

Parenteral corticosteroids followed by early surgical resection of large amblyogenic eyelid hemangiomas in infants

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Background: The purpose of this study was to evaluate the results and complications of early surgical resection of large amblyogenic subdermal eyelid hemangiomas in infants after prior short-term parenteral administration of corticosteroids.

Methods: Sixteen infants were given dexamethasone 2 mg/kg/day in two divided doses for three consecutive days prior to scheduled surgical excision of large eyelid hemangiomas. The lesions were accessed via an upper eyelid crease, subeyebrow incision, or a lower eyelid subciliary incision.

Results: In all cases, surgical excision of the entire lesion was possible with no significant intraoperative or postoperative complications. The levator muscle/aponeurosis complex was involved in 31.25% of cases and was managed by reinsertion or repositioning without resection. A satisfactory lid position and contour with immediate clearing of the visual axis was achieved in all but one case (93.8%).

Conclusion: Parenteral corticosteroids helped in reducing volume and blood flow from the hemangiomas, allowing for very early total excision of large subdermal infantile hemangiomas without significant intraoperative hemorrhage. This resulted in immediate elimination of any reason for occlusion amblyopia. Long-term follow-up of visual development in these patients would help to demonstrate the effectiveness of this strategy compared with more conservative measures.

Keywords: large eyelid hemangiomas, early surgical resection, parenteral corticosteroids

Introduction

Some authors have described large eyelid hemangiomas as “high-risk” or “clinically deceiving” lesions because, even after treatment with steroids, they display an “apparently open” visual axis.¹ Studies of the prevalence of amblyopia in such patients have reported rates of 50%–60%.² Present treatment strategies with either oral or local intralesional steroids, although very effective from the long-term cosmetic point of view, do not appear to achieve the desired goal as regards prevention of imminent amblyopia. Therefore, early primary surgical resection would provide an attractive alternative, but many surgeons are reluctant to perform the procedure for fear of postoperative scarring causing eyelid disfigurement or extensive bleeding potentially requiring blood transfusion.³ The aim of this work was to evaluate the results and complications of early surgical resection of large amblyogenic subdermal eyelid hemangiomas in infants after short-term parenteral administration of corticosteroids.

Materials and methods

In this prospective case series, 16 infants with large subdermal upper or lower lid hemangiomas, without orbital extension, were given parenteral dexamethasone

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2 mg/kg/day in two divided doses for three consecutive days prior to scheduled surgical excision of the tumor. The decision for early surgery was taken after parental counseling, explaining the benefits and risks and with the approval of the local research ethics committee. Steroids were given under the supervision of a pediatrician. A detailed history and physical examination were performed, including recording of body weight, height, skull circumference, and blood pressure, as well as hematologic investigations for baseline blood sugar, electrolytes, and abnormalities of coagulation. These measurements were repeated three months after steroid administration. Hemangiomas were included if the lesions were large enough to occupy more than half of the lid margin, extended beyond the eyelid region, and obstructed the visual axis. Preoperative computed tomography enabled specific localization, accurate measurement, and determination of the extent of the lesions. No Doppler studies were needed because all cases were restricted to the eyelids with no orbital involvement. Surgical removal was done by one of the authors (REE) and the lesions were biopsied. Any intraoperative or postoperative complication was recorded. Withdrawal of steroids was not necessary because the duration of therapy did not exceed three days, according to the protocol of withdrawal of steroid therapy.⁴ Treatment of amblyopia was initiated soon after surgery.

Results

The mean age of the infants was 3.93 ± 2.8 (1–11) months, and approximately two thirds were female ($n = 10$, 62.5%). Most of the hemangiomas ($n = 13$, 81.2%) affected the upper eyelid. All lesions caused obscuration of the visual axis with mechanical ptosis leading to corneal deformation. The average size of the hemangiomas was 3.22 ± 1.15 cm \times 2.63 ± 1.15 cm. Preoperative steroids caused a change in the texture and color as well as size of the lesions, and the overlying skin became wrinkled with ulceration and sloughing of some areas. The tumors were accessed via an upper eyelid crease incision (in 10 cases), a subeyebrow incision in the upper eyelid (in three cases), or a subciliary incision in the lower eyelid (in three cases). Dissection was mainly carried out in a blunt fashion and aided by careful cauterization. Complete resection was attempted, leaving only small residues of hemangioma around the lacrimal gland, lacrimal canaliculi, or near the roots of the eyelashes. The levator muscle and aponeurosis were identified, carefully separated and, if involved, reinsertion and/or repositioning of the residual muscle was done without further muscle resection. This was required in five cases (31.25%). Biopsy confirmed

infantile hemangioma in all cases. A satisfactory appearance and lid position with immediate clearing of the visual axis was achieved in all but one case which had severe postoperative ptosis. No intraoperative complications or uncontrollable intraoperative hemorrhage was encountered, and no donor blood transfusion was required. No postoperative hemorrhage occurred. Other than expected sloughing of the overlying skin due to necrosis of the hemangioma, no preoperative complications from steroids or withdrawal symptoms were noted, and there was no adrenal suppression or growth retardation. Three months after resection, mean corneal astigmatism was 0.87 ± 0.85 diopters. No recurrences have been noted during a follow-up period of 4.25 ± 1.77 (3–9) months.

Discussion

Hemangioma of the eyelid accounts for 1%–3% of all cases of hemangioma in newborns.⁵ It may be deep subdermal or superficial (known as a strawberry nevus) or sometimes a combination of both. Subdermal hemangiomas tend to undergo more rapid and more prolonged growth than superficial ones.⁶ Ultrasonographic examination shows that these lesions have a very high blood flow in feeding vessels, ie, about 2–3 orders of magnitude greater than in normal capillary beds.⁷ Large hemangiomas can result in amblyopia due to occlusion of the visual axis or astigmatism. Other authors have suggested that a size greater than 1 cm in largest diameter is an important predictor of amblyopia.⁸ Early surgical removal in infancy, if possible, would be beneficial, considering that even a 1–2-week period of visual deprivation in this critical period has been demonstrated to have the potential to cause permanent cortical visual deficits.¹

Surgical excision of eyelid hemangiomas is usually indicated for removal of small, involuted, well localized residual lesions (and likely to leave an esthetically more acceptable scar) and for lesions that continue to cause amblyopia despite treatment with oral or intralesional steroids, or both. However, there is limited experience in the literature of primary excisional surgery, especially in infants.^{3,5,9–11} In one report of six hemangiomas treated primarily with surgical resection,³ two cases required intraoperative donor blood transfusion to control severe intraoperative hemorrhage, and one case had postoperative wound transudation and wound dehiscence. The same patient later needed surgical scar revision. Other studies showing successful primary removal without complications either reported on a few moderately sized lesions,^{5,9} or removal done under ultrasonic guidance using a Dissectron.^{10,11} In this case series, preoperative parenteral corticosteroid injections, known to reduce volume and blood

flow in haemangiomas,¹² allowed for even large subdermal infantile hemangiomas to be completely and safely excised without postoperative fibrosis or disfigurement (Figure 1). The idea for this came from a previous report¹³ of using pulsed intravenous methylprednisolone followed by oral corticosteroids in children with periocular hemangioma in whom the visual prognosis was poor. That study showed that short-term parenteral corticosteroids alone achieved rapid shrinking of hemangiomas and complete resolution of their visual impact within two days, which would appear to be more rapid than with usual oral corticosteroid therapy.

Systemic side effects are reported in the literature to occur in 27%–44%^{14,15} of cases when corticosteroids are used for prolonged treatment of periorbital hemangiomas. These include behavioral changes, insomnia, cushingoid appearance, and hypertension, which were all avoided in our study. Despite the high parenteral steroid dose given, adrenal suppression and/or growth retardation were not noted during the short period of follow-up. However, the long-term repercussions, if any, of early parenteral steroid therapy on the natural growth and development of such very young infants remain to be assessed by longer follow-up.

Resection of the levator muscle/aponeurosis, despite involvement in almost one third of cases (31.25%), was not performed at this stage to avoid postoperative lid retraction. Postoperative ptosis was avoided in these cases by careful

identification and suturing of healthy tissue. Only one case of severe postoperative ptosis due to an unidentifiable levator muscle was encountered using this technique.

It has been demonstrated that the longer a complicated periocular hemangioma is observed, the greater the astigmatism and the less amenable it will be to correction following tumor removal.^{16,17} In one study,¹⁷ the degree of astigmatism improved from 2.75 to 0.80 diopters in infants younger than three months undergoing surgical resection and from 3 to 1.1 diopters in older children. In our series, astigmatism was only measured postoperatively because the large size of the lesion did not permit accurate preoperative measurements, and was found to be insignificant (0.87 ± 0.85) in accordance with the figures previously mentioned.

Conclusion

Early surgical resection of large eyelid hemangiomas following short, high-dose, parenteral corticosteroid administration is safe and results in immediate elimination of amblyogenic factors. Surgical removal from the very thin and underdeveloped infant eyelid requires an excellent knowledge of eyelid anatomy to avoid massive intraoperative bleeding and postoperative ptosis. Long-term follow-up of the visual development of these patients would help to confirm the advantages of this strategy compared with more conservative measures.

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Disclosure

This paper was presented in part as a poster at the European Society of Ophthalmic Plastic and Reconstructive Surgery annual meeting held on September 9–11, 2010, in Munich, Germany. Otherwise, the authors report no conflicts of interest in this work.

References

1. Frank RC, Cowan BJ, Harrop AR, Astle WF, McPhalen DF. Visual development in infants: visual complications of periocular haemangiomas. *J Plast Reconstr Aesthet Surg*. 2010;63(1):1–8.
2. Haik BG, Jakobiec FA, Ellsworth RM, Jones IS. Capillary hemangioma of the lids and orbit: an analysis of the clinical features and therapeutic results in 101 cases. *Ophthalmology*. 1979;86(5):760–792.
3. Walker RS, Custer PL, Nerad JA. Surgical excision of periorbital capillary haemangiomas. *Ophthalmology*. 1994;101(8):1333–1340.
4. Jacobs TP, Whitlock RT, Edsall J, Holub DA. Addisonian crisis while taking high dose glucocorticoids. An unusual presentation of primary adrenal failure in two patients with underlying inflammatory diseases. *J Am Med Assoc*. 1988;260(14):2082–2084.

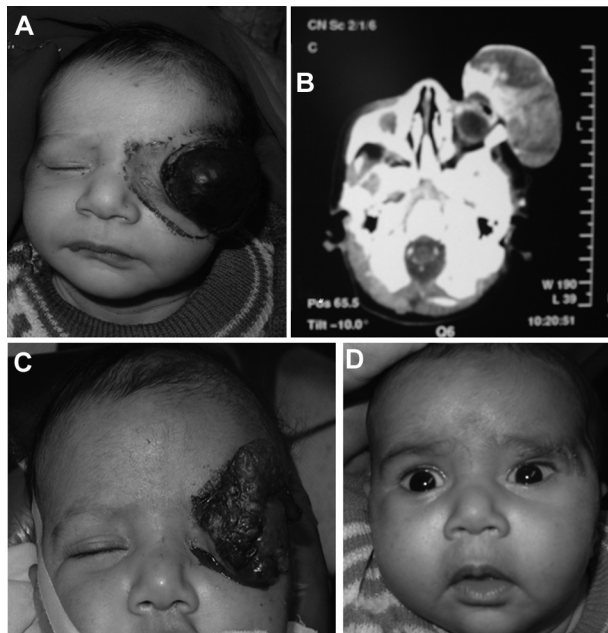


Figure 1 (A) Two-month-old boy presenting with a huge “tennis ball” hemangioma with (B) computed tomography scan showing no orbital extension. (C) Three days after parenteral steroids showing a marked decrease in size with sloughing of the overlying skin. (D) Two weeks after surgical removal.

5. Slaughter K, Sullivan T, Boulton J, O'Reagan P, Gole G. Early surgical intervention as definitive treatment for ocular adnexal capillary haemangioma. *Clin Experiment Ophthalmol*. 2003;31(5):418–423.
6. Tambe K, Munshi V, Dewsbury C, Ainsworth JR, Willshaw H, Parulekar MV. Relationship of infantile periocular hemangioma depth to growth and regression pattern. *JAAPOS*. 2009;13(6):567–570.
7. Verity DH, Restori M, Rose GE. Natural history of periocular capillary haemangiomas: changes in internal blood velocity and lesion volume. *Eye (Lond)*. 2006;20(10):1228–1237.
8. Schwartz SR, Blei F, Ceisler E, Steele M, Furlan L, Kodsi S. Risk factors for amblyopia in children with capillary hemangiomas of the eyelids and orbit. *JAAPOS*. 2006;10(3):262–268.
9. Aldave AJ, Shields CL, Shields JR. Surgical excision of selected amblyogenic periocular capillary hemangiomas. *Ophthalmic Surg Lasers*. 1999;30(9):754–757.
10. Momtchilova M, Pelosse B, Diner PA, Vazquez MP, Laroche L. Amblyopia and peri-ocular capillary hemangioma of infancy: screening and clinical course before and after surgery. *J Fr Ophthalmol*. 2004;27(10):1135–1140. French.
11. Claude O, Picard A, O'Sullivan N, et al. Use of ultrasonic dissection in the early surgical management of periorbital haemangiomas. *J Plast Reconstr Aesthet Surg*. 2008;61(12):1479–1485.
12. Verity DH, Rose GE, Restori M. The effect of intralesional steroid injections on the volume and blood flow in periocular capillary haemangiomas. *Orbit*. 2008;27(1):41–47.
13. Delesalle F, Staumont D, Houmany MA, Breviere GM, Piette F. Pulse methylprednisolone therapy for threatening periocular haemangiomas of infancy. *Acta Derm Venereol*. 2006;86(5):429–432.
14. Ranchod TM, Frieden IJ, Fredrick DR. Corticosteroid treatment of periorbital haemangioma of infancy: a review of the evidence. *Br J Ophthalmol*. 2005;89(9):1134–1138.
15. Pope E, Krafchik BR, Macarthur C, et al. Oral versus high-dose pulse corticosteroids for problematic infantile hemangiomas: a randomized, controlled trial. *Pediatrics*. 2007;119(6):e1239–e1247.
16. Levi M, Schwartz S, Blei F, et al. Surgical treatment of capillary hemangioma causing amblyopia. *JAAPOS*. 2007;11(3):230–234.
17. Arneja JS, Mulliken JB. Resection of amblyogenic periocular hemangiomas: indications and outcomes. *Plast Reconstr Surg*. 2010;125(1):274–281.

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