Journal of College of Medical Sciences-Nepal, Vol-21, No.2, Apr-Jun 2025

ISSN: 2091-0657 (Print); 2091-0673 (Online) Open Access
DOI: 10.3126/jcmsn.v21i2.76611

Recurrent Hyphema After Periocular Dog Bite in A 3-Year-Old: A Diagnostic Enigma

Kumudini Subedi[1], **Sangita Pradhan**[1], **Maria Gautam**[2], **Niva Tiwari**[3], **Raghunandan Byanju**[4]

[1]Department of Uveitis and Vitreoretinal Service, Bharatpur Eye Hospital, Bharatpur, Chitwan, Nepal, [2]Department of Oculoplasty, Bharatpur Eye Hospital, Bharatpur, Chitwan, Nepal, [3]Department of Paediatric Oncology, B. P. Koirala Memorial Cancer Hospital, Bharatpur, Chitwan, Nepal, [4]Department of Ophthalmology, KIST Medical College, Imadole, Lalitpur, Nepal.

ABSTRACT

Background

Dog bites have been reported to cause destructive and avulsive ocular injuries. There have been rare instances of intraocular effect with an intact globe. Hyphema in such context must be dealt with a high degree of clinical suspicion.

Case: A 3-year-old male was brought to the department with history of dog bite to the lateral aspect of left orbit following which his guardians noted persistent redness of the ipsilateral eye. Clinically, grade IV hyphema was noted which persisted even after anti-rabies prophylaxis was completed. Ultrasonography noted few vitreous opacities with no mass lesion. Hyphema was washed. Iris neovascularization with ectropion uveae was noted. Diffuse posterior segment abnormality was seen with no exact pathognomonic feature on table. The child returned with similar hyphema with clot in the anterior chamber with no other inciting factors 3 weeks later. Repeated imaging was misleading towards the exact diagnosis and the eyeball was enucleated whereby the histopathology established the exact diagnosis.

Conclusion: A variety of clinical diagnosis must be kept in mind when a suspicious clinical scenario presents. The vision as well as the life of the patient may be at stake when sinister diagnoses are not kept in mind.

Keywords: dog bite; hyphema; iris neovascularization; ectropion uveae.

Correspondence: Dr. Kumudini Subedi, Department of Uveitis and Vitreoretinal Service, Bharatpur Eye Hospital, Bharatpur, Chitwan, Nepal. Email: subedikumu@gmail.com, Phone: +977-9845100057. **Article received**: 2024-02-09. **Article accepted**: 2025-05-25. **Article published**: 2025-06-30.

INTRODUCTION

Dog bites are common causes of emergency room visits in the United States. Dog bites occur in over 1 million Americans a year with over 300,000 emergency room visits and result in significant morbidity and even mortality. In Nepal, dog bites account for more than 90% of the total cases of animal bite. According to Sukraraj tropical and infectious disease hospital, 150 people visit the hospital each day to receive anti-rabies vaccination. National data recorded 33204 cases of dog bite in the year 2017-2018. Many cases are not documented and there may still be many more in reality. These injuries can result in a diverse set of periocular complications including damage to the canaliculi, facial nerve, levator muscle, lacrimal gland, rectus muscles, bony orbit and globe.[1-4] Rarely ocular injuries from dog bites cause infections, reported as 2% only in a large case series of 91 cases of ocular dog bite injuries over a 10-year period (2003-2013).[5] Spontaneous hyphema in children is uncommon, the most common cause of hyphema per se being trauma. Previous case reports state that spontaneous hyphema may occur in retinoblastoma, retinoschisis, retinopathy of prematurity, persistent primary hyperplastic vitreous, blood dyscrasias, and certain other less frequent conditions including neoplasms (medulloepithelioma, and leukemia).[6] Spontaneous hyphema is also a common presenting sign of juvenile xanthogranuloma of the iris.[7] We consider a 3-year-old child with hyphema and a history of dog bite to the periorbital region just a day before presentation to hospital, a very rare and suspicious clinical scenario.

Figure 1. Grade 4 hyphema in left eye with dog bite scars in temporo-zygomatic region.

CASE REPORT

A three-year-old thin built child from a remote village in Gorkha presented to the pediatric eye department of our hospital 24 hours after a dog bite incident by a stray dog to the temporal-zygomatic region of the left orbit. The child was playing at a neighbor's house when the incident occurred and the parents did not witness the injury. The child was born at full term with no other ocular symptoms noted before the present incident. There was no relevant family history associated. The pediatric department noted a total hyphema in the ipsilateral eye and performed gentle B scan ultrasonography of the eye which was inconclusive except for few vitreous opacities.

The patient was then sent for anti-rabies prophylaxis as the dog was stray and had been killed by the villagers after the incident. The child returned 14 days later, after completion of anti-rabies prophylaxis, with a persistent grade III hyphema (Figure 1). Ultrasonography of eye showed mild retinal thickening with no pathognomonic features. Hence, he was planned for hyphema wash and proceed.

Hyphema wash was done which revealed neovascularization of the iris 360 degrees, producing ectropion uveae. There were no active bleeding sites. Pupil was fully dilated and the red glow was dull. Indirect ophthalmoscopy revealed multiple patches of cheesy exudates on the surface of the retina and multiple tortuous dilated vessels in between. The presentation was suspicious as there was no hypopyon associated yet the multiple minute masses looked like fungal colonies however no other signs were suggestive of fungal endophthalmitis. Due to suspicious presentation and grossly poor visual prognosis as the whole of the retina was involved, vitrectomy was not planned at the same sitting. The provisional diagnoses were retinoblastoma, infantile coats, followed by xanthogranuloma of iris, chronic uveitis, or a masquerade syndrome. Blood investigations were sent to rule out any form of uveitis or likely leukemia. All blood investigations were

normal. The guardians of the child were counseled regarding grave visual prognosis and the need for further imaging for investigation. The parents were economically strained and lost to follow up. Child was brought back on repeated beckoning by phone call after 2 weeks with a grade IV hyphem and clot in the anterior chamber following which CT scan was done which revealed enhancing non-calcified mass confined to left globe with subtle infiltration into vitreous with mild retinal detachment and involvement of the intraocular/intraorbital part of the left optic nerve. As this was suspicious for either retinoblastoma or Coat's disease, pediatric oncologists were consulted regarding the reports which they stated were still inconclusive to start treatment. After repeated multi-modal imaging at different time frames weeks apart and multiple radiologist's consultations, very minute nidus of suspected calcification were suspected on MRI. The guardians of the patient were informed that the eye had no visual potential and likely harbored a sinister lesion that may eventually be life-threatening. It was a very difficult decision on the part of treating ophthalmologists as the evidence was not conclusive but even more difficult on the guardians side as it involved many social issues. After approval, enucleation of the eye ball was done as per protocol and was sent for histopathological evaluation. Biopsy revealed retinoblastoma grade 4 with a maximum tumor dimension 1.4 cm at the optic nerve head and full thickness choroidal invasion with diameter of invasion 1mm. The retro-laminar portion of the optic nerve was invaded by the tumor but surprisingly iris and ciliary body were tumor free. The sclera was also free of tumor and the cut end of the optic nerve was also free of tumor. The tumor was assigned "pT3b not assigned" status as per biopsy reports and child was started on systemic chemotherapy under pediatric oncologist.

DISCUSSION

Spontaneous hyphema in a very young child should be considered an alarming clinical sign. A large case series of Massachusetts Eye and Ear Infirmary was the first to delineate the causes of spontaneous hyphema in children. The most common causes of hyphema in childhood were xanthogranuloma of the iris, retinoblastoma, retinoschisis, retrolental fibroplasia, persistent primary hyperplastic vitreous, blood dyscrasias.[6] In an attempt to delineate other causes of spontaneous hyphema in childhood, two cases were studied by Howard et al., of the two cases with similar presentation, one had metastasis into the iris from a primary lesion in the other eye and another had iris discoloration which was seeding from a retinoblastoma of the same eye.[6] However, in the present case, there was no mass lesion visible on the operating table or grossly identifiable features on standard imaging modalities including CT scan and MRI. The above reported case is considered as diffuse infiltrating retinoblastoma. The first report of diffuse infiltrating retinoblastoma was in 1989.[8] Diffuse infiltrating retinoblastoma comprises only 2% of the total retinoblastoma cases reported.[9]

Retinoblastoma is known to be the most common intraocular tumor of childhood. The major presentations of retinoblastoma vary through age. Children under 5 years of age present most commonly with leukocoria or strabismus.[10,11] Case reports published before 2000 report spontaneous hyphema as a presentation of retinoblastoma but there are very few cases where the hyphema associated with retinoblastoma is not associated with mass lesion.[12-14] A literature search and publication done in 2016 concerning diffuse infiltrating retinoblastoma considering English, German and Spanish cases and case series identified 77 patients in twenty-four case reports and 6 case series.[9,15-19] The above article defined diffuse infiltrating retinoblastoma as having no clear-cut definition but having broad consensus on a flat infiltration of the retina, with small tumor mass. Diffuse spread of tumor cells in the vitreous, iris, trabecular meshwork and anterior chamber often occurs. In our present case, although gross iris neovascularisation was seen and ectropion uveae was noted, histopathology reports stated no involvement of the iris or the ciliary body even though full thickness of the choroid was invaded by the tumor with diameter of choroidal invasion being

1mm). Of the 77 eyes reported in the above literature, the common presentations were vitreous cells (79%), pseudo-hypopyon (48%) and an increased intraocular pressure of more than 21 mm Hg (43%).[9, 15-19] Of those cases, although iris neovascularisation was reported in 34% cases, no cases were reported to present with hyphema.[9,15-19] Corroborative to the literature presented in the above article, the patient discussed in this article was male (male predominance 62%), had unilateral involvement (unilateral in 92% cases) and had no family history (family history absent in 96%). A remarkable aspect of this case and similar cases was the questionable diagnosis until the histopathological test reports were available. Earlier reports state that upto 56% cases of diffuse infiltrative retinoblastoma are misdiagnosed. Previous reports state that the differentials have included posterior uveitis, Toxocara canis, Endophthalmitis, Trauma and coats disease.[20,21] Similarly, in this case also we had a problem in confirming the diagnosis especially with fungal endophthalmitis as there was no spectator to the injury mechanism and even intraocular findings after hyphema wash simulated fungal colonies in the retina.

CONCLUSIONS

Retinoblastoma is a sinister lesion that may affect not only the vision and vision related quality of life. With an array of diverse clinical presentations, a high degree of clinical suspicion must be maintained for diagnosis and treatment.

Limitations

Economic constraints on the patients' family limited the genetic testing of the child so the exact heredity pattern could not be established. It would have been beneficial in terms of prognosis of the second eye as there are reports of second eye being involved even after 18 years.[22]

Patient Consent

Written consent to publish this case has not been obtained. This report does not contain any personal identifying information.

ACKNOWLEDGEMENTS

Authors would also like to acknowledge Dr. Kshamata Subedi for correspondence with patient party and help with data collection.

Conflict of interest: None

Funding: None

REFERENCES

1. Erickson BP, Feng PW, Liao SD, Modi YS, Ko AC, Lee WW. Dog bite injuries of the eye and ocular adnexa. Orbit (London). 2019. [DOI]
2. Weiss HB, Friedman DI, Coben JH. Incide nce of dog bite injuries treated in emergency departments. JAMA 1998;279:51-3. [DOI]
3. Bratton EM, Golas L, Wei LA, Davies BW, Durairaj VD. Ophthalmic Manifestations of Facial Dog Bites in Children. Ophthalmic Plast Reconstr Surg 2018;34:106-9.[DOI]
4. Hospital receives 150 Dog Bite Cases Per Day. [Link]
5. Prendes MA, Jian-Amadi A, Chang SH, Shaftel SS. Ocular Trauma From Dog Bites: Characterization, Associations, and Treatment Patterns at a Regional Level I Trauma Center Over 11 Years. Ophthalmic Plast Reconstr Surg. 2016 Jul-Aug;32(4):279-83. [DOI] [PMID]
6. Howard GM. Spontaneous Hyphema in Infancy and Childhood. Arch Ophthalmol. 1962;68(5):615–620. [DOI]
7. Samara WA, Khoo CT, Say EA, et al. Juvenile Xanthogranuloma Involving the Eye and Ocular Adnexa: Tumor Control, Visual Outcomes, and Globe Salvage in 30 Patients. Ophthalmology 2015;122(10):2130-8. [PMID] [DOI]
8. Mansour AM, Greenwald MJ, O'Grady R. Diffuse infiltrating retinoblastoma. J Pediatr Ophthalmol Strabismus. 1989 May-Jun;26(3):152-4. [DOI] [PMID]
9. Shields CL, Ghassemi F, Tuncer S, Thangappan A, Shields JA. Clinical spectrum of diffuse infiltrating retinoblastoma in 34 consecutive eyes. Ophthalmology. 2008; 115: 2253–2258. [PubMed]
10. Claudia Carolina Cruz-Gálvez, 1 Juan Carlos Ordaz-Favila, 2 Víctor Manuel Villar-Calvo,

3 Martha Edith Cancino-Marentes, 4 and Vanessa Bosch-Canto; Retinoblastoma: Review and new insights. Front Oncol. 2022; 12: 963780. Published online 2022 Nov [DOI] [PMCID] [PMID]

11. Grossniklaus HE. Retinoblastoma. fifty years of progress. the LXXI Edward Jackson memorial lecture. Am J Ophthalmol (2014) 158(5):875–91. [DOI]
12. Hyphema revealing retinoblastoma. Saraux H, et al. Bull Soc Ophtalmol Fr. 1974.[PMC]
13. Retinoblastoma presenting with spontaneous hyphema and dislocated lens. Byrnes GA, et al. J Pediatr Ophthalmol Strabismus. 1993. [PMID]
14. Retinoblastoma presenting as a total hyphema. Appelboom T, et al. Ann Ophthalmol. 1985. [PMID]
15. Bhatnagar R, Vine AK. Diffuse infiltrating retinoblastoma. Ophthalmology. 1991; 98:1657–1661. [Google Scholar]
16. Brisse HJ, Lumbroso L, Fréneaux PC, Validire P, Doz FP, Quintana EJ, et al. Sonographic, CT, and MR imaging findings in diffuse infiltrative retinoblastoma: report of two cases with histologic comparison. Am J Neuroradiol. 2001;22:499–504. [Google Scholar]
17. Nicholson DH, Norton EWD. Diffuse infiltrating retinoblastoma. Trans Am Ophthalmol Soc. 1980;78:265–289. [PMC]
18. Morgan G. Diffuse infiltrating retinoblastoma. Br J Ophthalmol. 1971;55:600–606. [DOI]
19. Flick H, Schwab B. Das infiltrativ wachsende Retinoblastom-eine schwierige Differentialdiagnose. Klin Mbl Augenheilk. 1980;177:220–224. [DOI]
20. Mansour AM, Greenwald MJ, O'Grady R. Diffuse infiltrating retinoblastoma. J Pediatr Ophthalmol Strabismus. 1989 May-Jun;26(3):152-4. [DOI] [PMID]
21. Traine PG, Schedler KJ, Rodrigues EB. Clinical Presentation and Genetic Paradigm of Diffuse Infiltrating Retinoblastoma: A Review. Ocul Oncol Pathol. 2016 Apr;2(3):128-32.[DOI] [PMID] [PMCID]
22. Khanfir K, Chompret A, Frau E, Bloch-Michel E, Tursz T, Le Cesne A. An unusual variant of diffuse retinoblastoma in an adult. Acta Oncol. 2008;47:973–974. [DOI]

Citation: Subedi K, Pradhan S, Gautam M, Tiwari N, Byanju R. Recurrent Hyphema After Periocular Dog Bite in A 3-Year-Old: A Diagnostic Enigma. JCMS Nepal. 2025; 21(2): 201-205.