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CASE REPORT

Conjunctival keratoacanthoma: a case report and literature review

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ABSTRACT

Keratoacanthoma is typically a skin lesion which rarely occurs in Conjunctiva. On the literature review, only 16 Cases of Conjunctival Keratoacanthoma have previously been reported in the English language. All previously described lesions occurred at the limbus.

We describe here a case of a 43-year-old Woman with a lesion that is compatible both clinically and histopathologically with conjunctival keratoacanthoma. The treatments given were excisional biopsy and amniotic membrane transplant. The outcome was a complete clinical remission during the follow-up period (3 months).

Conjuctival keratoacathoma is rare and making correct differential diagnosis between kerathoacanthoma and Squamous cell carcinoma is very important. We recommend complete surgical exicision and carrying out close monitoring after surgery due to the possibility of relapse and conversion to Squamous cell carcinoma.

Key words: conjunctival keratoacanthoma, squamous cell carcinoma, conjunctiva

INTRODUCTION

The term keratoacanthoma refers to a wide range of benign skin tumors characterized by rapid growth which usually leads to complete regression after approximately 6 months.[1] Keratoacanthoma first described by Sir Jonathan Hutchinson[2] in 1889 as a crateriform ulcer of the face. Periocular keratoacanthoma presents as papules usually on the lower eyelid in patients with chronic sun exposure or immunocompromised patients. Keratoacanthoma rarely appears in conjunctiva; squamous cell carcinoma is well-known in differential diagnosis of Keratoacanthoma, Histopathologically and clinically.[3] Only 16 cases of conjunctival keratoacanthoma have been reported in the literature. Among all described cases of conjunctival keratoacanthoma, only the one presented by Grossniklaus in 1990 invaded the anterior chamber which authors decided for enucleation. Here, we present a case of conjunctival keratoacanthoma and comment on the clinical findings of this case and previous reported patients in the literature.

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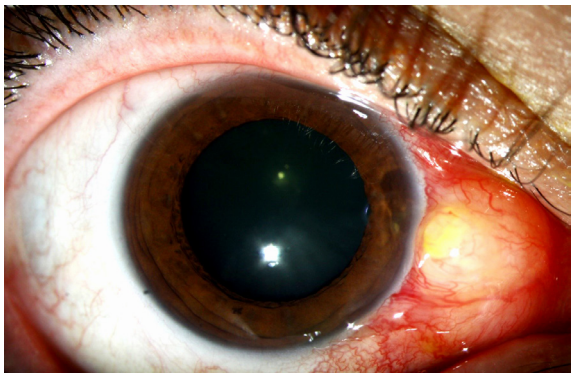

Figure 1, Conjunctival keratoacanthoma of right eye shows a generalized conjunctival redness on the nasal side surrounded with dilated and tortuous vessels.

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A 43-year-old diabetic woman referred to cornea clinic of Farabi Hospital due to redness of her right eye since a few weeks ago.

On examination the best corrected visual acuity (BCVA) was of 0.20 with a refraction of -4.25-2.5×175 in right eye. Her left eye BCVA was 0.90 with a refraction of -3.25-2.00×25. According to the patient, he has right visual loss from previous years. The patient referred with rapid progression of the injury (2–3 weeks), which greatly increased in size, causing ocular hyperemia and foreign body feeling. Anterior segment's examination of left eye presented normal results.

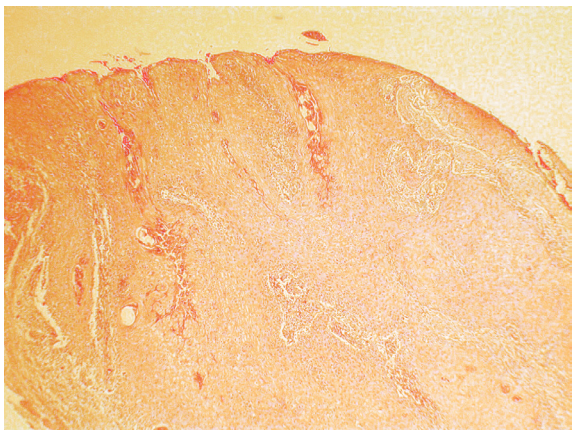

Figure 2, Section of conjunctival tumor showing proliferative squamous cells with mild atypia, pleormorphic vesicular nuclear, prominent nucleoli and sparse mitosis.

Biomicroscopic examination of right eye revealed a generalized conjunctival redness on the nasal side and a protruding hyperemic lesion in conjunctiva measuring 5 mm × 4 mm × 3 mm with an a vascular yellowish central area which invaded the nasal limbus and surrounded with dilated and tortuous vessels (Fig. 1). Dellen Ulcer was developing on the cornea adjacent to the tumor. Other anterior segment's examinations including iris, lens and anterior compartment were normal. Upper and lower lid were normal. Fundus examination revealed evidences of diabetic retinopathy in the form of mild non-proliferating diabetic retinopathy (NPDR) in both eyes. The Patient did not refer previous conjunctival disorders or relevant personal history. He has not revealed any history of eye trauma. No skin lesion was seen. There was no risk factor due to kerathoacanthoma.

The lesion was treated with complete excision and primary amniotic membrane transplant and sent for histological and cytological analysis. The histological report was as followed: section shows histology of conjunctiva with stratified squamous epithelium; reveal marked acanthosis with papillomatosis, hyperkeratosis, parakeratosis and dyskeratosis. Proliferative squamous cell shows mild atypia with pleormorphic vesicular nuclear, prominent nucleoli and sparse mitosis.

The squamous epithelial proliferations external downward the subepithelium, degeneration of collagen and moderate leukocytic infiltration at subepithelium are seen, predominantly composed of lymphocytes and plasma cells. Histological findings are consistent with keratoacanthoma.

Three months after resection the patient did not exhibit any signs of relapse.

DISCUSSION

One of the relatively common skin neoplasms originating from pilosebaceous glands is keratoacanthoma (KA) which is normally characterized by the rapid growth of a painless keratotic nodule over a few weeks to months. Most cases are known to regress over 4-6 months without any treatment; however tumors are normally excised due to the high probability of malignancy and the difficulty of distinguish between KA and conventional well differentiated squamous cell carcinoma (SCC). Actually the main differential diagnosis is SCC.

Freeman *et al.*, described the first case of conjunctival KA in 1961 in a 55-year old white man.[4] Only 16 cases have been described to date, mainly in white. Conjunctive keratoacanthoma usually involves younger adults with the age range of 26 to 65 years and male predominance, while skin kerataocanthoma affects 58-92 years old individuals mostly.[5] Our patient was woman and 43 years old.

To the best of our knowledge, there are 2 cases of conjunctival kerathocanthoma reported in Asia. The first report on 2001 was about a 41-year-old Thai woman presenting a cup shaped well-circumscribed squamous cells with central keratin crater as a robust marker for Kerathocanthoma.[6] On 2001 later, a 39-year-old Japanese patient was described as the second case of conjunctival keratoacanthoma in Asia.[7] Just similar to the Thai patient, the diagnosis was established on this case based on examinations resembling a keratin-filled crater surrounded by epidermis extending in a liplike manner over its sides.

Researches dedicated a great body of investigations to find out the exact external aggregations for conjunctival Keratoacanthoma including traumatic injury, sunlight, environmental carcinogens and viral infection.[8] In our case no risk factor was seen.

Previously reported cases of bulbar conjunctival Keratoacanthoma presented a rapid-growth leading to excision in three weeks. Earl D reported a 28-year-old Caucasian male presenting an initial white mass on the nasal bulbar conjunctiva of left eye which progressed to the complete excision with the purpose of interrupting the pathologic process of malignant neoplasm.[9] As characteristically observed in KA, our patient's lesion grew rapidly; it was removed as it could progress to malignancy .

A 2004 case report by Perdigão, FB *et al.* introduced a 34-year-old woman with a conjunctival tumor regarding a crateriform histopathologic appearance. The initial differential diagnosis was performed in order to distinguish between KA and squamous cell carcinoma (SCC)[10]; As such in our report, histopathological examination showed proliferative squamous cell with mild atypia and pleomorphic vesicular nucleoli and sparse mitosis which were in a robust consistency with keratoacanthoma.

Histopathological examination disclosed KA as heavily keratin-filled skin excavations surrounded by acanthotic epithelium which mostly show inflammatory infiltrates. Lesions might tend to invasive stage and thus some may interpret them as a variant of squamous cell carcinoma. A case of 40-year-old man from Barbados described by Hughes EH emphasizes the need of histological diagnosis as the appearance of

Table 1, *Review of the conjunctival keratoacanthoma cases to date*

Authors	Year	Age	Sex	Race	Eye	Anatomic place	City/ Country
Freeman *et al.*	1961	55	Male	White	Right	Temporal Limbus	Barbados
Bellamy *et al.*	1963	28	Male	-	Left	Nasal Limbus	New Orleans
Bellomio	1970	41	Male	-	Left	Nasal Limbus	-
Roth	1978	26	Female	White	Right	Temporal Limbus	-
		37	Male	White	Left	Temporal Limbus	
Hamed *et al.*	1988	49	Male	White	Right	Temporal Limbus	-
		40	Male	White	Right	Temporal Limbus	
Grossniklaus *et al.*	1990	65	Male	White	Left	Temporal Limbus	Atlanta
Munro *et al.*	1993	42	Male	-	Left	Temporal Limbus	-
Grossniklaus *et al.*	1996	23	Female	Black	Right	Eyelid	Atlanta
		75	Male	White	Left	Eyelid	
		60	Female	-	Left	Eyebrow	
Schellini *et al.*	1997	28	Female	Mulatto	Left	Nasal Limbus	Botucatu
Coupland	1998	37	Male	Black	Left	Nasal Limbus	Ethiopia
Tulvanta *et al.*	2001	41	Female	White	-	-	Thailand
Kifuku *et al.*	2001	39	Male	White	Right	Bulbar conjunctiva	Japan
Hughes *et al.*	2003	40	Male	White	Right	Nasal Limbus	Barbados
Perdiago *et al.*	2004	34	Female	White	Right	Nasal Limbus	Campinas
Oellers *et al.*	2014	83	Male	White	Right	Temporal Limbus	Miami
Ozge *et al.*	2016	24	Male	White	Left	Nasal Limbus	Ankara
Gohari *et al.*	2017	43	Female	White	Right	Nasal Limbus	Iran

conjunctival keratoacanthoma and in non-caucasians is rare.[11]

There has been a great concern toward the chance of malignant neoplasms and permanent scars; thus, this fact will shed light on the importance of surgical removal besides the potential ability of keratoacanthoma for spontaneous regression. One of the most recent reports of conjunctival keratoacanthoma has been introduced by P. Oellers in 2014.[12] The diagnostic uncertainties between KA and SCCA led to the treatment of this 83-year-old male patient with wide surgical excision. For that reason,complete exicision and amniotic membrane transplantation (AMT) was done in our patient.

CONCLUSION

Conjuctival keratoacathoma is rare and making correct differential diagnosis between kerathoacanthoma and squamous cell carcinoma is very important.We recommend complete surgical exicision and carrying out close monitoring after surgery due to the possibility of relapse and conversion to squamous cell carcinoma.

CONFLICT OF INTEREST

None.

REFERENCES

1. Gleich, T., *et al.*, Keratoacanthoma: a distinct entity? Exp Dermatol, 2016. 25(2): p. 85-91.
2. Hutchinson J. Morbid growths and tumours. The 'crateriform ulcer of the face', a form of acute epithelial cancer. Trans Pathol Soc London 1889; 40: 275–281.
3. Cabrijan, L., *et al.*, Differences between keratoacanthoma and squamous cell carcinoma using TGF-alpha. Coll Antropol, 2013. 37(1): p. 147-50.
4. Freeman, R.G., T.M. Cloud, and J.M. Knox, Keratoacanthoma of the conjunctiva: A case report. Archives of Ophthalmology, 1961. 65(6): p. 817-819.
5. Blessing, K., A.A. Nafussi, and P.M. Gordon, The regressing keratoacanthoma. Histopathology, 1994. 24(4): p. 381-384.
6. Tulvatana, W., P. Pisarnkorskul, and P. Wannakrairot, Solitary keratoacanthoma of the conjunctiva: report of a case. J Med Assoc Thai, 2001. 84(7): p. 1059-64.
7. Kifuku, K., *et al.*, Conjunctival keratoacanthoma in an Asian. Arch Ophthalmol, 2003. 121(1): p. 118-9.
8. Schellini, S.A., *et al.*, Conjunctival keratoacanthoma. Acta Ophthalmol Scand, 1997. 75(3): p. 335-7.
9. Bellamy, E.D., J.H. Allen, and N. Hart, Keratoacanthoma of the bulbar conjunctiva. Archives of Ophthalmology, 1963. 70(4): p. 512-514.
10. Perdigão, F.B., *et al.*, Conjunctival keratoacanthoma. Revista do Hospital das Clínicas, 2004. 59: p. 135-137.
11. Hughes, E.H., *et al.*, Keratoacanthoma of the conjunctiva. Eye, 2003. 17(6): p. 781-782.
12. Oellers, P., *et al.*, Conjunctival keratoacanthoma. British Journal of Ophthalmology, 2014. 98(2): p. 275-276.