing numerous mitochondria.[4]

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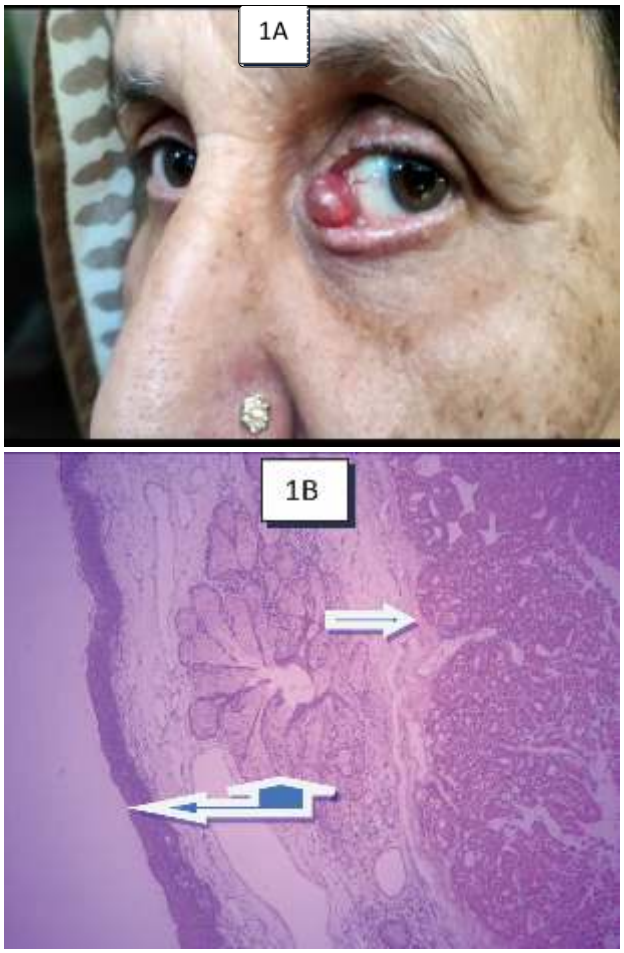
Case

A 60-year-old female patient presented in an Ophthalmology outdoor clinic. There was a small, nodular, painless, reddish-colored swelling at the inner canthus of the left eye for three years (Figure 1A). Clinically, the patient did not report any symptoms such as lacrimation, pain, discharge, or bleeding. On visual examination, visual acuity and ocular motility were unremarkable. She did not give any history of eye trauma, weight loss, or surgery was provided. On physical examination, a mass with overlying, reddish-colored mucosa was located at

the medial canthus of the left eye. Her baseline blood investigations were normal in range. The patient was prepared for surgery and the lesion was excised via a transcaruncular approach by giving a vertical incision in the conjunctiva between the caruncle and plica semilunaris. The mass was excised in toto and sent for histopathological examination. Gross examination showed there was a 10 mm × 5 mm × 5 mm smooth surfaced, red-colored mass with round contours. The cut surface was cystic brown. Microscopy showed a well-encapsulated mass with overlying thinned-out stratified squamous epithelium and sebaceous glands (Figure 1B). The underlying lesion was composed of tall, columnar, uniform cells with round to oval nuclei, fine chromatin, and abundant granular cytoplasm. Dark round to ovoid paracentral nuclei were noted. The oncocytes were arranged in solid cords and tubular structures with cystic spaces. Interspersed lymphoplasmacytic infiltrate was also noted. No cytological atypia, mitosis, and necrosis were found (Fig 1C &D).



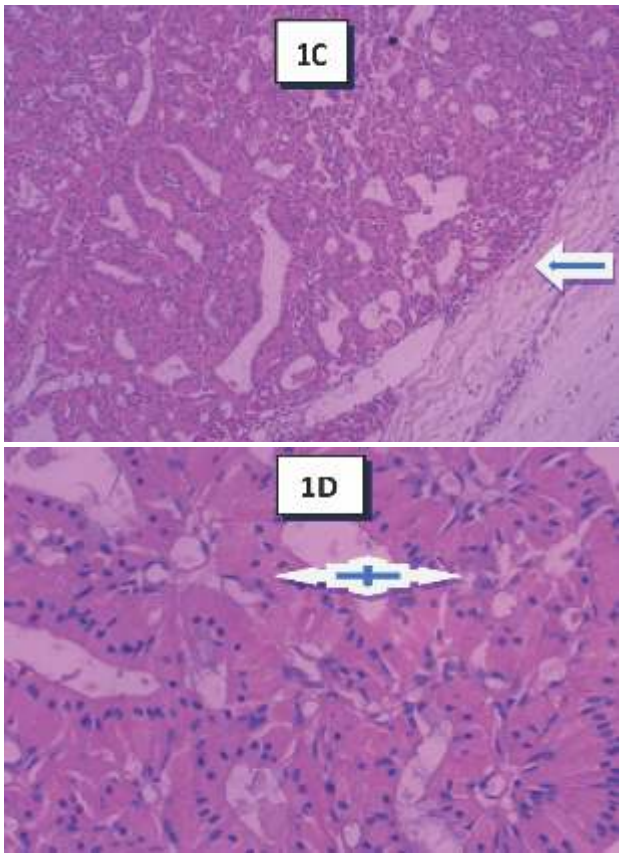


Fig 1A: Presence of a tumor in the medial canthus of left eye. Fig 1B: A panoramic view shows skin (arrows) with underlying sebaceous glands and well circumscribed lesion arranged in lobules. Fig 1C: H&E stain shows a well circumscribed lesion predominantly arranged in glandular pattern. Fig 1D: H&E shows a high-power view of lesion arranged in glandular pattern. The neoplastic are large containing abundant eosinophilic cytoplasm and small round nuclei on the apical side.

Discussion

The case of CO in an elderly Pakistani female highlights the rarity and distinctive pathology of this benign tumor, characterized by its prevalence in the elderly and its primary manifestation at the caruncle. The successful excision with no recurrence emphasizes the effectiveness of surgical intervention. Despite being rare, CO is being reported by various case reports and studies around the globe. OC are oncocytes, which are modified epithelial cells with large size and voluminous granular, eosinophilic cytoplasm. Electron microscopic examination showed a cytoplasm packed with abnormal and dysfunctional variable-sized irregular mitochondria. Clinically, differentials include squamous papilloma, lobular capillary

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hemangioma, melanocytic nevi, and basal cell carcinoma.[1-4]

This case report from Pakistan contributes to the limited available data on CO in South Asia, where few cases have been documented. For example, Alam MS *et al*.,[2] and Mitra S *et al*.,[4] reported one and two cases over two decades, and Clemens AC *et al*.,[1] documented nine cases over 22 years. Our case is unilateral and solitary, consistent with reports by Clemens AC *et al*.,[1] Alam MS *et al*.,[2] Heathcote JG *et al*.,[3] and Fenelon EM *et al*.,[6] although some cases of bilateral CO have been noted. The slow growth over three years aligns with the findings by these authors. The duration of symptoms for CO typically ranges from two months to five years, with Alam MS *et al*.,[2] reporting a mean of 36 months and Heathcote JG *et al*.,[3] noting 3-4 years, while Mitra S *et al*.,[4] reported the earliest duration at four months. Rest of the cases are described in Table I and II for detailed demographic comparison of index case with total cases, CO cases, site of lesion, and time-period noted.

The reported average age is 68 years, and most COs were seen at more than 55 years of age, with female preponderance. The index study showed a 60-year-old female like the one reported by AC Clemens *et al*.,[1] and Heathcote JG *et al*.,[3]. However, in contrast, Luthra *et al*.,[7] reported a young male patient of 30 years in 1978, and Fenelon EM *et al*.,[6] reported an oncocytoma of the lacrimal gland in a 4-year-old African girl. The differential diagnosis is narrowed down according to the color and size of tumors noted in the caruncle. Our study showed that the left eye's inner medial canthus is 10mm in size. Similarly, Indian author Mitra *et al*.,[4] reported 7 and 11 mm-sized tumors. However, one case reported by Heathcote JG *et al*.,[3] was 7mm, and in contrast, another case was 23mm in size. The clinical appearance of CO in the index case was a reddish round mass, similarly, reported by AC Clemens *et al*.,[1] however, the color and size of lesions reported do not affect the prognosis.

Conclusion

CO are rare benign tumors frequent in elderly females. No clinical complaints, and visual disability were noted. Excision in Toto is the treatment of

Table I: A Detailed Comparison of Index Case with Case Series with International Studies

S .No	Author	Year of publication	Country	Age in years	Gener	Site of tumor
1	Fenelon *et al* [6]	2015	Brazil	4o	Female	Lacrimal Gland Presented with proptosis
2	Surakiatchanukal *et al* [5]	2017	USA	81	Female	Caruncle Clinically mimicking melanona
3	Mitra *et al* [4]	2018	India	72 and 80	Female and male	Caruncle Cystic and solid with papillary configuration Solid with central degeneration
4	Heathcote *et al* [3]	2022	USA	70 and 87	Male Female	Left Caruncle Mainly cystic Right Lacrimal sac Solid and cystic

Table II: A Detailed Comparison of Index Case with Case Reports with International Studies

S. No	Author	Year of Publication	Country	Centre	Total Cases	Oncocytoma Cases	Time Period in Years	Gender Female: Male	Site of Tumor	Significant Findings
1	Levy et al[10]	2008	Israel	Single center	42	3	17	2:1	Caruncle	Pigmented lesions
2	Sola ri et al[8]	2009	Canada	Single center	42	3	15	2:1	Caruncle	-----
3	Shiel ds et al[11]	2016	USA	Multicenter	5002	6	40	51:49	Lacrimal Glands	-----
4	Alam et al[2]	2002	India	Single Center	87	1	20	49:51	Caruncle	-----
5	Clemens et al[1]	2002	Germany	Single Center	82	9	22	4:5	Lacrimal Gland and Caruncle	5 Solid and 4 Cystic Morphologies

choice with no recurrence. Given the rarity of CO, the timely and accurate reporting of these lesions is paramount. Our case report adds to the limited literature available on these benign tumors.

Conflict of Interest

Authors declared no conflicts of Interest.

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Data Sharing Statement

The data that support the findings of this study are available from the corresponding author upon request

REFERENCES

1. Clemens AC, Loeffler KU, Holz FG, and Herwig-Carl MC. Clinico-pathological correlation of lacrimal caruncle tumors: a retrospective analysis over 22 years at the University Eye Hospital Bonn. *Graefes Arch*. 2022 Apr 1:1-1. doi:10.1007/s00417-021-05464-x.
2. Alam MS, Pal SS, and Krishnakumar S. Clinical and histopathological profile of primary caruncular lesions. *Indian J. Ophthalmol*. 2022 Jun;70(6):1975. doi:10.4103/ijo.IJO_2724_21.
3. Heathcote JG, Archibald CW, and Valenzuela AA. Oncocytic lesions of the ocular adnexa: A review of the histopathology with a brief discussion of the pathogenesis. *Saudi J. Ophthalmol*. 2021 Jul;35(3):179. doi:10.4103/SJOPT.SJOPT_93_21.
4. Mitra S, Lath K, Samanta R, Saikia UN. Caruncular oncocytoma: report of two cases with review of literature. *Indian Dermatol. Online J.* 2018 Sep;9(5):324. doi:10.4103/idoj.IDOJ_241_17.
5. Surakiatchanukul T, Sioufi K, Pointdujour-Lim R, Eagle Jr RC, Shields JA, *et al*. Caruncular oncocytoma mimicking malignant melanoma. *Ocul. Oncol. Pathol.* 2017 Nov 28;3(4):320-3. doi:10.1159/000468524.
6. Fenelon EM, Balby IT, Neves NT, Figueiredo F, Duarte EC, Tzelikis PF. Oncocytoma of the lacrimal gland: a case report. *Arq. Bras. Oftalmol.* 2017 Mar;80: 128-30. doi:10.5935/0004-2749.20170031.
7. Luthra CL, Doxanas MT, Green WR. Lesions of the caruncle: a clinicohistopathologic study. *Surv. Ophthalmol*. 1978 Nov 1;23(3):183-95. doi:10.1016/0039-6257(78)90155-8.
8. Solari, H.P., Ventura, M.P., Orellana, M.E. Histopathological study of lesions of the caruncle: a 15-year single-center review. *Diagn. Pathol*. 4, 29 (2009). doi:10.1186/1746-1596-4-29.
9. Kaeser PF, Uffer S, Zografos L, Hamédani M. Tumors of the caruncle: a clinicopathologic correlation. *Am. J. Ophthalmol.* 2006 Sep 1;142(3):448-55. doi:10.1016/j.ajo.2006.04.035.
10. Levy J, Ilsar M, Deckel Y, Maly A, Pe'er J. Lesions of the caruncle: a description of 42 cases and a review of the literature. *Eye.* 2009 May;23(5):1004-18. doi:10.1038/eye.2008.316.
11. Shields CL, Alset AE, Boal NS, Casey MG, Knapp AN, Sugarman JA, *et al*. Conjunctival tumors in 5002 cases. Comparative analysis of benign versus malignant counterparts. The 2016 James D. Allen Lecture. Am. J. *Ophthalmol.* 2017 Jan 1;173:106-33. doi:10.1016/j.ajo.2016.09.034.

CONFLICT OF INTEREST

Authors declared no conflicts of Interest.

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Authors have declared no specific grant for this research from any funding agency in public, commercial or nonprofit sector.

DATA SHARING STATMENT

The data that support the findings of this study are available from the corresponding author upon request.