Case Report

Canine Bilateral Conjunctivo-Palpebral Dermoid: Description of Two Clinical Cases and Discussion of the Relevance of the Terminology

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Two young dogs were presented for the evaluation of an abnormally haired appearance of both eyes since adoption. In one dog, the lesions were symmetrical and appeared as disorganized skin tissue located on the cutaneous aspect of the lateral portion of both lower eyelids, and continuing to the palpebral and the bulbar conjunctiva, thus forming continuous lesions. In the other dog, a similar lesion was present in the right eye (OD), but the lesion of the left eye (OS) was of discontinuous, disorganized skin tissue located midway on the lower eyelid and on the lateral bulbar conjunctiva. The lesions were surgically removed and routinely processed for histopathological analysis. Definitive diagnosis was conjunctivo-palpebral dermoids for each dog. Dermoids are usually considered to be choristoma (normal tissue in an abnormal location) when they are located on the ocular surface (cornea and/or conjunctiva) and as hamartoma when located on the palpebral skin. The lesion presentation in these two dogs reveals that names of “choristoma” alone or “hamartoma” alone are not accurate to depict the continuous, composite, conjunctivo-palpebral dermoids. These cases suggest that choristoma and hamartoma might develop subsequently from the same abnormal event during the embryonic development, which means that the lesion location might be the only difference between the two terms.

1. Introduction

Dermoids result from the formation of histologically normal cutaneous tissue in an abnormal location during embryonic development [1–3]. These developmental anomalies are usually classified in the choristoma group [1]. The dermoid tissue has all of the characteristics of skin: an epidermis, dermis, fat tissue, sebaceous glands, hair follicles, and hairs [1]. Numerous types of ocular surface dermoid have been described in dogs [4–7], cats [8, 9], horses [10–12], and anecdotally in various other species [13–19]: corneal dermoid [4–7, 15, 19, 20], conjunctivo-corneal dermoid [9, 10, 16, 17], conjunctivo-palpebral dermoid [3, 20], and dermoid of the nictitating membrane [10]. Human ocular dermoids have also been described. They can be isolated lesions or a component of Goldenhar syndrome, which also affects other organs [21–24]. In dogs, the most frequent form is the conjunctivo-corneal dermoid [3]. However, the conjunctival-palpebral dermoid, for which a genetic predisposition has been identified, is a rare finding [3]. The predisposed breeds are German Shepherd, Dalmatian, and Saint-Bernard dogs [3]. In the latter breed, the presence of a dermoid is thought to be associated with other developmental anomalies such as eyelid coloboma [25]. The small number of cases described in the literature does not allow any particular sexual predisposition to be identified.
2. Clinical Cases

2.1. Case 1. A 4-month-old male German Shepherd dog presented with development of corneal pigmentation in both eyes (OU). The dog’s owners reported an abnormal ocular appearance in OU, associated with a copious mucopurulent discharge, since the dog’s adoption several weeks earlier. Medical treatment instituted by the referring veterinarian, of topical administration of fusidic acid ointment (Fucithalmic Vet, Léo) BID led to a temporary improvement in the dog’s symptoms. The general physical examination was unremarkable. An abundant mucopurulent ocular discharge was present bilaterally (Figure 1). Vision testing gave unreliable results. The pupillary response to light was difficult to establish due to the poor visibility of intraocular structures. Adnexal examination revealed the presence of haired skin tissue in the lateral conjunctival fornix. These hairs were numerous, approximately 3 cm long, and straight and were matted together by the chronic ocular discharge and oriented towards the external canthus. The lesion was approximately 8 mm in length, and was slightly elevated and separated from the corneo-scleral limbus by a 2 mm wide strip of bulbar conjunctiva in the temporal canthal region. It continued to reach the cutaneous aspect of the lateral lower eyelid and resulted in the absence of the lid margin in this area. The identified lesions were bilateral and symmetrical (Figure 2). The palpebral length was measured with calipers (Castroviejo compass): the upper eyelid length was 29 mm and the lower eyelid was 28 mm (4 mm laterally and 24 mm medially from the skin tissue) in length. Corneal examination with a slit lamp (SL 15, Kowa, Düsseldorf, Germany) revealed diffuse pigmentation of low density in the temporal quadrant of OU. Schirmer’s tear test (STT, MSD Santé Animal, Clermont-Ferrand, France) was within the normal range of OU. The rest of the ocular examination was unremarkable. The epidemiological data, combined with the characteristic clinical presentation, led us to the diagnosis of bilateral conjunctivo-palpebral dermoids. The corneal pigmentation, associated with signs of ocular discomfort and chronic ocular discharge, was considered a consequence of the irritation produced by the hairs growing from the dermoid.

2.1.1. Surgical Procedure. Surgical removal of the dermoid was undertaken. General anaesthesia was induced using intravenous administration of medetomidine (Domitor ND, Sogeval, Laval, 40 μg/kg), ketamine (Imalgene 1000 ND, Merial SAS, Villeurbanne, France, 5 mg/kg), and morphine hydrochloride (Morphine 10 mg, Lavoisier, Paris, France, 0.05 mg/kg). Anaesthesia was maintained using inhaled isoflurane (Isoflo ND, Axaence, Pantin, France, 1.8%) and oxygen following endotracheal intubation. The eyelids were routinely prepared using a povidone-iodine solution (Vetidine ND, Vetoquinol SA, Lures, France, diluted to 2%) after the hair was carefully cut. The conjunctival dermoid was incised around its periphery, at the limit of the lateral palpebral conjunctiva adjoining the external canthus and the bulbar conjunctiva close to the corneo-scleral limbus. The excision was continued ventrally on the cutaneous aspect of the eyelid with a V-shape incision, in order to achieve full excision of the dermoid and reconstruction of the lower palpebral margin. The resulting conjunctival defect was sutured in a simple continuous suture pattern using polyglactin 6-0 (Vicryl 6-0, Elanco, Neuilly-sur-Seine, France). A two-layered closure of the lower eyelid was performed. The palpebral conjunctiva was sutured using a simple continuous pattern of polyglactin 6-0 (Vicryl 6-0, Elanco, Neuilly-sur-Seine, France). The skin was then sutured with a simple interrupted pattern of nylon 5-0 (Prolene 5-0, Elanco, Neuilly-sur-Seine, France). The palpebral margin was restored by means of a “figure-of-eight” suture, using the same suture material. The same procedure was followed on each side.

The postoperative medical treatment consisted of topical administration of chloramphenicol ointment (Ophtalon ND, TVM, Lempdes, France) TID OU for a period of 2 weeks. The wearing of an Elizabethan collar was prescribed for one week, together with strict precautions related to the animal’s convalescence (short sanitary walks on a leash).

2.2. Case 2. A 1-year-old male French bulldog was referred with an abnormal ocular appearance in OU. The dog’s owners reported chronic profuse bilateral ocular discharge, an abnormally haired eyelid appearance, and a whitish lesion of the left eye since his adoption. Topical chloramphenicol ointment (Ophtalon pommade, TVM, Lempdes, France) prescribed
by the referring veterinarian for 2 weeks resulted in a slight reduction of the discharge.

The general physical examination was normal. Ophthalmic examination of the dog revealed slightly elevated haired tissue arising from the temporal palpebral conjunctival fornix OD, continuous with disorganised hair implantation on the adjacent skin. The palpebral margin was absent at the junction of the conjunctival and the cutaneous portion of the disorganised skin tissue. Pigmentary keratitis of the corneal region adjacent to the haired lesion was secondary to chronic irritation (Figure 3). A similar lesion was identified on the ventrolateral portion of the bulbar conjunctiva OS, with an area of disorganised skin tissue in the central portion of the lower eyelid, also involving the lid margin. The rest of the ocular examination revealed a corneal leukoma and anterior synechia OS, a consequence of a prior corneal perforation (Figure 4). Menace responses and dazzle reflexes were absent in OS and present in OD. The direct pupillary light reflex (PLR) was absent in OS and present in OD, and the indirect PLR was absent in OD and present in OS. The STT was within normal limits in OU. Intraocular pressure was 16 mm Hg OD and 15 mm Hg OS, as measured with a rebound tonometer (Tonovet, Icare, Helsinki, Finland). The clinical diagnosis was a conjunctival–palpebral dermoid affecting both eyes. The corneal scar OS was unrelated to these adnexal lesions.

2.2.1. Surgical Procedure. The anaesthesia and preparation of the patient were as described in Case 1. As the OD dermoid did not affect the bulbar conjunctiva, the surgery consisted of the removal of the temporal conjunctival fornix and the adjacent aberrant palpebral conjunctiva and skin. The reconstruction was carried out as described in Case 1. For the OS lesion, the disorganised skin tissue affecting the conjunctiva and the cutaneous palpebral aspect was not continuous and it required separated excisions. The abnormal tissue affecting the cutaneous aspect of the eyelid was resected with a simple V-shaped, full thickness incision to ensure removal of all hair follicles. The incision was closed in 2 layers with a continuous pattern (polyglactin 6-0 for the conjunctival repair and nylon 5-0 for the cutaneous repair). The palpebral margin was restored with a “figure-of-eight” suture using polyglactin 6-0. The isolated bulbar conjunctival lesion was removed by gentle dissection and then complete resection with straight Castroviejo scissors was carried. The bulbar conjunctiva was repaired using a continuous suture pattern of polyglactin 7-0. The excised tissues were routinely processed for histopathological analysis.

3. Histopathological Diagnosis (Figure 5)

The evaluated surgical excised samples consisted of fully differentiated skin tissue, regardless of their location (palpebral skin or conjunctiva) and were composed of multiple piloappendageal units, embedded in a fibrous connective tissue and deep adipose lobules. Overlying squamous stratified epithelium exhibited focal basal pigmentation, more pronounced in the conjunctival location, and variable amount of superficial keratinisation. These observations fully supported the microscopic diagnosis of conjunctivo-palpebral dermoid.

4. Follow-Up

Two weeks after surgery, the skin sutures were removed. A discreet cutaneous oedema limited to the lateral canthus was noticed on each side for both dogs. The palpebral margin of the lower eyelid was continuous. The owners noted that the dogs’ behaviour had improved and their behaviour was more playful and less irritable. A steroidal anti-inflammatory ointment (Fradexam ointment, TVM, Lempdes, France) was prescribed BID OU for 2 weeks. Four weeks after surgery, the patients showed no signs of discomfort. The dogs’ owners were satisfied with the cosmetic result (Figure 6) and the functional result, with complete resolution of any signs of ocular pain or irritation and a reduction in the degree of corneal pigmentation. There was no progression of the corneal leukoma OS of the second dog following surgery (Figure 7).
Figure 5: Case 2 histopathological image of excised lesion of OD, showing characteristics of normal skin. (a) At low magnification, it is impossible to differentiate the sample from another skin sample. Note the anatomical continuum from the left (eyelid hamartoma, L) to the right (conjunctival dermoid, R) and the conjunctival margin of the sample (#). Medium magnification shows pilosebaceous structures (∗) with hair shafts (→), sweat glands (S), and adipose lobules (A) on the cutaneous side (b) and conjunctival side (c) of the dermoid. High power aspect highlights differences between the keratinized epidermis (d) and the smooth, non-, or slight-keratinized thinner epidermis of the conjunctival side (e). Haematoxylin and eosin staining. Original magnification ×10 (a), ×40 (b, c), and ×200 (d, e).

Figure 6: Case 1 appearance three month after surgery. No sign of discomfort is observed.

Figure 7: Case 2 appearance OS, 1 month after surgery. A discreet oblique scar is still present on the lower eyelid. No sign of discomfort is observed.

5. Discussion

The physiopathological mechanism leading to dermoid formation is unknown. The most likely hypothesis is an abnormal differentiation of the surface ectoderm during embryonic development [1]. This tissue then develops into a segment, with all the histological characteristics of normal skin [1–3]. Although dermoids affecting the eye and its adnexa are mostly unilateral, some authors have reported cases of bilateral disease [6, 12–16, 21]. The hereditary nature of this affliction has been demonstrated in cats [8, 9], cows [14], and horses [11], as well as humans [24].

The conjunctival-palpebral dermoid in dogs is an uncommon form which has previously only been described in veterinary textbooks [1, 3] and not in peer reviewed clinical case reports. The most frequent location is at the external canthus [3], where newly formed tissue interrupts the continuity of the lid margin and continues into the conjunctival fornix [3]. In general, the length of the palpebral opening remains normal, but the presence of this tissue has functional and physical consequences that result from the presence of hairs [3].

In both of these cases, histopathological analysis of the removed modified tissues confirmed the diagnosis of dermoid. In cases of ocular surface dermoids (i.e., conjunctival or corneal dermoids), the skin tissue appears histologically normal but in an abnormal location, corresponding to the definition of choristoma [1]. Embryologically, choristoma results when one or two germ layers form mature tissue that is not normally found in that topographic location [1]. To this definition the notion of mass effect can be added [1]. In our
cases, the only difference between the conjunctival portion of the dermoid and normal skin is that the keratinization of the epidermis disappeared or significantly decreased, probably due to the lacrimal fluid that permanently bathed the skin. In cases of cutaneous palpebral dermoids, the location is normal, but the development of the skin is abnormal, with a larger number and size of skin components as compared to normal skin, corresponding to the definition of hamartoma [1, 26]. According to Dubielzig et al., hamartoma is an “excessive amount of mature tissue (hypertrophy and/or hyperplasia) occurring in a location in which tissue is usually found” [1]. The mass effect is also reported as a component of the definition of hamartoma [1].

Both eyes of Case 1 and OD of Case 2 presented with continuous conjunctivo-palpebral dermoid, thus forming composite lesions, whereas OS of Case 2 presented with two distinct lesions affecting the cutaneous portion of the eyelid and the conjunctiva. The abnormal skin island located on the lower eyelid of OS of Case 2 corresponds to the definition of hamartoma. The abnormal skin island located on the conjunctiva of the same eye corresponds to the definition of choristoma. However, we believe that the current accepted terminology inadequately describes the entire composite lesion (OU of Case 1 and OD of Case 2). Indeed, the cutaneous portion of the lesion occurs in a normal location, excluding the term “choristoma.” In comparison, the term “hamartoma” does not fit with the conjunctival portion of the lesion, because the anomaly occurs in a location in which skin tissue is usually not found. To the best of our knowledge, there is no specific or accurate term to define the entire cutaneoconjunctival lesion. Both parts of the lesion are probably secondary to the same abnormal event during embryonic development, affecting two adjacent areas of different nature (e.g., the skin and the conjunctival mucosa). The coexistence of distinct separated lesions on OS of Case 2 and continuous cutaneoconjunctival lesion on OD of the same patient make us hypothesize that choristomas and hamartomas may be due to the same developmental process, meaning that the notion of location might be the only difference between the two terms.

The use of the term “dermoid” also seems controversial. For some authors, dermoids are strictly defined as choristomas [1, 2, 4] and cannot be applied for a skin location, whereas for some others [3], by usage and extension, the term “dermoid” can be used for either hamartomas (when occurring on the eyelid) or choristomas (when occurring on the eye surface). In our opinion, as illustrated in these two cases with continuous cutaneoconjunctival lesions, the developmental abnormality and biologic mechanism causing the lesion are probably the same. This is the reason why the term “dermoid” remains accurate for the three possible entities: strictly palpebral lesions, strictly conjunctival (or corneal) lesions, or composite conjunctivo-palpebral lesions with a physical continuum. For this last entity, as the nature of the lesion (dermoid) cannot be associated with “hamartoma” alone or “choristoma” alone, we propose to describe it as a “choristohamartoma.”

Independently from the terminology, the clinical presentation of such a lesion is comparable with that observed in cases of conjunctival of corneal dermoid. Hairs chronically traumatize the ocular surface, leading to superficial inflammation characterised by conjunctivitis, corneal neovascularisation, and pigmentation [3]. This abrasion is painful for the patient and provokes a chronic ocular discharge as well as an intense blepharospasm. The patient’s abnormal ocular appearance and pain are usually the primary reasons for consultation [3].

The treatment is always surgical and obviously requires resection of the malformed area, sometimes associated with graft procedures [3, 27–29]. During the surgery, particular care must be taken in reconstruction of the palpebral margin in order to conserve palpebral function and remove any risk of iatrogenic ocular irritation. When the palpebral margin deficit is small following resection, a simple edge-to-edge suture can be used, with continuity of the margin being ensured by means of a “figure-of-eight” suture [3]. If the resection leads to a more significant deficit, a more complex oculoplastic procedure is indicated [3]. When the developmental anomaly is severe and has serious consequences (ocular pigmentation or even ulceration and chronic ocular pain), early surgery is recommended [3]. In the case of more discrete conditions having less significant physical and functional consequences, surgery can be carried out at the age of three months [3]. In Case 1, the condition was serious and caused considerable pain as well as intense corneal pigmentation, but surgery was performed at the age of 4 months, following the referring veterinarian’s unsuccessful medical treatment. In both cases, the malformation was characteristically located in the outer canthal region except in OS of Case 2. Although it covered a significant portion of the conjunctival fornix, alteration of the palpebral margin was minimal, and so simple resection and classical edge-to-edge reconstruction were sufficient to permanently remove the source of irritation, restore normal palpebral function, and result in the rapid resolution of discomfort and recovery of the dog’s normal temperament.

In conclusion, despite the simple but uncommon clinical presentation of these two cases, they illustrate the limitation of the terminology to describe development of abnormalities affecting the ocular surface and adnexa.

**Conflict of Interests**

The authors declare that there is no conflict of interests regarding the publication of this paper.

**References**


