Case Report

Epistaxis-related hemolacria in a 6-year-old female after midfacial trauma: Epistaxis-related hemolacria

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ABSTRACT

Hemolacria (HL) is a rare condition characterized by the presence of blood in the tears. Bleeding associated with tears can occur in the context of infection, inflammation, trauma to the eye or surrounding structures, vascular tumors of the eye or surrounding structures, or retrograde epistaxis. We herein present a pediatric case with HL in the setting of epistaxis due to post-traumatic vessel injury without fracture, along with a literature review. Bleeding from the inferior lacrimal punctum was controlled by successful cauterization of damaged vessels and follow-up has remained uneventful for over a year. Bleeding from the eye is an alarming symptom, especially if it developed after facial trauma. However, physicians should be more aware that, in the absence of a fracture, HL due to retrograde epistaxis resulting from facial trauma can be a self-limited condition that is easily treated by controlling nasal hemorrhage.

Key words: Hemolacria, Epistaxis, Vessel injury, Midfacial trauma

emolacria (HL) is a rare condition characterized by the presence of blood in the tears. Bleeding associated with tears can occur in the context of infection, inflammation, vascular tumors, or trauma to the eye. The source of bleeding may also be the puncta [1-3]. Due to the intimate connection of nose and eye via the lacrimal apparatus, an increase in pressure within the nasal cavity can cause retrograde blood flow through the system and lead to bloody tears emerging from the ipsilateral punctum. Post-traumatic fractures of the nasal bone or walls of the sinuses, particularly Le Fort fracture type 1, cause HL as a commonly associated injury [4]. Hematologic conditions (coagulopathies) and medications (anticoagulants) may also cause bloody tears [5]. In cases with no organic cause of bleeding, Munchausen syndrome should be considered [6]. To date, there have been no previously reported cases of HL associated with epistaxis due to pure post-traumatic vascular injury.

CASE PRESENTATION

The presented case here is a 6-year-old female patient with an unremarkable past medical history. The patient fell from a height of 1 m on her nose and this triggered a massive nosebleed in the right nostril (Fig. 1). The application of direct pressure on the bleeding point by pinching the nose resulted in bleeding from

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the ipsilateral eye. Therefore, she was brought to the pediatric emergency department with complaints of bleeding from both nose and eye.

The physical examination of the child was unremarkable with normal blood pressure. No lesion of the eye or lids was present. Detailed ocular assessment including the orbital cavity revealed no abnormality.

Radiological studies of the area were unremarkable. An endoscopic examination of the nasal cavity revealed several injured vessels. A full hematological assessment was carried out to exclude hemostatic defects. The platelet count, prothrombin time, activated partial thromboplastin time, plasma fibrinogen concentration, vWF assay and F8 levels, PFA-100 test for platelet dysfunction, and serum clotting factor levels revealed normal results.

She had successful cauterization of the bleeding vessels and was discharged with complete recovery. During the 2-year follow-up, she had further three episodes of epistaxis, each associated with ipsilateral HL, and hence required additional cauterization. A meticulous assessment of the nasal cavity revealed no vascular malformations. There was no bleeding recurrence for a year. When last seen a week ago, no intranasal lesions were detected.

DISCUSSION

In this case, HL developed after midfacial trauma without a fracture. Blunt facial traumas cause Le Fort fractures which

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Figure 1: Hemorrhage from the right lacrimal puncta. Photograph reproduced with the written consent of the patient's parents

Table 1: Literature review of reported cases of hemolacria associated with epistaxis

Previous studies	Age/ Gender	Epistaxis	Management of epistaxis	Hemolacria	Additional bleeding	Underlying clinical condition	Medication	Hemostatic defect	Prognosis
Abel [7]	78/Male	Left nostril	Resolved itself	Left lacrimal punctum	Ø	Alcoholism	Ø	Ø	Resolved after digital pressure over the sac
Drake and Packer [8]	81/Male	Right nostril	Rhino rocket nasal tamponade	Right lacrimal punctum	Ø	End-stage renal disease	Aspirin	Ø	Resolved after treatment with intranasal oxymetazoline
Wiese [9]	56/ Female	Right nostril	Pinching her nose	Right lacrimal punctum	Bloody otorrhea	Not described	Ø	Ø	Controlled by nasal sponge tamponade
Algoraini et al. [10]	12/ Female	Location not reported	Subsided by itself	Bilateral lacrimal punctum	Ø	Psychological stress	Ø	Ø	Resolved spontaneously
Kosałka- Węgiel et al. [11]	18/Male	Location not reported	Glucocorticoids and antihistaminic drug	Bilateral lacrimal punctum	Bloody otorrhea, hemoptysis, hematuria	Irritant exposure	Ø	Ø	Excellent clinical prognosis by avoiding inhaled irritants
Humber et al. [12]	17/Male	Right nostril	Nasal precautions and observation	Right lacrimal puncta	Ø	Orthognathic surgery	Ø	Ø	Resolved spontaneously
	41/ Female	Right nostril	Nasal packs	Right lacrimal puncta	Ø	Orthognathic surgery	Ø	Ø	Controlled by nasal packs
Present report	6/Female	Right nostril	Pinching her nose	Right lacrimal punctum	Ø	Intranasal vessel injury after midfacial trauma	Ø	Ø	Recovery after successful cauterization of bleeding vessels

are associated with other severe injuries to the head and neck, including HL. The most common mechanism of HL seen in facial trauma includes Le Fort fracture type 1, in which the fracture line passes through the alveolar ridge, lateral nose, and inferior wall of the maxillary sinus [4]. Apart from fractures, facial trauma can rarely cause ocular bleeding through damage to the apparatus or nasal cavity. For example, a rupture in the apparatus (sac, ductus, or valves) may lead to punctate bleeding or damage to the intranasal vessels, particularly maxillary arteries or their terminal branches, which may result in retrograde epistaxis and subsequent HL (as suspected in our case). Pinching or blowing the nose to manage epistaxis may further intensify HL by augmenting the intranasal pressure.

Anecdotal evidence may suggest that epistaxis-related HL is commonly encountered. However, to date, only a few cases have been reported and none of the reported cases were associated with post-traumatic vessel injury. Abel reviewed these cases for the first time in 1950 [7], looking at all the cases reported between 1916 and 1947 and presenting a new case in detail. The clinical condition was not yet denoted as HL, instead expressed as "ocular hemorrhage," "bleeding from the eye," "hemorrhage from the lacrimal punctum," or "bleeding from the lacrimal duct." In this review, it was clearly emphasized that ocular hemorrhage was not related to the sac or ductus; the bleeding did not originate from the eye per se; instead, it was associated with epistaxis. In accordance, subsequent publications have preferred the phrase "HL associated

with retrograde epistaxis." In this situation, HL may develop due to the original cause of epistaxis, the increased intranasal pressure by epistaxis, or during the management of epistaxis. For example, in the patient by Drake and Packer [8], HL developed after the insertion of a "Rhino Rocket" nasal tamponade device to treat persistent epistaxis. Epistaxis is idiopathic in some cases [9] or may be secondary to underlying conditions with systemic effects (e.g., end-stage renal disease, psychological stress, irritant exposure, and alcoholism) [7,8,10,11] and to maxillary orthograthic surgery [12]. The two patients by Humber et al., after having orthognathic surgical correction of Class III malocclusion, presented with complaints of epistaxis and retrograde hemorrhage from ipsilateral nasolacrimal puncta, one on the 8th day postoperatively and the other early on after her transfer to the post-anesthesia care unit [12]. Table 1 shows the summary of the previously reported cases. In our case, vessel injury due to facial trauma caused epistaxis, epistaxis increased the intranasal pressure leading to retrograde blood flow, and pinching the nose further increased the retrograde epistaxis leading to punctate drainage. The review of cases also shows that ocular bleeding is on the same side as nosebleeds (ipsilateral), drainage is via the punctum (particularly the lower punctum), and the condition is self-limited despite its scary appearance. All of the reported cases have made a complete recovery. Of note, the recurrence of bleeding observed in our case might be due to the development of target vessels following traumatic injury and the bilateral nature of HL observed in cases by AlGoraini et al. and Kosalka-Wegiel et al. might be related to the underlying systemic conditions.

CONCLUSION

Regardless of the reason, bleeding from the eye is very concerning for both the patient and the physician. Among multiple underlying etiologies, epistaxis-related HL even resulting from facial trauma is usually self-limited and can be treated by controlling nasal hemorrhage. This report draws attention to this rare but alarming clinical entity to increase awareness and help to provide enhanced outcomes for affected patients.

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REFERENCES

- Tripathy K, Salini B. Hemolacria. In: StatPearls. Treasure Island, FL: StatPearls Publishing; 2022. Available from: https://www.ncbi.nlm.nih.gov/books/NBK539774 [Last accessed on 2022 Dec 04].
- Haemolacria (Bloody Tears). Available from: https://anaheimeyemd.com/ haemolacria-bloody-tears [Last accessed on 2022 Nov 21].
- Di Maria A, Famà F. Hemolacria crying blood. N Engl J Med 2018:379:1766-9.
- 4. Phillips BJ, Turco LM. Le fort fractures: A collective review. Bull Emerg Trauma 2017;5:221-30.
- Nandakumal G, Ismail F, Mohamad NF, Lott PW, Chew KS, Ab Rahman S, et al. A rare case of von willebrand disease presenting as hemolacria and literature review. J Pediatr Hematol Oncol 2021;43:101-3.
- Audelan T, Best AL, Ameline V. Hemolacria: A pediatric clinical case report. J Fr Ophtalmol 2019;42:e15-7.
- Abel S. Haemorrhage from the lacrimal punctum. Br J Ophthalmol 1950;34:754-5.
- Drake AE, Packer CD. Epistaxis complicated by hemolacria: A case report. Clin Med Res 2020;18:99-101.
- Wiese MF. Bloody tears, and more! An unusual case of epistaxis. Br J Ophthalmol 2003;87:1051.
- AlGoraini Y, Şeyhibrahim A, Jawish M. A healthy young girl crying out blood: A case report. Heliyon 2021;7:e07143.
- Kosałka-Węgiel J, Wawrzycka-Adamczyk K, Matyja-Bednarczyk A, Siwiec A, Włudarczyk A, Bazan-Socha S. Hemolacria, epistaxis, bloody otorrhea, hemoptysis, and hematuria in an 18 -year old man. Pol Arch Intern Med 2021;735:735-6.
- 12. Humber CC, Lanigan DT, Hohn FI. Retrograde hemorrhage (hemolacria) from the lacrimal puncta after a Le Fort I osteotomy: A report of 2 cases and a review of the literature. J Oral Maxillofac Surg 2011;69:520-7.

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