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CASE REPORT

Treatment of an eyelid infantile hemangioma with potential ptosis development

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ABSTRACT

Background: Infantile hemangiomas are prevalent benign vascular tumors in children, frequently found on the head and neck, and pose a notable risk of visual impairment when situated on the eyelids. **Case presentation:** This report presents the case of a 9-year-old girl with a right upper eyelid hemangioma that has been present since the age of 3. Initially, she received intralesional triamcinolone acetonide injections, which resulted in limited improvement, prompting the decision for surgical excision. The procedure was conducted via an anterior approach, successfully excising the tumor. Following the surgery, the patient experienced no complications, and her condition significantly improved. This case underscores the critical need for prompt intervention in eyelid hemangiomas to avert visual complications. **Conclusion:** Management strategies for infantile periocular hemangiomas are influenced by factors such as the stage and location of the lesion, as well as the risk of functional impairment, with surgical excision proving to be an effective treatment option during the involution phase.

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Highlights

1. Treatment of infantile periocular hemangioma: emphasizing the efficacy of intralesional triamcinolone acetonide injections and surgical excision as treatment options for a hemangioma located in the right upper eyelid. It underscores the significance of prompt intervention to avert potential visual impairment.
2. Surgical approach in the involution phase: surgical excision of the hemangioma was conducted once the lesion entered the involution phase, illustrating that surgery can serve as a viable option when pharmacological treatments prove to be less effective during this stage.

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BACKGROUND

Hemangioma is a prevalent benign vascular tumor in children characterized by the proliferative changes of endothelial cells, with an incidence rate ranging from 5% to 10% (Selvi, 2019). Hemangiomas are most frequently located on the head and neck, accounting for 60% of cases, followed by the trunk at 25%, and the extremities at 15% (Baihaqi and Wibowo, 2024; Kunimoto et al., 2022). While infantile hemangiomas are generally not present at birth, they may develop alongside various preceding lesions and can lead to several complications, including disfigurement, scarring, vision loss, airway obstruction, congestive heart failure, and even death (Smith et al., 2016). Risk factors for infantile hemangioma include being female, having a preterm birth, being of low birth weight, experiencing anemia during pregnancy, having placenta previa, a history of threatened miscarriage, premature rupture of membranes (PROM), abnormal amniotic fluid volume, Caucasian ethnicity, progesterone therapy, and a family history of miscarriages (Ding et al., 2020; Tavakoli et al., 2017). In most instances, the diagnosis is primarily established through clinical evaluation; however, a skin biopsy may be conducted to aid in identifying the lesion (American Academy of Ophthalmology, 2016; Smith et al., 2016).

Infantile periocular hemangioma (IPH) may resolve spontaneously without treatment, except in certain cases where intervention is necessary, such as in the presence of visual axis obstruction, anisometropia, or strabismus (Tavakoli et al., 2017). The primary treatment for infantile periocular hemangioma (IPH) has traditionally been systemic corticosteroids; however, recent years have seen the introduction of beta-blockers. Nevertheless, because of the adverse effects associated with systemic corticosteroids, local corticosteroid injections are now preferred (Sethuraman et al., 2014). In instances where vital structures, such as the orbit or airway, are compromised, surgical removal of the infantile hemangioma is considered necessary to prevent additional dysfunction (Krowchuk et al., 2019).

CASE

A 9-year-old girl presented with a lesion on her right upper eyelid that had been present since she was 3 years old. Last year, she received intralesional injections of triamcinolone acetonide every two weeks. After five injections, the tumor became firmer, and surgery was scheduled for its removal; however, she did not attend and was lost to follow-up. Currently, she reports that the lump has not increased in size and denies any pain, itching, or burning sensation associated with it. Nonetheless, she experiences a sensation of fullness when opening her eye. The lump does not bleed easily, and no lesions were found on other parts of her body.

Figure 1. Hemangioma mass on the right upper eyelid

The patient's birth history indicates she was born at term with a birth weight of 2,700 grams, and the lump was not present at birth. There is no family history of hemangiomas or other tumors. Physical examination revealed a single, skin-colored tumor on the lateral aspect of the right upper eyelid. The lesion was soft, compressible, immobile, and exhibited no thrill, bruit, or pulsation (Fig. 1 A-B). The patient's visual acuity was 10/10 in both eyes, and no abnormal findings were noted during the slit-lamp examination. She received additional intralesional injections of triamcinolone acetonide every two weeks, and after a total of eight injections, she is scheduled for surgery.

Total excision of the hemangioma was performed using an anterior approach, 6 mm above the eyelid margin. Prior to excision, an intralesional injection of triamcinolone acetonide was administered to

facilitate lesion identification during the surgical procedure. Following the injection, a skin incision was made, revealing the hemangioma located in the subcutaneous layer, just superficial to the orbicularis oculi muscle. The hemangioma was composed predominantly of fibrofatty tissue with minimal vascularization. Surrounding tissues were carefully dissected until the mass was fully visible, after which total excision of the mass, along with the affected skin, was carried out. Post-surgery, no mass remained on the upper right eyelid.

The day following the surgery, the patient was evaluated, and no signs of bruising or significant edema were observed on her upper right eyelid (Fig. 2). She was prescribed a topical application of chloramphenicol-hydrocortisone eye ointment once daily, along with sodium hyaluronate-polyvinylpyrrolidone eye drops to be administered four times a day in her right eye.

Figure 2. Right upper eyelid condition on the day after surgery.

One week post-surgery, the patient returned for a follow-up examination. She reported no complaints. Upon examination of the right upper eyelid, there were no signs of infection or significant edema (Figure 3).

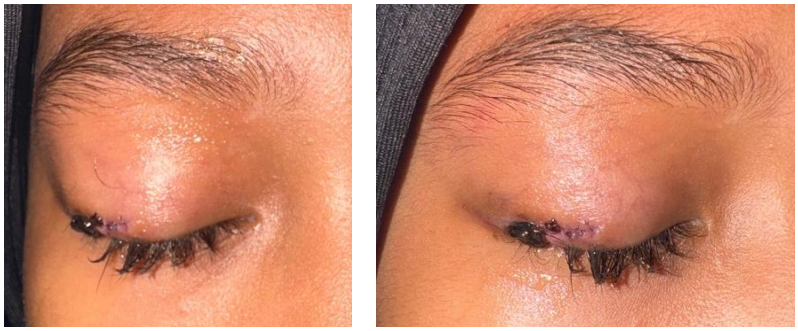

Figure 3. Right upper eyelid condition one week after surgery.

DISCUSSION

Based on The International Society for the Study of Vascular Anomalies (ISSVA), infantile hemangioma is categorized as vascular tumor, characterized as the proliferation of endothelial cells (Kunimoto et al., 2022). Infantile hemangioma are the most common vascular tumor marked by the presence of glucose transporter-1 protein (GLUT-1). This expression is a defining feature of infantile hemangioma (Daniel P Krowchuk et al., 2019). Several risk factors are associated with infantile hemangioma, including female sex, low birth weight, placental anomalies, and a family history of hemangiomas (Sethuraman et al., 2014). The head and neck region is the most frequent location for infantile hemangioma lesions, accounting for 60% of cases, followed by the trunk at 25% and the extremities at 15%. Based on the depth of the lesion, infantile hemangiomas are classified into three categories: superficial (located in the epidermis and dermis), deep (beneath the skin surface), and mixed .[2,8] In the deep type of infantile hemangioma, the lesion typically appears either bluish or skin-colored. This condition undergoes three distinct phases of growth: (1) the early proliferative phase, marked by rapid lesion growth; (2) the plateau phase, characterized by stability and quiescence; and (3) the involution phase, during which the lesion becomes softer and more compressible. Notably, the proliferative phase of deep infantile hemangiomas occurs later than that of superficial hemangiomas (Smith et al., 2016). In our case, the patient presented with the risk factor of being female. Given her age and the examination findings, which revealed a skin-colored lesion with a soft and compressible

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consistency, the hemangioma is determined to be in the involution phase. Additionally, Nguyen et al. reported a case of multifocal infantile hemangiomas, including one on the left upper eyelid, in a fraternal twin infant who also had the risk factors of being female and born preterm (Nguyen et al., 2023).

The treatment of infantile hemangioma typically depends on its stage, extent, and location. Lesions that threaten function—such as those located on the eye, nasal tip, ear, or lip—require intervention (Jung, 2021). Various treatment options are available for infantile hemangioma, including systemic beta-blockers, systemic corticosteroids, intralesional corticosteroid injections, topical beta-blockers, and surgical intervention (Krowchuk et al., 2019). Currently, oral propranolol is regarded as an alternative treatment for infantile hemangioma, particularly in instances that necessitate systemic therapy, such as during the proliferative stage. In cases involving vision-threatening, extensive, or deep orbital involvement, oral propranolol is indicated alongside oral corticosteroids. For lesions confined to the eyelid, options include intralesional corticosteroids or topical timolol gel. Topical timolol is typically used for the treatment of thin and/or superficial infantile hemangiomas (American Academy of Ophthalmology, 2016; Krowchuk et al., 2019).

Corticosteroids inhibit the production of vascular endothelial growth factor A (VEGF-A) in hemangioma-derived stem cells. VEGF-A levels are elevated during the proliferative phase compared to the involuting phase and serve as the primary regulator of angiogenesis and vasculogenesis. Additionally, corticosteroids modulate the activity of nuclear factor κ-light-chain enhancer of activated B cells (NF-κB) by suppressing its function. This suppression results in the downregulation of VEGF-A, leading to anti-endothelial effects and the apoptosis of endothelial cells. In contrast, beta blockers induce vasoconstriction and inhibit both angiogenesis and the production of nitric oxide (Greenberger and Bischoff, 2011). Systemic corticosteroids are linked to various adverse effects, such as growth retardation and immune suppression. In contrast, topical timolol is associated with minimal adverse effects; however, for patients who cannot tolerate beta-blockers, intralesional steroid injections may be considered as an alternative treatment to mitigate the side effects of systemic corticosteroids. Although topical timolol generally has few side effects, it can cause local irritation and bronchospasm in some individuals (American Academy of Ophthalmology, 2016).

In this case, the patient's hemangioma lesion was in the involution stage and located on the right upper eyelid, which posed a risk of impairing her vision due to the potential for mechanical ptosis. Therefore, the author opted for intervention. An intralesional injection of the steroid triamcinolone acetonide was selected because the lesion was confined to the upper eyelid, minimizing the risk of systemic adverse effects. Similarly, Kurachi et al. reported a case of infantile hemangioma on the upper eyelid of a low birth weight infant, in which oral propranolol was used to reduce the lesion during the proliferative phase (Greenberger and Bischoff, 2011). Another treatment for infantile hemangioma was reported by Ballesteros et al., who utilized a 0.1% timolol ophthalmic gel to address an eyelid hemangioma in a 2-month-old infant (Fernández-Ballesteros et al., 2012). The selection of these diverse treatments is influenced by the patient's stage, age, and the location of the lesion. Surgical excision of infantile hemangiomas is feasible for well-circumscribed lesions; however, meticulous hemostatic control is essential during the procedure. Typically, surgical intervention is avoided in infants due to the risk of excessive bleeding from the highly vascular tumor, unless the lesions cause ulceration, compress or deform critical structures (such as the airway or orbit), or affect cosmetically sensitive areas (American Academy of Ophthalmology, 2016). In our case, we conducted excisional surgery to remove the fibrofatty lesion, thereby alleviating the risk of mechanical ptosis and enhancing the visual function of the eye. Similarly, Kurachi et al. performed surgery on an infant due to the lesion's location on the upper eyelid, which posed a threat to her vision. The procedure was carried out while the lesion was in the involution stage (Kurachi et al., 2019).

Limitations

The case needs further long-term follow-up to assess tumor's growth in the future.

CONCLUSION

Infantile hemangioma of the eyelid is a benign vascular tumor that can obstruct the visual axis, lead to anisometropia or strabismus, and result in considerable disfigurement. Various treatments are available

for infantile hemangioma, with the choice of treatment determined by several factors, including the lesion's stage, location, and extent. Surgical intervention may be performed during the involution stage to excise the lesion, as pharmacological therapies often prove ineffective at this stage.

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Conflict of Interest

The authors declare that there are no conflicts of interest.

Ethical Consideration

The patient has provided consent for the publication of this case report, including all information presented herein.

Author Contribution

DNF: concept, literature search, manuscript preparation, manuscript editing. YR: concept and design, definition of intellectual content, literature search, manuscript editing, manuscript review.

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