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Intramuscular hemangioma of the pretarsal orbicularis oculi muscle in an adult treated with intralesional bleomycin injections

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ABSTRACT

Purpose: Intramuscular hemangiomas of the head and neck are rare and account for fewer than 1% of total cases. Reports of intramuscular hemangiomas in adults' eyelids, orbital and periorbital regions are especially scarce, conceivably because occurrence in the masseter muscle is more common. Herein we report a highly unusual case of hemangioma located in the pretarsal orbicularis oculi muscle of an adult patient. This report describes the clinical and pathological assessment of the patient, subsequent diagnosis of periorbital intramuscular hemangioma, and conception and implementation of a treatment approach using intralesional bleomycin injections.

Observations: A 29-year-old female patient without other clinical complaints presented with an enlarged, painless mass in the upper left eyelid. Physical examination of the mass suggested a vascular origin, and hemangioma diagnosis was confirmed by computed tomography and incisional biopsy. The anatomical location was determined as the pretarsal orbicularis oculi muscle involving mixed capillary-sized and cavernous-sized vessels. The treatment strategy involved monthly intralesional bleomycin injections (1 mL volume; 3 IU/mL) for 4 consecutive months resulting in notable size reversion. The patients experienced no associated complications, and the size remained stable over the 2-year follow-up period.

Conclusions: Intralesional bleomycin injections may offer an effective and safe treatment option for intramuscular hemangioma in the periorbital region. However, larger studies are needed to substantiate these findings further.

1. Introduction

Hemangiomas are benign vascular tumors found in skin, subcutaneous tissue or in the tissue of deep organs (e.g., the liver). Intramuscular hemangiomas (IMH) frequently occur in the trunk and extremities but are rare in the head and neck region, mostly in the masseter muscle or the trapezius.[1,2] Congenital capillary hemangiomas of the eyelids are more common than the acquired type suffered by adults, but both are scarcely reported. Existing reports include IMH in the sternocleidomastoid, temporalis, and extraocular muscles.[1,3] However, to the best of our knowledge, only a single case of IMH in the periorbital portion of the orbicularis oculi muscle has been described previously,[4] and the current case is the first to report an IMH specifically located in the pretarsal region.

Preoperatively, soft tissue masses are rarely diagnosed as IMHs, and magnetic resonance imaging (MRI) has been used to determine the nature of the lesion without surgery. Hemangiomas are vascular in appearance and embedded in fibrofatty tissue. By MRI, IMHs often appear as bright and sharply demarcated intramuscular lesions in T2 weighted images because of blood stagnation in surrounding cavernous vessels.[5,6] The septa of the fibrofatty tissue appear as repeating lines. In this report, the diagnostic imaging modality applied was computed tomography (CT) because of its availability in our institute. However, IMHs are poorly defined by CT,[7] and an incisional biopsy was used to support a conclusive diagnosis.

To date, the conventional treatment is surgical excision; however, the risk of recurrence is substantial (50%).[1,8] Here, we present a case of IMH in the periorbital region successfully treated with intralesional bleomycin injections (IBI). This report aims to describe the pathophysiology, clinical implications, and treatment response using our findings.

2. Case report

This study was approved by the institutional review board in

Abbreviations: IMH, Intramuscular hemangiomas; MRI, magnetic resonance imaging; CT, computed tomography; IBI, intralesional bleomycin injections; FDA, Food and Drug Administration.

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Dhahran Eye Specialist Hospital, and signed, written consent from the patient was obtained for publication of the case and use of the images. The study was conducted in accordance with the 1964 declaration of Helsinki.

A 29-year-old female patient presented at our facility with the chief complaint of a painless mass in the left upper eyelid that had become noticeable 7 years back and had been progressively enlarging since then. The patient has no medical history of similar childhood lesions and reported no trauma. However, the patient did indicate a previous surgical excision of a persistent chalazion in the same region.

Upon examination, she had full vision in both eyes and no abnormalities in the right eye or surrounding structures. Inspection of the left eyelid revealed a reddish and ill-defined mass measuring 3 × 1.5 cm and associated mechanical ptosis with tortuous blood vessels in the overlying skin. The mass was non-tender, non-pulsatile, fixed to the skin, and cool to the touch. No thrill was detected, and the mass was negative to retropulsion. The patient was then lost to follow-up for 14 months and returned pregnant; further enlargement of the mass was noted. The CT was only performed after the birth and revealed a highly enhanced soft tissue mass in the left upper eyelid with a vascular pattern suggestive of hemangioma. An incisional biopsy was subsequently performed, revealing a mixture of small capillary- and large cavernous-sized vessels, skeletal muscle fibers, and fibro-adipose tissue. The diagnosis of IMH was thus confirmed.

Conventional excision surgery was precluded by the location of the hemangioma and the risk of recurrence. The patient received a single intralesional dexamethasone injection (1 mL; 4 mg/mL) as treatment. One month later, a 2 mL mixture of dexamethasone (1 mL; 4 mg/mL) and triamcinolone (1 mL; 40 mg/mL) was injected. These treatments produced a 20% size reduction. The patient was again lost to follow-up for 5 months before presenting with ulceration and bleeding in the region of the mass managed with topical antibiotics after the wound was sutured. There was a need for an alternative treatment approach that was effective, safe, and long-lasting.

Before IBI, a negative history of respiratory disease was confirmed, and informed written consent was obtained. Baseline laboratory tests, chest X-rays, and photos of the eyelids were taken (Fig. 1A). Monthly intralesional injections of IBI (1 mL; 3 IU/mL) were administered under topical anesthesia using a 27-gauge needle with a multi-puncture technique. A reduction in venous tortuosity was noted after the first injection. A continuous size reduction was observed after the second injection (Fig. 1B), and the venous tortuosity disappeared after the third injection (Fig. 1C). A total of 4 injections were given. The hemangioma size reduction was maintained at the 2-year follow-up with no associated bleomycin-induced pneumonitis or pulmonary fibrosis symptoms (Fig. 1D). The follow-up chest X-ray remained clear.

3. Discussion

The origin of IMHs remains controversial. Some believe they are congenital and become slowly enlarged until their size makes them symptomatic[2]; others favor a traumatic or hormonal etiology.[9,10] In this

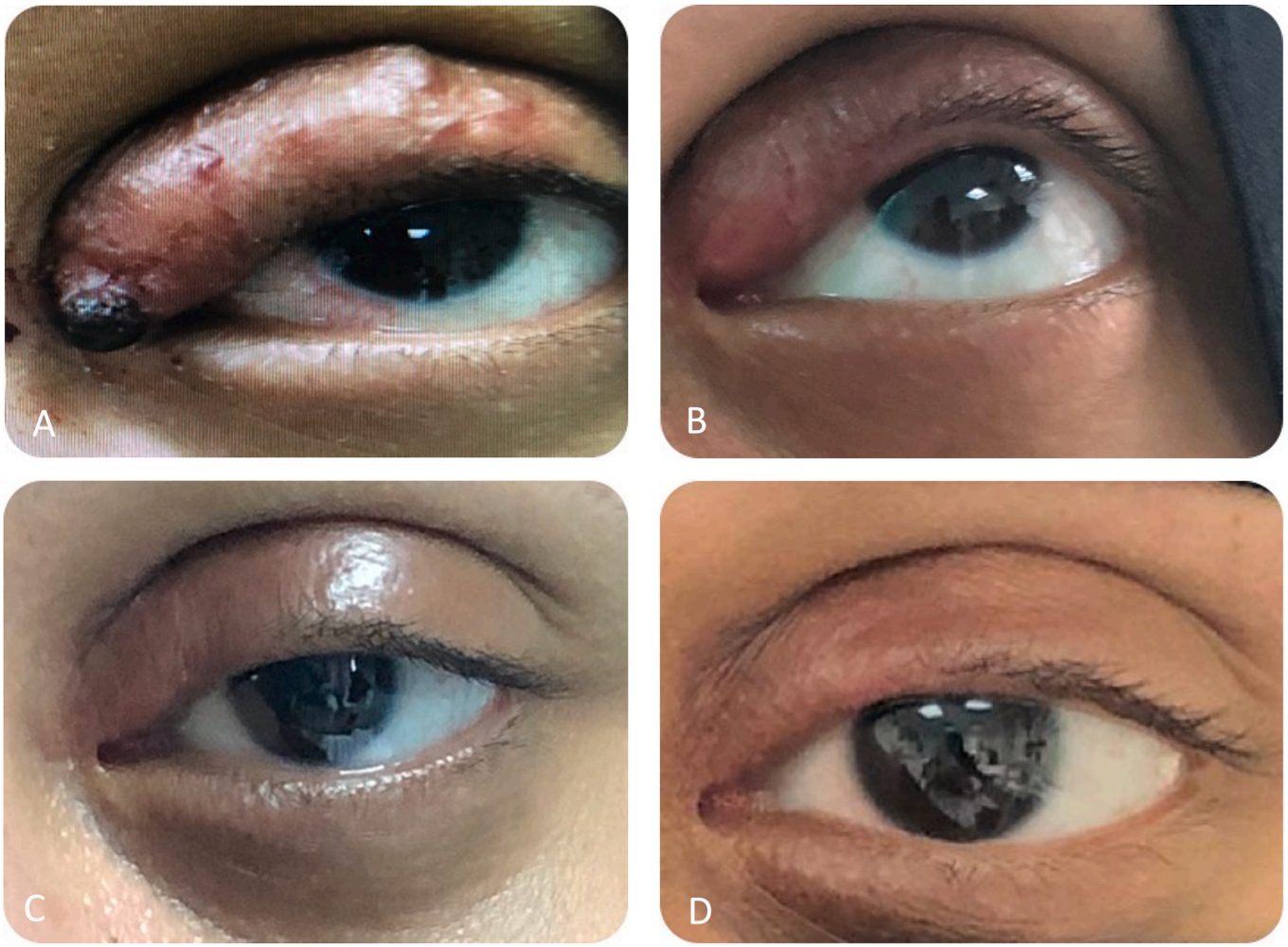

Fig. 1. Clinical photos of the intramuscular hemangioma in the left upper eyelid (A) prior to initiation of the intralesional bleomycin injections (IBIs), (B) after the second IBI, (C) after the third IBI, and (D) one year after the final IBI.

case, it is possible that the previous surgical removal of a chalazion from the patient's left upper eyelid may have contributed to IMH development. Furthermore, enlargement of the IMH was exacerbated during her pregnancy, indicating the potential involvement of hormonal changes. The pathophysiology of IMH development in our case appears supportive of a traumatic or hormonal origin.

Allen & Enzinger studied 89 patients with diverse IMH-types and histologically classified the masses into small-, large- and mixed-types based on the predominant size of associated vessels.[11] Importantly, IMH-type cannot be distinguished by radiology alone.[6,7] Histological assessment of incisional biopsy material from our patient confirmed a mixed-type IMH.

The management plan for IMH is usually patient-dependent and considers several factors, including tumor location, tumor depth, and functional and aesthetic consequences for the affected area. Different treatment modalities are described in the literature. Some examples include steroid therapy, embolization, cryotherapy, and blood-vessel ligation.[12] However, surgical excision is the standard approach despite the risk of recurrence (9–50%), with recurrence being more likely with incomplete excision.[11,13] For mixed-type IMH, the post-surgery recurrence rate was previously recorded as 28%, with a median of 2 years before surgery was repeated.[11] Surgery is a temporarily effective treatment option.

The previously reported orbital IMH of the orbicularis oculi muscle was identified as large cavernous-type and treated by surgical excision[4]; no recurrence was reported 2-years post-surgery. Our patient presented with a mixed-type IMH in a location considered high-risk for surgical excision. There was potential for serious complications like retro-orbital hemorrhage and extensive bleeding, and poor aesthetic outcomes. Hence, the less invasive approach of intralesional bleomycin injections was adopted with success. The patient sustained a recurrence-free condition over the 2-year follow-up period.

Systemic bleomycin administration has been approved by the Food and Drug Administration (FDA) for certain malignancies (e.g., squamous cell carcinoma and lymphoma). The use of IBI for vascular tumors is not yet FDA-approved but is used off-label for lymphangioma and infantile hemangioma refractory to other treatment modalities.[14–16] The dosage of bleomycin is variable in the literature. In one report, a refractory infantile hemangioma was treated with three injections at a dose of 15 mg/month. In another study, bleomycin dosage was calculated by weight (0.5 mg/kg) and administered in 5–9 injections, and the stability of lesion size was noted 12–16 months after the final injection. However, the natural regression of infantile hemangioma could not be ruled out as contributing to the recorded stabilizing effect. No side effects related to bleomycin injection were reported in either report.[14,15] We chose to use IBI for our patients to produce safe and satisfactory size reduction rather than complete removal. To the best of our knowledge, this is the first reported case of IMH treated with IBI.

4. Conclusion

This report describes a rare case of IMH in the pretarsal orbicularis oculi muscle of an adult patient. Standard surgical excision of the IMH posed a significant risk for recurrence and serious complications. As an alternative, the patient received IBIs resulting in the long-term stability of the reduced lesion size. Although no side effects were reported in our case, studies with larger sample sizes are needed to determine the efficacy and safety profile in a more diverse participant population.

Patient consent

Written consent from the patient was obtained to use her information and pictures in this report.

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Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Intellectual property

We confirm that we have given due consideration to the protection of intellectual property associated with this work and that there are no impediments to publication, including the timing of publication, with respect to intellectual property. In so doing we confirm that we have followed the regulations of our institutions concerning intellectual property.

Research ethics

We further confirm that any aspect of the work covered in this manuscript that has involved human patients has been conducted with the ethical approval of all relevant bodies and that such approvals are acknowledged within the manuscript.

IRB approval was obtained (required for studies and series of 3 or more cases).

Authorship

All listed authors meet the ICMJE criteria.

We attest that all authors contributed significantly to the creation of this manuscript, each having fulfilled criteria as established by the ICMJE.

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Declaration of competing interest

The authors declare no conflict of interest related to the study. The following authors have no financial disclosures: AlAbdulhadi H.A., AlBadri K.S., AlFayyadh M.A.

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