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Brief Clinical Studies

Title

Ulcerated infantile hemangioma of the hard palate: diagnostic treatment with oral propranolol

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Abstract

Infantile hemangiomas arising in the palate are rare. The authors describe a case of ulcerated infantile hemangioma of the hard palate with feeding difficulty. To our knowledge, this is the first reported case of immunohistochemically diagnosed palatal infantile hemangioma successfully treated using oral propranolol.

Keywords

Glucose Transporter Type 1; Hemangioma; Immunohistochemistry; Palate; Propranolol; Soft Tissue Neoplasms; Ulcer

Introduction

Infantile hemangiomas (IHs) are the most common soft-tissue tumors of infancy and are characterized by a proliferating phase in early infancy followed by a prolonged involuting phase.¹ Although approximately 60%–80% of IHs occur in the head and neck region^{2,3} and mucous membranes are involved in 10%,¹ IHs of the palate have rarely been reported.

Patient

The patient was delivered with a birth weight of 2.5 kg at 37 weeks of gestation following an uneventful pregnancy and delivery. She was noted to have an opalescent lesion on the palate with feeding difficulty and was referred at age 9 weeks to a local city hospital. A swollen lesion with small ulceration was observed on the left side of the hard palate (Fig. 1a), which showed high signal intensity on T2-weighted magnetic resonance imaging (Fig. 1b).

Following enlargement of the ulceration because of poor body weight gain owing to her feeding difficulty, a feeding tube was placed and she was referred to us at age 12 weeks.

On examination, the left side of the hard palate was ulcerated, with the crater measuring approximately 1 cm in diameter, surrounded by reddish mucosa with slight swelling (Fig. 2a). There were no cutaneous lesions suggestive of IHs on the entire body. Her body weight at the time was 4.6 kg. We suspected the possibility of ulcerated IH but were unable to reach a definitive diagnosis.

We opted to institute diagnostic treatment, so she was hospitalized at age 13 weeks for treatment with oral propranolol. Administration was started at a dose of 1 mg/kg/day twice daily and was increased by 1 mg/kg/day over the next 2 days to a maximum dose of 3 mg/kg/day with cardiovascular assessment. The ulceration healed after discharge following 2 weeks of propranolol treatment and she became able to suckle and nurse properly.

Incisional biopsy under general anesthesia (Fig. 2b) showed immunopositivity for glucose transporter protein isoform 1 (GLUT1) (Fig. 3) at age 6 months. There has been no recurrence of ulceration in the 4 months of follow-up with propranolol maintained at 3 mg/kg/day as of this writing.

Discussion

The prevalence of IHs in term neonates is around 4.5%, with a female and white predominance. Placental anomalies, prematurity, and low birth weight are also vital risk factors.⁴

Ulceration is the most prevalent complication in patients with IHs, at approximately 16%.⁵ Infants with ulcerated IHs are often irritable, feed poorly, and are unable to sleep.⁶ Ulcerated IHs are commonly seen in the head and neck region, with a preponderance on or adjacent to the lip, in the postauricular sulcus, or in the neck folds. Thus, maceration and frictional stress may be contributory factors to ulceration.⁷

All ulcerated and obstructive IHs require immediate treatment.⁸ Due to its impressive efficacy and safety profile, propranolol has become considered first-line therapy for complicated IHs.⁸ The response rate of oral propranolol at a dose of 2–3 mg/kg/day for a mean of 6 months is 96%–98%, and complete or near-complete regression occurs in 60% of cases.^{9–11} Thus, 6 months is considered the standard duration of propranolol treatment,¹⁰ although the relapse rate is around 17%–20% after 6 months of treatment.^{12,13}

IHs in the head and neck region involve the mucous membranes, such as the lips, buccal mucosa, palate, nasal mucosa, conjunctiva, tongue, and floor of mouth,¹ as well as the airway, including the subglottis and trachea.¹⁴ Despite several reports on "hemangioma of the palate" in the literature our review identified no cases of immunohistochemical diagnosis of IHs arising in the palate.

In our case, ulceration occurred in the palate without obvious cause at age 9 weeks. Because the clinical appearance was not characteristic of IHs, a diagnosis of exclusion was made based on the response to treatment with propranolol. The diagnosis was confirmed at age 6 months by positive endothelial staining for the immunohistochemical marker GLUT1, which is highly specific and diagnostic for IHs during all phases.¹⁵ Based on this confirmed histological diagnosis of IH, we plan to continue propranolol treatment beyond the patient's first year of life.

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Figures

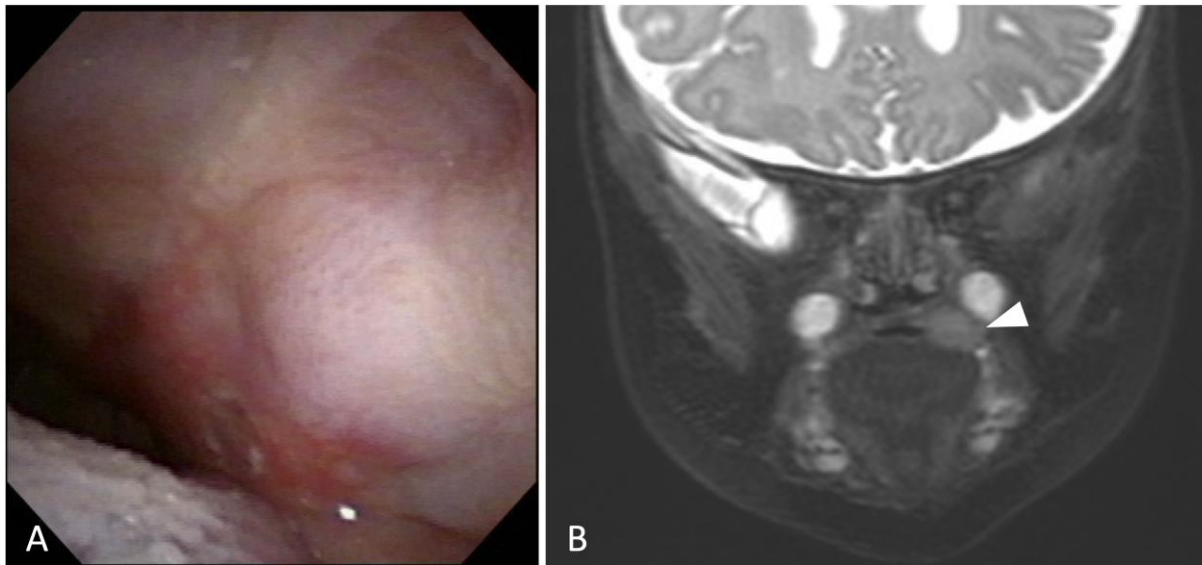


Figure 1. (A) A swollen lesion with small ulceration observed on the left side of the hard palate. (B) Relatively high signal intensity on T2-weighted magnetic resonance imaging observed on the left side of the hard palate (arrowhead).

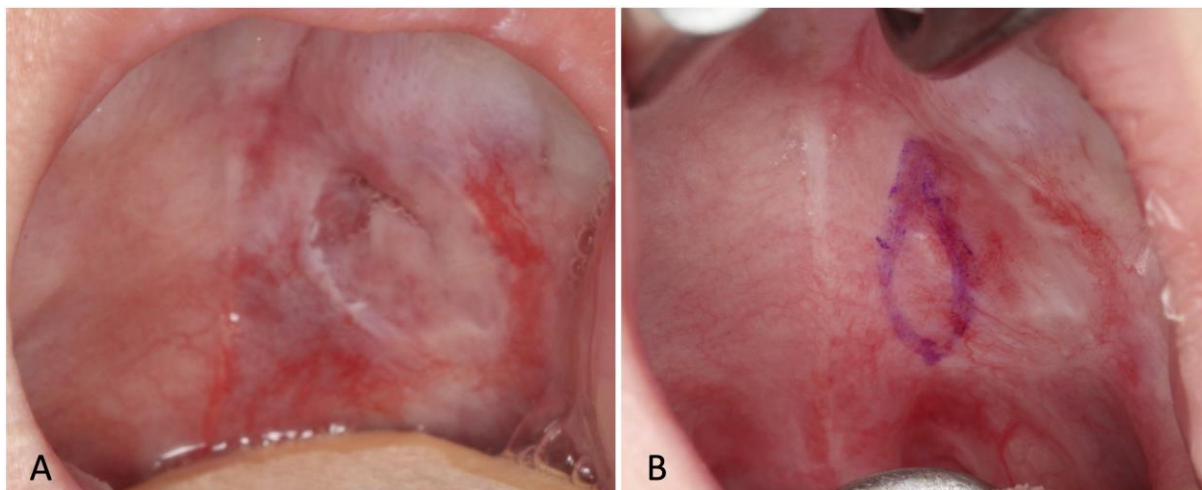


Figure 2. (A) Left side of the hard palate showing ulceration measuring approximately 1 cm in diameter, surrounded by reddish mucosa with slight swelling. (B) Intraoral view at the time of incisional biopsy under general anesthesia at age 6 months. The marked lesion was incised for pathological examination.

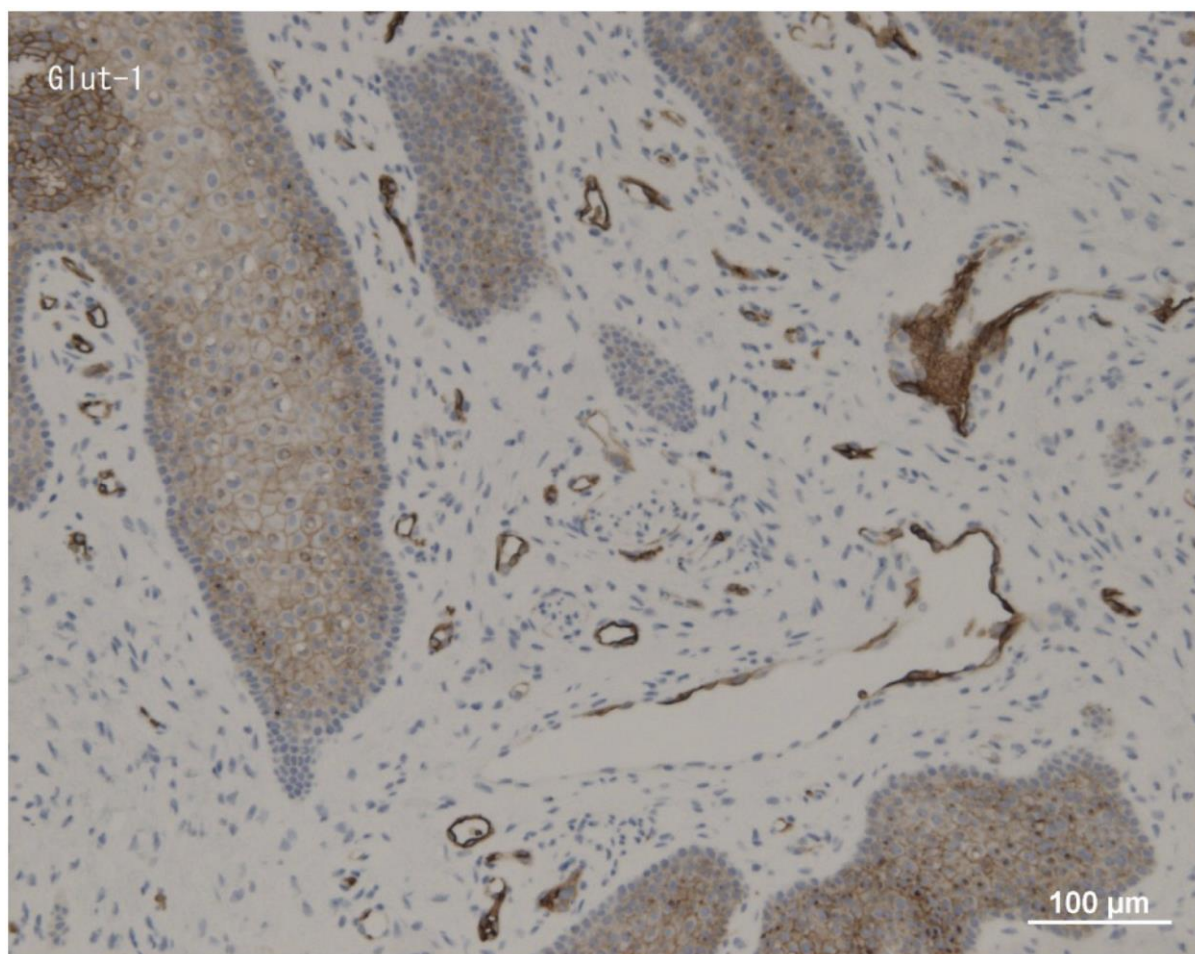


Figure 3. Endothelium in the biopsy specimen of the scar in the hard palate is positive for glucose transporter protein isoform 1 on immunohistochemical staining, confirming the diagnosis of infantile hemangioma.