RESEARCH LETTER

Orbital Intra-osseous Infantile Haemangioma in an 8-month-old Infant: A Case Report

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To the Editor,

Infantile haemangiomas (IH) are common benign vascular tumours of early childhood, characterized by a stereotypical course of progression and regression. However, rare forms of IH do exist, for which diagnostic approaches and management strategies are less well established and standardized.

Here, we report the case of an 8-month-old female who was referred to our multidisciplinary consultation for vascular anomalies because of erythema of the left upper eyelid with slight swelling (**Fig. 1**A) that had appeared at 15 days of age and had gradually increased. She was born prematurely (34 weeks of gestation by Caesarean section because of heart rhythm abnormalities). She had 1 brother who died of Brugada syndrome at age 8, and her father died of Brugada syndrome during the girl's gestation. The mother was visually impaired.

Physical examination of the infant, including ophthalmological examination, was normal except for the erythema of the left upper eyelid. Palpation revealed no subcutaneous mass but an irregular aspect of the bony contour of the eye, which was not painful. MRI revealed intraosseous tissular lesion of the roof and lateral wall of the orbit, with intense and homogeneous enhancement (**Fig. 2**A), and CT scan revealed bone lysis of the superior and lateral wall of the orbit.

We obtained intra-osseous samples under general anaesthesia; pathological examination showed hyperplastic endothelial cells within the bone, with the cells expressing glucose transporter 1 (GLUT-1; Fig. 1B). We diagnosed infantile intraosseous haemangioma.

After multidisciplinary consultation, we decided on a "wait-and-see" attitude and planned to introduce propranolol in case of worsening of the condition because of the family history of Brugada syndrome and the mother's reluctance to initiate treatment. After 1 year of monitoring, the IH had decreased in size by half on MRI, and the ophthalmological examination remained normal. The erythema had subsided. Two years later, the IH had completely regressed clinically and on MRI, with normalization of the bone (Fig. 2B).

IH is the most common paediatric vascular tumour, occurring in 5% to 10% of infants, with a higher frequency in premature newborns (1). It usually appears on skin in the first days of life, then follows a proliferative phase until approximately 6 months; there is then an involution phase that leads to partial or complete regression. IH can present in various forms: superficial haemangiomas, which appear bright red on the skin surface; deep (or "subcutaneous") haemangiomas, located beneath the skin and appearing blue or skin-coloured; and mixed haemangiomas, which exhibit both superficial and deep components (1). The treatment of complicated cutaneous IH relies on beta-blockers, with propranolol (2 to 3 mg/kg/day) being the first-line option, provided there are no contraindications to its use (2–4). Moreover, IH can oc-

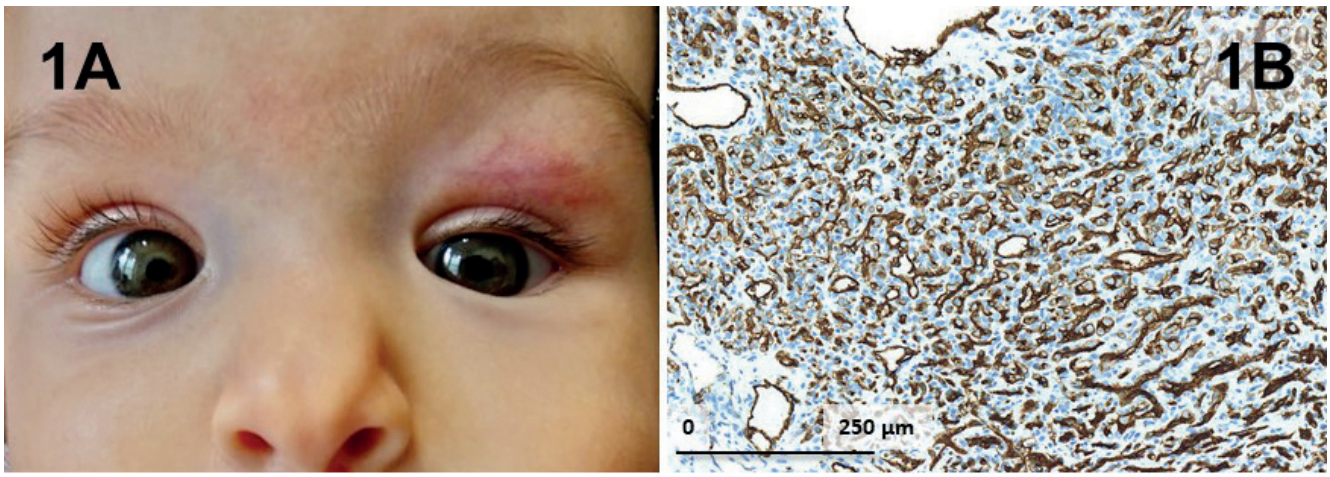

Fig. 1. (A) Clinical aspect at diagnosis: erythema of the left upper eyelid with slight swelling. (B) Histological section showing clear diffuse GLUT-1 labelling of endothelial cells.

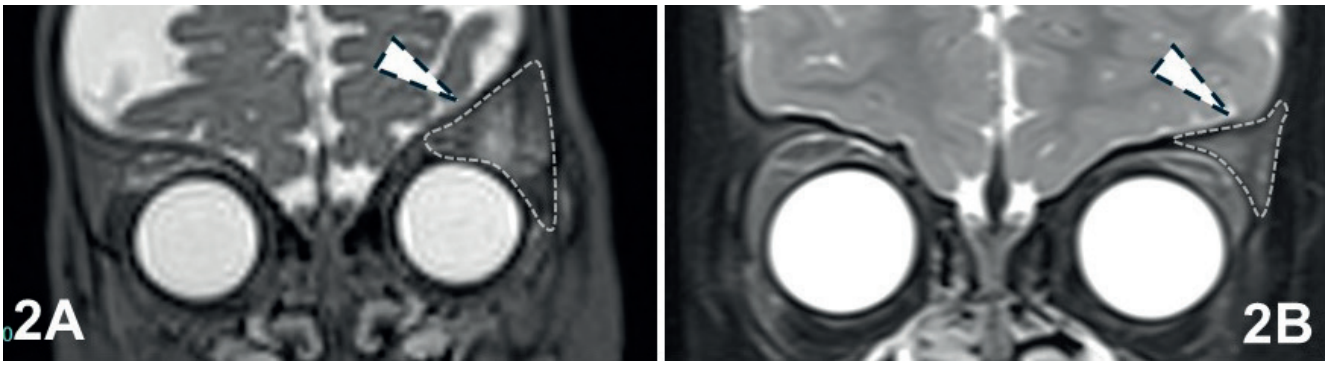

Fig. 2. (A) MRI section at 8 months of age showing intraosseous tissular lesion with intense and homogeneous enhancement. (B) MRI section 30 months of age showing complete regression of infantile haemangioma.

cur in locations other than the skin, such as the mucous membranes or the liver (5), with other sites being even rarer. In these very rare locations, data on evolution and on the efficacy of propranolol are lacking.

Intraosseous haemangiomas are benign vascular tumours that can be categorized into intraosseous haemangiomas and infantile intraosseous haemangiomas. The former is a rare and poorly known condition that occurs mainly in young adults and does not share the same clinical and pathological features as infantile intraosseous haemangiomas, notably lacking GLUT-1 expression (6, 7). Regarding infantile intraosseous haemangioma, only 1 case has previously been published to our knowledge. That case occurred in a maxillary location associated with bone lysis in a 1-month-old male (8). It was treated with oral corticosteroids (2 mg/kg/day) for 2 months, which was gradually decreased, and no propranolol. In this case, the lesion completely regressed after 4 months of treatment. This case, like ours, was localized in the bones of the head but in a different anatomical location. Also, both cases presented with bone lysis, which is often concerning as it suggests an aggressive lesion, requiring a biopsy for diagnosis. However, complete bone recovery was observed within the first 2 years of life in both cases.

In conclusion, we report a rare case of infantile intraosseous intra-orbital haemangioma with bone lysis in a premature infant, which regressed spontaneously and entirely within 2 years, including the restoration of normal bone structure. It is important to recognize this very rare location and clinical presentation to ensure accurate diagnosis and personalized management.

The authors have no conflict of interest to declare.

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