Solitary Keratoacanthoma of the Conjunctiva: Report of a Case†

WASEE TULVATANA, M.D.*,
PETCH PISARNKORSKUL, M.D.*,
PONGSAK WANNAKRAIROT, M.D.**

Abstract

Keratoacanthoma characteristically occurs on the skin and is rarely found in the conjunctiva. We, herein, report a case of a healthy 41-year-old Thai woman presenting with a rapidly growing conjunctival mass. The tumor was excised to exclude squamous cell carcinoma. Histopathology revealed a cup shaped well-circumscribed proliferation of squamous cells, with a central keratin crater, consistent with a keratoacanthoma. On the literature review, only 12 cases have previously been reported in the English language. The mean age of presentation was 40.75 years, with a male preponderance. Presentation of a mass was mostly within 4 weeks after onset. History of foreign material getting into the eye was positive in 5 cases. All cases were successfully treated by excision, except for one case in which malignancy transformation was found, and enucleation was required eventually.

Conjunctival keratoacanthoma is a rare disease. However, this lesion should be well recognized and not misinterpreted as malignancy. As far as we are aware, this is the first report of conjunctival keratoacanthoma from this region.

Key word : Keratoacanthoma, Conjunctiva, Squamous Cell Carcinoma

**TULVATANA W,
PISARNKORSKUL P, WANNAKRAIROT P
J Med Assoc Thai 2001; 84: 1059-1064**

Keratoacanthoma was first described in 1950 by Rook and Whimster[1]. It is a common benign tumor on the cutaneous surface. The distincive finding is a rapidly growing dome-shaped nodule of 1.5-2 cm in diameter with a horn filled crater in its center. Most tumors spontaneously in-

* Department of Ophthalmology,
** Department of Pathology, Faculty of Medicine, Chulalongkorn University, Bangkok 10330, Thailand.
† Presented at the 6th Annual Meeting of the Royal College of Ophthalmologists of Thailand, Bangkok, Thailand, November 26, 1999.

volute after 6-8 weeks[2]. Although it is common on the skin, keratoacanthoma of the conjunctiva is extremely rare. We report a case of a conjunctival keratoacanthoma diagnosed and treated at the Department of Ophthalmology, Faculty of Medicine, Chulalongkorn University.

CASE REPORT

This case was a 41-year-old female farmer from the northeastern region of Thailand, presenting with a growth on her left temporal limbus. She reported that the mass developed rapidly within 2 weeks. She was otherwise healthy. She was not certain about any prior major eye injury, but recalled some dust getting into her eye. She went to a local clinic and was given steroid-antibiotic eye drops, which showed no improvement. The lesion quickly enlarged and caused irritation. On physical examination, a round, 5-mm conjunctival mass was seen on her left temporal limbal area. The mass had a smooth surface and protruded anteriorly, resembling a doorknob. (Fig. 1) The surface was dry because of the mass exposure, which could be demonstrated by presence of a mass between the upper and lower eyelids even with lid closure. (Fig. 2) It did not cause pain and could be freely moved over the sclera. Vascular dilatation around the mass was also seen.

The patient was treated by total excision of the mass including a 2-mm edge of normal looking conjunctiva. Gross pathology of the specimen revealed a button of conjunctival mass with a white cut surface. Microscopic examination showed a cup shaped configuration of markedly acanthotic conjunctival epithelium. (Fig. 3) Multiple keratin pearls were seen extensively within the central crater. Distinctive eosinophilic glassy cytoplasm was demonstrated in the proliferated squamous cells. (Fig. 4) There was no pleomorphism or any evidence of malignant changes. The basal layer of the epithelium was intact. Invasion into the substantia propria was not seen. Solar elastosis was demonstrated in the substantia propria as well as patchy inflammation. Surrounding conjunctiva was unremarkable. The pathological diagnosis of conjunctival keratoacanthoma was made. The patient was seen at 2 weeks post operation without recurrence. (Fig. 5) The patient reported no pain, irritation or mass recurrence after follow-up to 6 months.

On review of literature in the English language, there have been only 12 case reports of this entity. The summary of patients' demographic data, clinical manifestation, treatment, and recurrence are in Table 1. The mean age of presentation was 40.75 years (age range 25-65 years). Almost all of the reported lesions occurred in white patients.

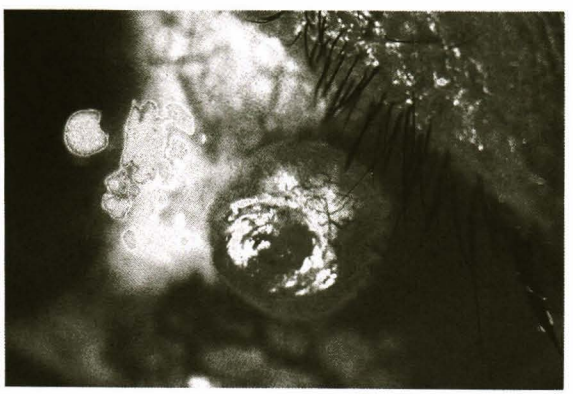

Fig. 1. Keratoacanthoma of the conjunctiva. The patient's left eye shows a well-circumscribed, raised, round, nodular lesion of the temporal limbus, resembling a doorknob. Conjunctival vascular dilatation around the mass is seen.

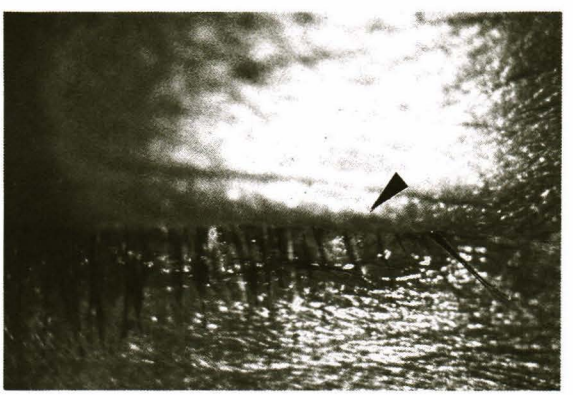

Fig. 2. Keratoacanthoma of the conjunctiva. The photograph shows a protruding mass (arrow) through lid closure causing exposure of the mass.

Fig. 3. **Keratoacanthoma of the conjunctiva. Histopathology shows a cup shaped configuration of acanthotic squamous epithelium. (Hematoxylin-eosin stain, original magnification 20X).**

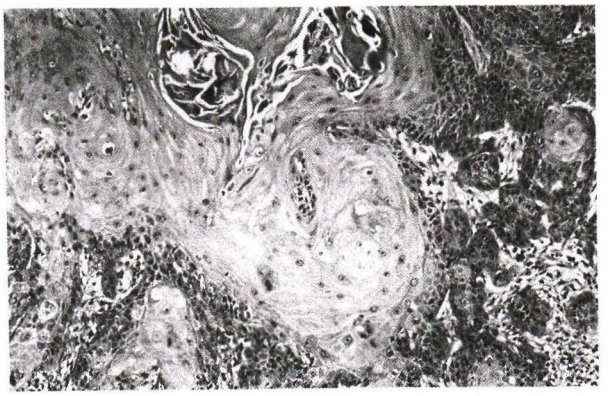

Fig. 4. **Keratoacanthoma of the conjunctiva. Histopathology, higher magnification, shows keratin pearls within the central crater. The cytoplasm of the proliferated squamous cells demonstrates typical eosinophilic glassy appearance. (Hematoxylin-eosin stain, original magnification 100X).**

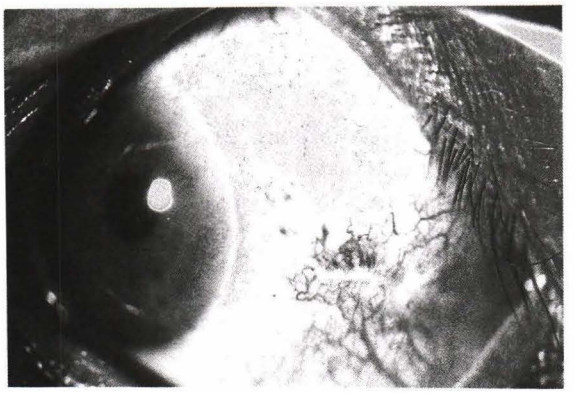

Fig. 5. **Keratoacanthoma of the conjunctiva, post-excision. The patient's left eye 2 weeks post-operatively shows well healed conjunctiva without recurrence of the mass.**

Gender predilection was male, with a male to female ratio of 5:1. Most of the patients (8/12) were outdoor workers. In most cases, the patient sought medical attention 2-4 weeks after disease onset. All patients reported having a rapid growing mass, and almost half of them (5/12) had a history of minor eye injury. All growths occurred on the bulbar conjunctiva. The location of the masses was usually on the temporal limbal area (8/12). Treatment by initial surgical excision was successful in 10/12 cases. Two cases demonstrated recurrence. One of these recurrent cases was successfully treated with additional excision. The other case was diagnosed later to have invasive squamous cell carcinoma and was eventually enucleated due to intraocular tumor invasion.

DISCUSSION

Conjunctival keratoacanthoma was first described by Freeman et al in 1961[3]. Only a small number of cases were described thereafter[4-11]. For the definite pathological diagnosis of keratoacanthoma, architecture of the lesion is as important as cellular characteristics[2]. Since it is difficult to make such a diagnosis unless a cup shaped configuration is obviously present, under-reporting maybe one of the explanations of the disease rarity.

Keratoacanthomas can be easily mistaken for squamous cell carcinoma since the clinical presentations and histopathologic findings are very similar. There were two conjunctival lesions in the literature misled to be squamous cell carcinoma, and pseudocarcinomatous hyperplasia, both of which were later believed to be keratoacanthoma[3,4]. The major difference in the clinical presentation is the rapid onset of the disease in keratoacanthoma.

Table 1. Keratoacanthoma of the conjunctiva: summary of patients' demographic data, clinical manifestation, treatment and recurrence.

Authors	Age	Sex	Race	Occupation	Onset (wks)	Symptoms	Eye	Site of tumor	History of injury	Treatment	Recurrence
1. Freeman et al[3]	55	M	White	Farmer	3	Mass	R	Temporal limbus	No	Wide excision	No
2. Freeman et al[3]	25	M	White	Unknown	Unknown	Mass	R	Nasal limbus	Unknown	Excision & beta ray	No
3. Walter[4]	57	M	White	Seaman	2	Mass	R	Temporal bulbar	Struck with a large hailstone	Wide excision twice	Yes
4. Bellamy et al[5]	28	M	White	Bulldozer operator	2	Mass & irritation	L	Nasal limbus	Dust getting into eye	Wide excision	No
5. Roth[6]	26	F	White	Ranch worker	4	Mass	R	Temporal limbus	Unknown	Wide excision	No
6. Roth[6]	37	M	White	Ranch worker	4	Mass	L	Temporal limbus	Unknown	Wide excision	No
7. Hamed et al[7]	49	M	White	Outdoor worker	4	Mass	R	Temporal limbus	Eardrops into eye	Wide excision	No
8. Hamed et al[7]	40	M	White	Outdoor worker	8	Mass	R	Temporal limbus	Struck with a piece of building siding	Wide excision	No
9. Grossniklaus et al[8]	65	M	Unknown	Unknown	2	Mass	L	Temporal limbus	No	Eventual enucleation	Yes
10. Munro et al[9]	42	M	Unknown	Unknown	3	Mass & irritation	L	Temporal limbus	No	Excision & cryotherapy	No
11. Schellini et al[10]	28	F	Mulatto	Housewife	4	Mass & irritation	L	Nasal limbus	No	Wide excision	No
12. Coupland et al[11]	37	M	Ethiopian	Civil engineer	2	Mass & irritation	L	Nasal limbus	Occur after grinding metal	Wide excision	No
13. Tulvatana et al	41	F	Thai	Farmer	2	Mass & irritation	L	Temporal limbus	Dust getting into eye	Wide excision	No

For histopathology, keratoacanthoma is typically cup shaped. Most proliferating cells are benign. Dysplastic epithelium and dyskeratosis may be seen, but no invasion is present. Multiple keratin pearls are the characteristic features, and contribute to the central keratin crater of the lesion.

The pathogenesis of keratoacanthoma is not known. The theory of viral genesis in cutaneous keratoacanthoma has not been confirmed. Exposure to carcinogens, sunlight, and trauma are not validated[2]. In this literature review, history of prolonged sun exposure strongly relates to conjunctival keratoacanthoma. Most patients were outdoor workers as well as the present patient, who is a farmer. Presence of solar elastosis of the substantia propria in this patient also supports the presumption. Almost half of the published cases noted minor eye injury. The history of dust getting into the eye in the present case is another example of insignificant eye injury. As a matter of fact, history of trauma might not be contributory to the lesion because there were no previous reports of actual detectable conjunctival injury both clinically and pathologically. The importance of trauma history is possibly what brings such lesions to the attention of the patients.

In contrary to skin lesions, conjunctival keratoacanthomas have never been reported to spontaneously involute. All cases were excised, partly because of the possibility of true malignancy and for a definite pathological diagnosis. Treatment by wide excision is usually adequate. Repeated recurrence is not usual unless the tumor undergoes malignant transformation.

For cutaneous keratoacanthoma, multiplicity of lesions is important since it could refer to a rare syndrome namely, Muir-Torre Syndrome. Multiple or eruptive conjunctival lesions without simultaneous skin lesions have never been reported. All of the earlier case reports were solitary conjunctival keratoacanthomas. Furthermore, none of the cases were associated with visceral malignancies as found in Muir-Torre syndrome.

SUMMARY

We report a typical case of solitary keratoacanthoma of the conjunctiva in a 41-year-old Thai woman. The disease is rare and generally occurs in middle aged outdoor workers. A rapid growth on temporal limbus is a common presenting feature.

Due to close resemblance to keratoacanthoma, squamous cell carcinoma should be considered as the primary differential diagnosis. However, excisional biopsy is generally sufficient to treat these lesions. To our knowledge, this is the first case report from this region.

(Received for publication on May 18, 2000)

REFERENCES

1. Rook A, Whimster IW. Le kératoacantome. Arch Belg Dermatol Syph 1950; 6: 137-46.
2. Kirkham N. Tumors and cysts of the epidermis. In: Elder D, Elenitsas R, Jaworsky C, Johnson B Jr, eds. Lever's histopathology of the skin. 8th ed. Philadelphia: Lippincott-Raven, 1997: 685-746.
3. Freeman RG, Cloud TM, Knox JM. Keratoacanthoma of the conjunctiva. A case report. Arch Ophthalmol 1961; 65: 817-9.
4. Walter WL. Pseudo-epitheliomatous hyperplasia of the conjunctiva. A lesion simulating squamous-cell carcinoma. A case report. Am J Ophthalmol 1962; 53: 999-1003.
5. Bellamy ED, Allen JH, Hart NL. Keratoacanthoma of the bulbar conjunctiva. Arch Ophthalmol 1963; 70: 512-4.
6. Roth AM. Solitary keratoacanthoma of the conjunctiva. Am J Ophthalmol 1978; 85: 647-50.
7. Hamed LM, Wilson FM 2nd, Grayson M. Keratoacanthoma of the limbus. Ophthalmic Surg 1988; 19: 267-70.
8. Grossniklaus HE, Martin DF, Solomon AR. Invasive conjunctival tumor with keratoacanthoma features. Am J Ophthalmol 1990; 109: 736-8.
9. Munro S, Brownstein S, Liddy B. Conjunctival keratoacanthoma. Am J Ophthalmol 1993; 116: 654-5.
10. Schellini SA, Marques MEA, Milanezi MFG, Bacchi CE. Conjunctival keratoacanthoma. Acta Ophthalmol Scan 1997; 75: 335-7.
11. Coupland SE, Heimann H, Kellner U, et al. Keratoacanthoma of the bulbar conjunctiva. Br J Ophthalmol 1998; 82: 586.

เคอราโทเอแคนโธมาของเยื่อบุตา: รายงานผู้ป่วย 1 ราย†

วสี ตุลวรรธนะ, พ.บ.,*
เพชร พิศาลก่อสกุล, พ.บ., พงษ์ศักดิ์ วรรณไกรโรจน์, พ.บ.***

เคอราโทเอแคนโธมาเป็นเนื้องอกผิวหนังที่พบบ่อยชนิดหนึ่ง แต่ที่เยื่อบุตากลับพบได้น้อยมาก รายงานผู้ป่วยนี้ได้กล่าวถึงผู้ป่วยหญิงไทยอายุ 41 ปี ที่มาพบแพทย์ด้วยเรื่องก้อนโตเร็วที่เยื่อบุตาซ้าย หลังจากผ่าตัดเอาก้อนเนื้อออกและส่งพิสูจน์ผลทางพยาธิวิทยา พบว่ามีการเพิ่มจำนวนของเซลล์เยื่อบุผิวเป็นรูปถ้วยตรงกลางถ้วยมีเคอราตินอยู่เต็ม เข้าได้กับเคอราโทเอแคนโธมา จากการทบทวนวรรณกรรมพบว่าก่อนหน้านี้มีรายงานผู้ป่วยเพียง 12 ราย อายุเฉลี่ยของผู้ป่วย 40.75 ปี ส่วนใหญ่เป็นเพศชาย ผู้ป่วยมักมาพบแพทย์หลังเริ่มมีอาการประมาณ 4 สัปดาห์ การซักประวัติพบมีสิ่งแปลกปลอมเข้าตา 5 ราย ผู้ป่วยเกือบทุกรายได้รับการรักษาให้หายขาดด้วยการผ่าตัดเอาก้อนเนื้องอกออก มีเพียง 1 รายที่ภายหลังพบว่ากลายเป็นเนื้อร้ายและต้องควักตาออกในที่สุด

เคอราโทเอแคนโธมาของเยื่อบุตาเป็นโรคที่พบได้น้อยมาก แต่จำเป็นต้องวินิจฉัยแยกโรคนี้ออกจากมะเร็งเยื่อบุชนิดสแควมัสเซลล์ การรายงานผู้ป่วยรายนี้เป็นรายงานแรกจากภูมิภาคนี้

คำสำคัญ : เคอราโทเอแคนโธมา, เยื่อบุตา, มะเร็งเยื่อบุชนิดสแควมัสเซลล์

วสี ตุลวรรธนะ, เพชร พิศาลก่อสกุล, พงษ์ศักดิ์ วรรณไกรโรจน์
จดหมายเหตุทางแพทย์ ฯ 2544; 84: 1059–1064

* ภาควิชาจักษุวิทยา,
** ภาควิชาพยาธิวิทยา, คณะแพทยศาสตร์ จุฬาลงกรณ์มหาวิทยาลัย, กรุงเทพ ฯ 10330
† นำเสนอในที่ประชุมวิชาการประจำปีราชวิทยาลัยจักษุแพทย์แห่งประเทศไทย กรุงเทพมหานคร ครั้งที่ 6 วันที่ 26 พฤศจิกายน 2542