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Case Report

Dual-threat: Extensive airway haemangioma in a context of focal subcutaneous haemangioma

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Abstract

Introduction: Infantile haemangiomas are the most common vascular tumours in children, with higher prevalence in females and low birth weight infants. Beard distribution haemangiomas, involving the lower face, chin, preauricular areas, and anterior neck, are often associated with airway involvement in the supraglottic or subglottic regions. Even isolated head and neck haemangiomas may carry a risk of airway complications. Diagnosis relies on clinical evaluation, with bronchoscopy reserved for complex cases.

Aim: To emphasise early recognition and treatment of airway involvement in focal head and neck haemangiomas.

Case study: A 4-month-old former 27-week preterm infant with a birth weight of 1.07 kg required oxygen support since birth. He was referred for upper airway assessment due to stridor during crying. Examination revealed a stridor with subcostal and suprasternal recession and a 1.5 × 1.0 cm submental haemangioma. Direct laryngoscopy and bronchoscopy under anaesthesia identified an extensive airway haemangioma extending from the nasal cavity to the bronchi. The infant was treated with oral propranolol. At 11-month follow-up, the airway haemangioma had resolved, and the submental haemangioma had significantly reduced in size.

Results and discussion: Recognizing the association between head and neck haemangiomas and airway involvement is essential for prompt and effective management.

Conclusions: Head and neck haemangiomas, even if they appear isolated, may still pose a risk of airway involvement. Early recognition and a multidisciplinary approach are essential for managing these complex cases. Propranolol therapy plays a critical role in the treatment of these lesions, but close monitoring and ongoing follow-up are necessary to ensure successful outcomes and prevent airway complications.

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1. INTRODUCTION

Infantile hemangiomas (IH) are the most common vascular tumors of infants, particularly affecting females and low birth weight infants. The unique behaviour of haemangiomas, with their rapid proliferation phase over the first 9–12 months followed by spontaneous involution, often allows for conservative management unless complications arise, such as airway obstruction, ulceration, bleeding, visual impairment, disfigurement, or psychosocial impact. The association of hemangiomas in the "beard distribution" (lower face, chin, preauricular areas, and anterior neck) with airway involvement is crucial to recognize, especially as 65% of these cases can lead to airway hemangiomas, typically located in the supraglottic or subglottic regions.[1] Early recognition of this connection is crucial, as airway involvement can lead to life-threatening complications.

2. AIM

To emphasise early recognition and treatment of airway involvement in focal head and neck haemangiomas.

3. CASE STUDY

A 4-month-old boy, born prematurely at 27 weeks with a birth weight of 1.07 kg, was delivered via emergency caesarean section due to placental abruption in the mother. At birth, the infant had a low Apgar score and required immediate intubation. Although he was extubated on day 7, he was reintubated on day 18 due to respiratory distress. During his 4-month hospital stay, he experienced respiratory distress syndrome, recurrent pneumonia, and feeding difficulties. Suspected of having a strangulated hernia, he was transferred to our hospital for further management by the paediatric surgical team. He was referred to the Otorhinolaryngology (ORL) team for airway assessment because of stridor after successful extubation.

On physical examination, the infant presented with soft biphasic stridor accompanied by subcostal and suprasternal retractions, necessitating nasal prong oxygen support. A subcutaneous haemangioma, which developed postnatally at around 1 month of age and measured 1.5 × 1.0 cm, was noted in the submental region. (Figure 1a–1c). He underwent examination under anaesthesia, during which direct laryngoscopy and bronchoscopy revealed extensive airway haemangiomas. Multiple haemangioma spots were observed on the bilateral middle turbinate, axilla, right superior turbinate (Figure 2),

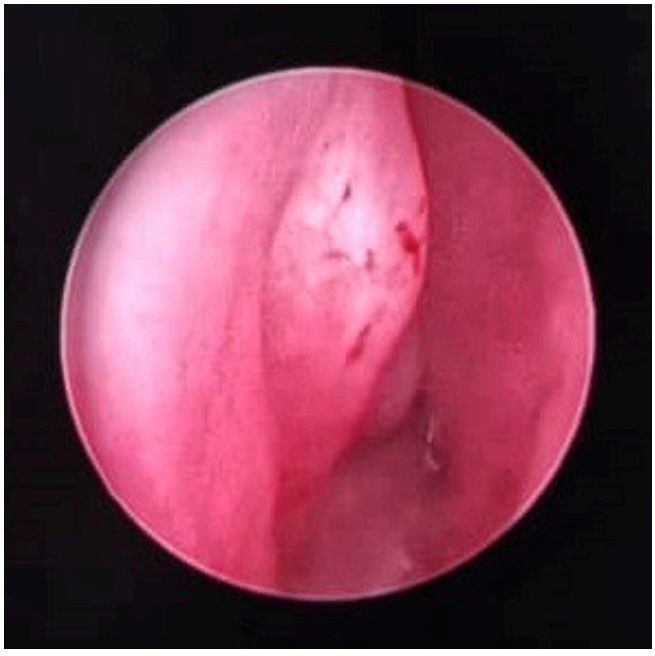

Figure 2. Nasoendoscopy view showing haemangiomatous spots at right middle turbinate.

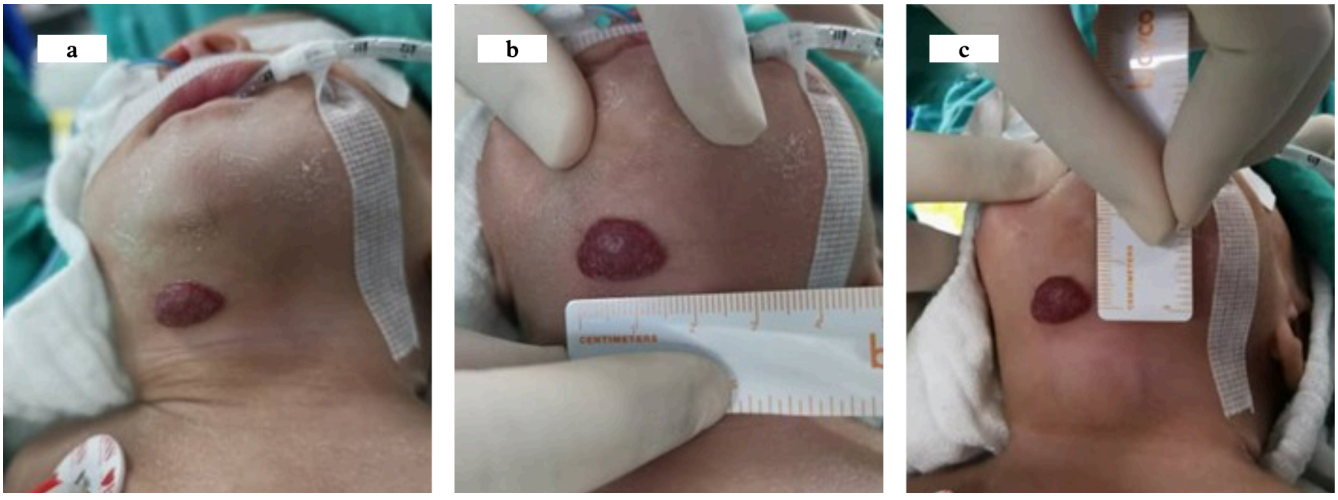

Figure 1a–c. Subcutaneous haemangioma at submental region measuring 1.5 × 1.0 cm.

vocal cord, subcordal region, and circumferentially from the subglottis to the carina and right main bronchus (Figure 3a–3c). The patient was commenced on oral propranolol, starting at 0.5 mg/kg/day for 5 days, then increased to 1.0 mg/kg/day for the next 5 days, followed by 1.5 mg/kg/day for another 5 days, and gradually titrated up to a final dose of 2 mg/kg/day. The echocardiogram revealed normal cardiac structure and function, and the abdominal ultrasound demonstrated normal findings.

Repeated airway assessments were conducted after two weeks of oral propranolol treatment, revealing tracheitis and persistent airway haemangiomas. Haemangioma spots were observed on the right septum, middle turbinate, left middle turbinate, vocal cords, and circumferentially narrowing the entire tracheal wall, with a thin slough over the lower tracheal wall and right bronchus, which bled easily upon touch and suctioning (Figure 4a–4c). The patient was treated with intravenous metronidazole and tazocin and was extubated on the third postoperative day with further weaning. Due to bronchopulmonary dysplasia, he required home nasal CPAP following discharge.

The patient continued on oral propranolol at a dosage 2 mg/kg/day and was monitored regularly in the clinic. A follow-up scope performed four months postoperatively showed that the airway haemangioma had completely resolved. The submental haemangioma had significantly reduced in size, and the parents reported that the noisy breathing had completely resolved.

At 15 months old, the patient underwent direct laryngoscopy and bronchoscopy, which revealed a resolved airway haemangioma (Figure 5a–5c). He was subsequently discharged on a tapering dose of oral propranolol: 4 mg twice daily for one week, then 4 mg once daily for one week, followed by 2 mg once daily for the final week. Eventually, he was able to discontinue CPAP, and the submental haemangioma fully resolved (Figure 6).

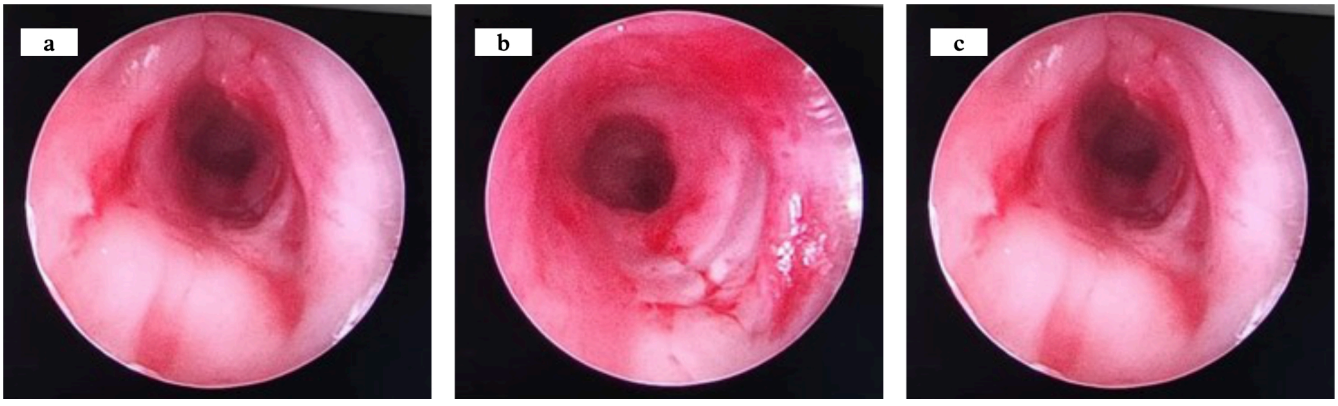

Figure 3. (a) Haaemagiomatous lesions at the vocal cord and subcordal. (b) Circumferential haemangiomatous at the subglottic and tracheal wall. (c) Haemangiomatous lesion over right main bronchus.

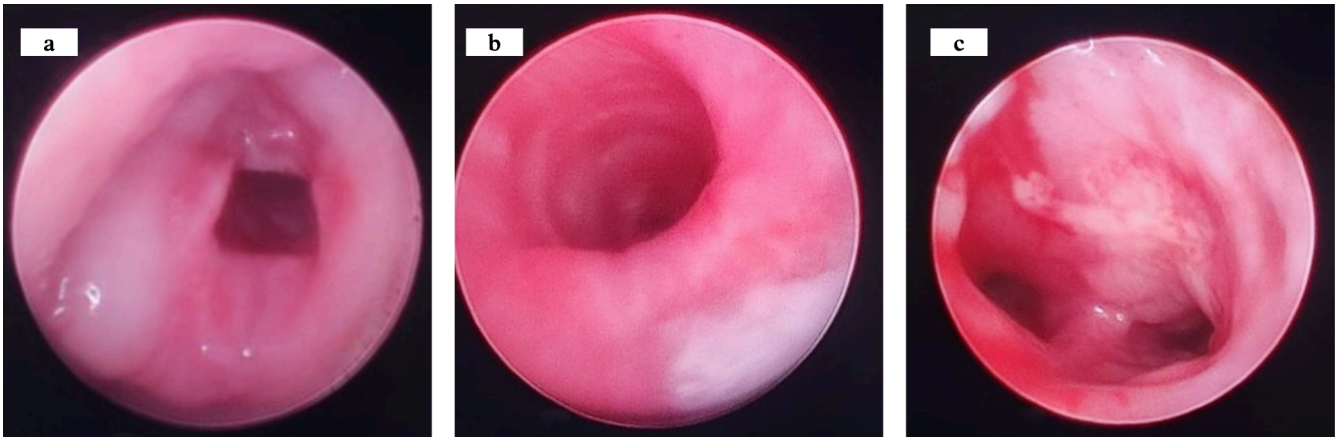

Figure 4. (a) Haemagiomatous lesions at the vocal cord. (b) Circumferential narrowing the entire tracheal wall. (c) Thin slough over the lower tracheal wall and right bronchus.

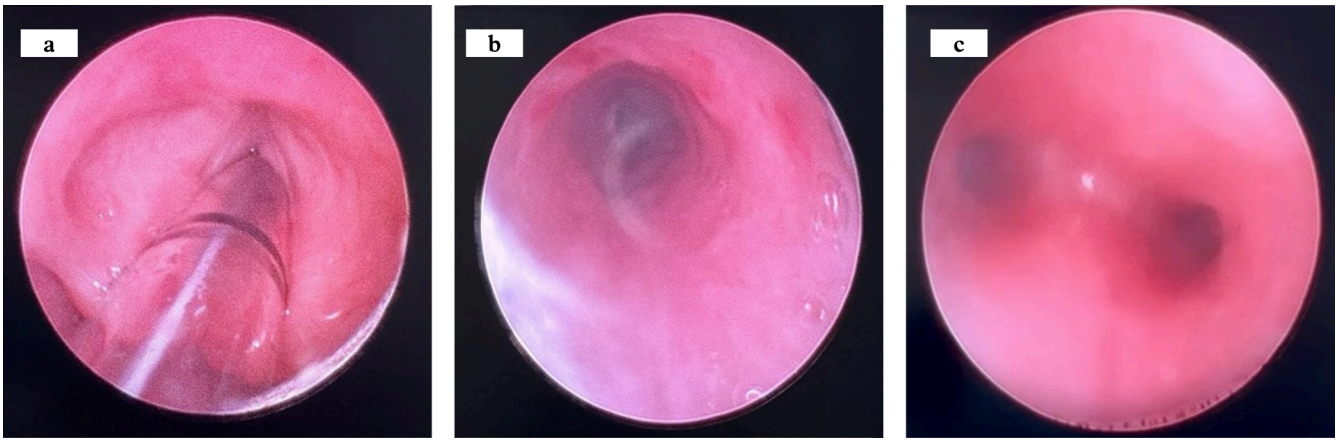

Figure 5. (a) Shows no haemagiomatous lesions at the vocal cord. (b) Trachea. (c) Right and left main bronchus.

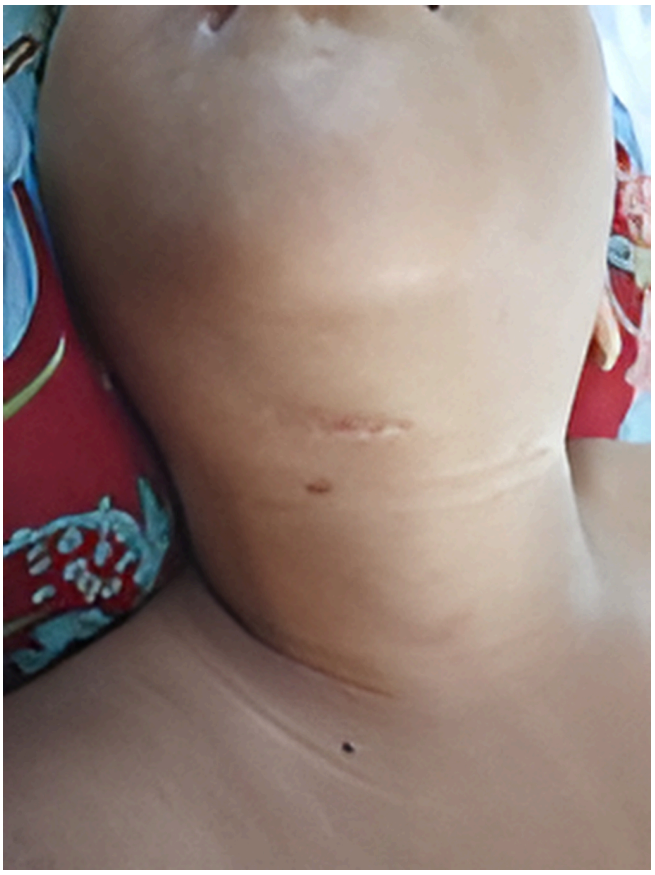

Figure 6. Submental Hemangioma fully resolved.

4. RESULTS AND DISCUSSION

Haemangiomas are benign vascular tumors that proliferate and cause hyperplastic abnormal endothelial and other vascular cells, followed by gradual tumor regression. Infantile hemangioma (IHs) are the most common vascular tumor and the most common benign tumor of infants in 5–10% of cases.[1] These tumours are more common in females, premature infants, those with low birth weight, and infants from multiple gestations.[3] This pattern is reflected in our case as well.

Lesions generally present cutaneously in the head and neck (60%) and can involve extremities (15%), visceral organs, and the brain. IHs can be categorized as localized (focal), segmental, or multifocal based on their anatomical configuration. Orlow et al. were the first to describe the association between beard distribution (chin, jawline, preauricular area, and anterior neck) subcutaneous haemangiomas and a high risk of airway IH, with 63% of patients showing airway involvement.[4] Although airway haemangiomas can occur without an associated cutaneous haemangioma, over 50% of patients with a subglottic haemangioma present with an accompanying cutaneous haemangioma[5] with an even higher rate of 81.6% reported by Uthurriague et al.[6]

Clinically characterized by a proliferative phase followed by an involutive phase, because of spontaneous involution, most IHs do not require treatment. However, 10% of cases require early treatment due to size, location, and complications. IH usually develop within the first four weeks of life and reach their peak size by the age of five months. IH involution typically starts at about twelve months of age and is largely completed by the time the child reaches four years old.[7] This is evident in our case report, where the patient developed symptomatic obstructive airway IHs at 4 months of age during the proliferative phase of the lesion.

IH is typically diagnosed based on clinical presentation and history. The most effective method for rapid initial laryngeal evaluation in an infant with stridor is nasopharyngoscopy.[8] Imaging techniques like CT and MRI are recommended when the diagnosis is uncertain, to assess the extent of lesion, or to monitor treatment response. MRI is the preferred imaging method for evaluating IH-associated anomalies, such as spinal dysraphism, ano genitourinary anomalies, and PHACE syndrome. Choi et al. highlighted that 3D-CT/bronchoscopy is valuable for identifying submucosal haemangiomas missed by laryngoscopy. It also provides a detailed assessment of the lesion's extent, airway narrowing, and response to treatment.[9] When diagnosing IH and screening patients

with multifocal IHs for liver or visceral involvement, ultrasonography is an appropriate first imaging modality.[10]

Propranolol, a nonselective beta-blocker, is the first-line treatment for infantile hemangiomas (IH). Initially reported by Leaute-Labreze et al. in 2008 to cause rapid hemangioma regression[11] it was later confirmed in 2016 to be well-tolerated with few adverse effects.[12] Many published clinical guidelines recommend starting with a dose of 1 mg/kg/day, increasing to 2–3 mg/kg/day.[13–15] According to available studies, the target dosage for treating IH is usually 2–3 mg/kg/day, based on the empirically established maximum dose.[16] However, Tan et al. recommended starting with a dose of 0.7–1.0 mg/kg/day and increasing to 2.0–2.5 mg/kg/day in three divided doses, with dose adjustments made at intervals of at least 3 days.[17] Propranolol's effectiveness in treating cutaneous IH is well-supported by research, including systematic reviews, large studies, and clinical trials.[18] Elluru et al. demonstrated that propranolol is a safe and effective alternative to surgery for treating airway haemangiomas and cutaneous haemangiomas, with fewer complications compared to other therapies. The median treatment duration was 15 months.[2] In our case, we initiated treatment at 0.5 mg/kg/day, increased to 1.0 mg/kg/day after 5 days, and gradually titrated up to 2 mg/kg/day in three divided doses. The propranolol treatment was stopped after 11 months, with the patient showing a consistently good response, including the resolution of both airway and subcutaneous haemangiomas, and no recurrence of symptoms throughout the therapy period. The response of airway haemangiomas to propranolol should be monitored through serial endoscopic evaluations. An initial laryngotracheobronchoscopy is recommended six weeks after initiating treatment to assess response, with follow-up endoscopies scheduled performed at three-month intervals.[19] In this patient, repeat endoscopic evaluations were performed two weeks after initiating treatment and again after an 11-month interval, both demonstrating a favourable treatment response.

The use of propranolol has significantly reduced the need for surgical management of airway infantile haemangiomas (IHs), but surgery remains necessary in cases of severe airway obstruction or failure of medical therapy. Surgical options include intralesional corticosteroid injections, partial or total excision, and lesion debulking. Intralesional steroids, such as triamcinolone and betamethasone, are effective adjunct therapies but may require postoperative intubation. For focal obstructions, endoscopic techniques like laser ablation (CO2, KTP, or Nd:YAG lasers) and microdebriders are preferred, despite risks of recurrence and subglottic stenosis. Open surgical excision is generally considered a second-line therapeutic option for patients unresponsive to propranolol.[20] Postoperative care typically includes intensive monitoring, intubation, and ongoing medical therapy to reduce the likelihood of recurrence.[18]

Untreated airway haemangiomas carry a significant risk and are associated with critically high mortality rates. The prognosis for uncomplicated infantile haemangiomas (IHs) is highly favourable, with most resolving spontaneously by age 4 and causing minimal cosmetic concerns. However, untreated IHs persisting beyond age 6 or those in high-risk anatomical regions are more likely to result in permanent changes such as telangiectasias or scarring, observed in 55% to 69% of cases.[8,14] Frongia et al. reported a recurrence rate of 18% following the discontinuation of propranolol, with a median time of 1.5 months between treatment cessation and regrowth. Recurrences are typically managed by reinitiating therapy and extending the duration of treatment.[21] However, in our case, the patient has remained well up to 2 years and 10 months of age, with no recurrence. Therefore, more frequent monitoring and follow-up are necessary after initiating oral propranolol therapy.

5. CONCLUSION

Infantile haemangiomas are common tumours in infancy, and if unrecognized or untreated, can lead to airway obstruction. Haemangiomas in the beard distribution are strong indicators of potential airway involvement, but even focal subcutaneous haemangioma in the head and neck should raise suspicion. Early recognition and a multidisciplinary approach are essential for effective management. Propranolol is an effective treatment, though it requires regular monitoring to prevent complications and ensure successful outcomes.

Informed consent
The patient's parent provided consent to write and publish this case report.

Ethics approval
None declared.

Conflicts of interest
The authors declare that they have no known financial interests of interest or personal relationships that could have influenced the work reported in this paper.

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Author Contributions
Study design: WQN, AA, FBAR
Data collection: WQN, AA
Statistical analysis: WQN, AA
Data interpretation: WQN, AA
Manuscript preparation: WQN, AA, FBAR
Literature search: WQN, AA, FBAR

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