Case Reports in Ophthalmology

Case Rep Ophthalmol 2017;8:535–538

DOI: 10.1159/000480725
Published online: December 5, 2017

Case Report

Lacrimal Caruncle Nevus with Papilloma

Eri Ishikawa Maria Suzanne Sabundayo Yasuhiro Takahashi
Hirohiko Kakizaki

Department of Oculoplastic, Orbital and Lacrimal Surgery, Aichi Medical University Hospital, Nagakute, Japan

Keywords
Lacrimal caruncle · Intradermal nevus · Papilloma

Abstract
Purpose: The aim of this article is to report a case of lacrimal caruncle nevus with papilloma. ***Methods:*** This is a case report of a 39-year-old female with a progressively enlarging pigmented lesion on the left lacrimal caruncle. She had been aware of a raised whitish wart on the top of this pigmented lesion for several months before her initial visit. Slit lamp examination revealed a papillomatous lesion over a well-circumscribed, pigmented lesion on the left lacrimal caruncle. ***Results:*** The histopathological examination of the excised tumor disclosed 2 characteristic findings, which include nests of nevus cells within the dermis and papillomatous structures which had fibrovascular cores overlying squamous cell epithelia with variable levels of acanthosis. The findings were consistent with an intradermal nevus and a papilloma arising from the conjunctival epithelium of the nevus. ***Conclusion:*** This is the first case report of a lacrimal caruncle nevus with papilloma. The clinical history and pathological findings of this case underscore the fact that an intradermal nevus primarily occurred on the lacrimal caruncle, after which a papilloma arose from the epithelium of the nevus as a consequence of human papillomavirus autoinoculation.

KARGER

Hirohiko Kakizaki
Department of Oculoplastic, Orbital and Lacrimal Surgery
Aichi Medical University Hospital
1-1 Yazako-Karimata, Nagakute, Aichi 480-1195 (Japan)
E-Mail cosme@d1.dion.ne.jp

Introduction

Various kinds of tumors arise from the lacrimal caruncle, of which nevus [1–5] and papilloma [6, 7] are the most frequent. However, only 1 case report has illustrated a compound caruncle tumor composed of a papilloma with scattered intradermal nevus cells [8].

We present the case of a lacrimal caruncle nevus with a papilloma, the pathology of which is different from the previous report [8].

Case Report

A 39-year-old female presented with a slowly progressing pigmented lesion on the left lacrimal caruncle. She had been aware of a raised whitish wart on the top of this pigmented lesion for several months before her initial visit. She had no history of a wart on the same site.

Slit lamp examination revealed a papillomatous lesion over a well-circumscribed, pigmented lesion on the left lacrimal caruncle (Fig. 1). The combined tumor was completely removed under local anesthesia. The histopathological examination disclosed 2 characteristic findings (Fig. 2), which include nests of nevus cells within the dermis and papillomatous structures which had fibrovascular cores overlying squamous cell epithelia with variable levels of acanthosis. Nevus cells and squamous epithelial cells showed no atypia. The findings were consistent with an intradermal nevus and a papilloma arising from the conjunctival epithelium of the nevus.

At the 6-month follow-up, there was no evidence of recurrence.

Discussion

We described the first case of a lacrimal caruncle tumor which contained 2 lesions, a nevus and a papilloma. Nevus and papilloma are the 2 most common tumors that independently arise from the lacrimal caruncle [1–7]. The frequency of these lesions ranges from 33.7 to 59.5% for nevus [1–5] and from 21.5 to 31.6% for papilloma [6, 7]. No report has, however, demonstrated the occurrence of a papilloma on a nevus as a combined lesion.

A nevus mainly has 3 pathological types depending on the location of the nevus cells: an intradermal nevus, located in the dermis, such as in this case; a junctional nevus, located in the dermoepidermal junction; and a compound nevus, involving both the dermoepidermal junction and the dermis [9]. While junctional and compound nevi may undergo malignant changes, an intradermal nevus shows a benign course as its epithelium remains intact [9].

Conjunctival papillomas, caused by human papillomavirus (HPV), have a tendency to be located medially especially on the lacrimal caruncle because of the tear flow direction (and hence the flow of HPV particles) from the lacrimal lake towards the medial conjunctiva [10]. Rubbing of eyes also involves the medial canthus, which may precede viral autoinoculation [10]. Most conjunctival papillomas (81%) harbor HPV, particularly low-risk HPV types 6 and 11 [11, 12], while normal conjunctival specimens are HPV negative [11], although hybrid capture II and polymerase chain reaction assays were not performed in this case.

As the epithelium of an intradermal nevus remains intact, the papilloma can arise from the normal caruncular epithelium in the intradermal nevus as a consequence of HPV autoinoculation. The clinical history and pathological findings of this case also underscore the de-

velopment of papilloma from an underlying intradermal nevus. On the other hand, it is unclear whether a papilloma can arise from an abnormal epithelium in a junctional or compound nevus.

In conclusion, we presented the first case of a lacrimal caruncle nevus with papilloma. Combined lesions should also be considered as a differential diagnosis for lacrimal caruncle lesions.

Statement of Ethics

This study was approved by the ethics committee and adhered to the tenets of the 1964 Declaration of Helsinki. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Disclosure Statement

The authors declare that they have no conflicts of interest.

Funding Sources

There was no funding for this article.

References

1 Luthra CL, Doxanas MT, Green WR: Lesions of the caruncle: a clinicopathologic study. Surv Ophthalmol 1978;23:183–195.
2 Santos A, Gómez-Leal A: Lesions of the lacrimal caruncle: clinicopathological features. Ophthalmology 1994;101:943–949.
3 Hirsch C, Holz FG, Tetz M, Volcker HE: Clinical aspects and histopathology of caruncular tumors. Klin Monbl Augenheilkd 1997;210:153–157.
4 Kaeser PF, Uffer S, Zografos L, Hamédani M: Tumors of the caruncle: a clinicopathologic correlation. Am J Ophthalmol 2006;142:448–455.
5 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;23:1004–1018.
6 Evans WH: Tumors of the lacrimal caruncle: a study of 200 collected cases. Arch Ophthalmol 1940;24:83–106.
7 Shields CL, Shields JA, White D, Augsburger JJ: Types and frequency of lesions of the caruncle. Am J Ophthalmol 1986;102:771–778.
8 D'Hermies F, Meyer A, Morel X, Halhal M, Vu A, Elmaleh C, Bourgade JM, Cisneros B, Behar-Cohen F, Parent De Curzon H, Renard G: Papillomatous and nevus of the caruncle (in French). J Fr Ophthalmol 2001;24:209–212.
9 Pe'er J: Pathology of eyelid tumors. Indian J Ophthalmol 2016;64:177–190.
10 Sjo N, Heegaard S, Prause JU: Conjunctival papilloma: a histopathologically based retrospective study. Acta Ophthalmol Scand 2000;78:663–666.
11 Sjö NC, von Buchwald C, Cassonnet P, Norrild B, Prause JU, Vinding T, Heegaard S: Human papillomavirus in normal conjunctival tissue and in conjunctival papilloma: types and frequencies in a large series. Br J Ophthalmol 2007;91:1014–1015.
12 Sjö NC, Heegaard S, Prause JU, von Buchwald C, Lindeberg H: Human papillomavirus in conjunctival papilloma. Br J Ophthalmol 2001;85:785–787.

DOI: 10.1159/000480725

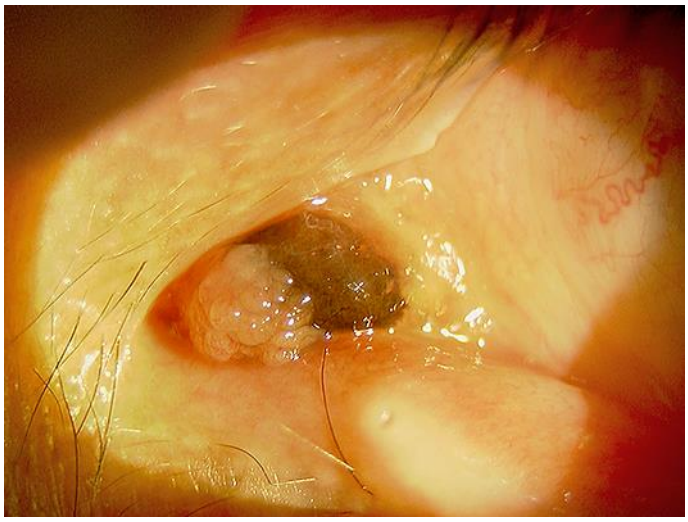

Fig. 1. Clinical appearance of the combined tumor on the left lacrimal caruncle. A papillomatous structure on the pigmented lesion is shown.

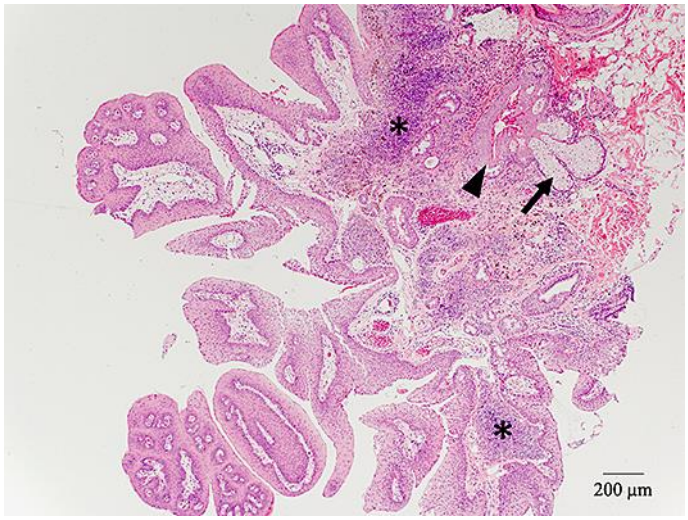

Fig. 2. Microscopic appearance of the combined lesion. Nests of nevus cells (asterisk) within the dermis with some pigmented melanocytes. The papilloma is composed of central fibrovascular cores covered by acanthotic squamous epithelium. Elements of the lacrimal caruncle, such as conjunctival epithelium with goblet cells, sebaceous glands (arrow), and hair follicles (arrowhead) are shown (hematoxylin and eosin stain; magnification ×75).