

Association of Infantile Cutaneous Haemangioma on the Face and Neck with Respiratory Distress in Infancy

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Accepted July 24, 2003.

Sir,

Infantile haemangioma is a fairly common vascular tumour of the skin in infancy. In contrast, infantile subglottic haemangioma is rare, accounting for about 1.5% of all congenital laryngeal anomalies (1). However, it often causes stridor in infancy and sometimes leads to life-threatening respiratory distress (2). We report here a case of infantile haemangiomas on the face, neck and chest that presented with stridor and respiratory distress caused by subglottic haemangioma.

CASE REPORT

A 3-month-old Japanese girl, the product of a normal pregnancy and vaginal delivery, was referred for laser therapy to treat infantile cutaneous haemangiomas. The haemangiomas were noticed just after birth. On physical examination, dark to bright red, irregularly shaped, protuberant tumours were seen on the lower lip, chin, anterior aspect of the neck and upper chest (Fig. 1). The patient was otherwise healthy with no other congenital anomalies, except severe biphasic stridor. Examination by laryngoscopy revealed a submucosal mass in the subglottis causing stenosis of the upper airway. Magnetic resonance imaging demonstrated a sharply demarcated mass extending beyond the tracheal wall and that obstructed more than 50% of the tracheal lumen (Fig. 2). The signal intensity of the lesion was high on T2-weighted sequence, which was enhanced by gadolinium. These findings clearly indicated that her stridor was being caused by a subglottic haemangioma. Over the next few days her biphasic stridor worsened and respiratory distress was noted. A diagnosis of rapidly enlarging subglottic haemangioma was suggested. Systemic steroids were administered ($2 \text{ mg kg}^{-1} \text{ day}^{-1}$ for 2 weeks, $1 \text{ mg kg}^{-1} \text{ day}^{-1}$ for the following week and $0.5 \text{ mg kg}^{-1} \text{ day}^{-1}$ for 2 weeks) and her respiratory condition improved dramatically. Laryngoscopic findings revealed that the size of the subglottic haemangioma was slightly reduced. There was no relapse of the respiratory symptoms. Only for the cutaneous lesions was a flash-lamp-pulsed dye laser treatment performed using a round 7 mm spot size at 7 J/cm^2 .

DISCUSSION

Infantile cutaneous haemangiomas are known to be occasionally accompanied by the presence of haemangiomas



Fig. 1. Infantile haemangiomas on the patient's chin, neck and chest.

internally. Subglottic haemangioma sometimes causes episodic biphasic stridor and respiratory distress.

Approximately 50% of subglottic haemangiomas are reported to have some cutaneous involvement (1). However, in the dermatological literature, the association of infantile haemangioma on the face and neck and subglottic haemangioma has rarely been mentioned. Orlov et al. (3) reported that the presence of cutaneous haemangiomas in a beard-like distribution, i.e. the preauricular regions, chin, lower lip and neck, was highly suggestive of subglottic involvement. In fact, in our case, the infantile haemangiomas were distributed on the lower lip, chin, neck and chest.

The treatment of subglottic haemangioma is still controversial. Some therapeutic approaches, including interferon alfa-2a, oral steroids, submucosal injection of methylprednisolone, CO_2 laser therapy and surgical excision have been tried (4). Laser treatment has been reported to be effective, although this treatment is thought to be inappropriate for bilateral or circular

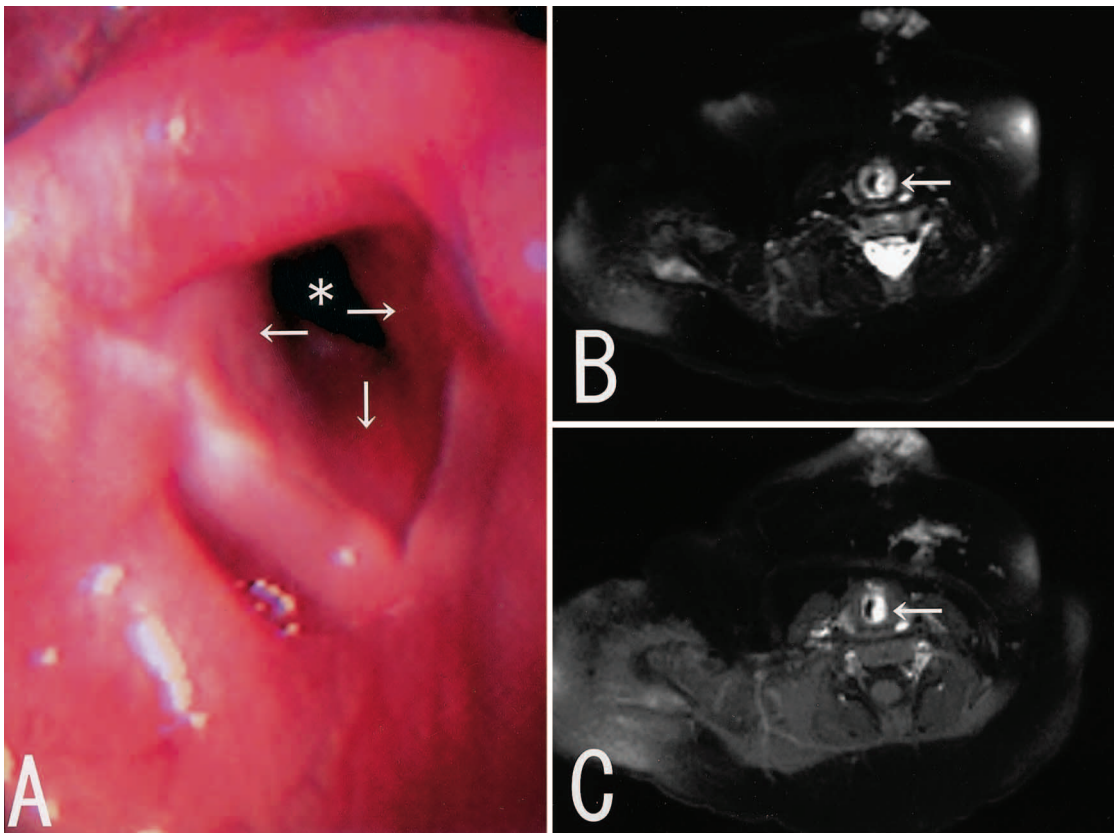


Fig. 2. Laryngoscopy demonstrates the subglottic haemangioma (arrows) protruding into the tracheal lumen (*) and obstructing more than 50% of the airway (A). The signal intensity of the subglottic lesion (arrows) is high on the T2-weighted sequence (B) and is enhanced by gadolinium (C).

haemangiomas because intense laser often results in cicatricial stenosis (4). As for surgical treatment, a new technique of single-stage laryngotracheoplasty (SS-LTR) with short-term intubation has been developed that does not require tracheostomy. This approach is worth trying for endangering cases including bilateral or circular subglottic haemangiomas and corticoreistant patients (5). Systemic steroid therapy, used in our case, is reported to be one of the efficient treatments for this tumour (6).

Subglottic haemangioma has a rapid growth phase as infantile cutaneous haemangiomas. Thus, the sudden onset of respiratory symptoms caused by subglottic haemangiomas, as seen in our case, is not rare and can be life-threatening. In this context, a full examination of the patient's respiratory conditions, including chest auscultation and, if necessary, laryngoscopic observation and magnetic resonance imaging, is recommended for a baby with infantile haemangioma distributed on the face and neck.

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