

CASE REPORT

Airway obstruction caused by rapid enlargement of cervical lymphangioma in a five-month-old boy

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Key Clinical Message

Cervical lymphangioma can cause airway obstruction secondary to enlargement following infection. Physicians should be aware that the airway obstruction can progress rapidly when patients with cervical lymphangioma have respiratory symptoms. Sclerotherapy for lymphangioma can cause both transient swelling and airway obstruction; thus, prophylactic and elective tracheostomy should be considered.

Keywords

Airway obstruction, lymphangioma, sclerotherapy, tracheostomy.

Introduction

Lymphangiomas are rare congenital malformations of the lymphatic system that tend to affect the head and neck [1]. These lesions are histologically benign and usually asymptomatic; therefore, they are usually followed up in the outpatient department. However, life-threatening complications including airway obstruction can arise due to sudden enlargement of a lymphangioma by hemorrhage or infection [1]. Percutaneous sclerotherapy for lymphangioma, such as that by intralesional injection of OK-432, has recently been proposed as an alternative to surgical resection [2]. However, sclerotherapy may take a long time to exhibit an effect, and airway obstruction may occur due to temporary enlargement of the lesion by inflammation. In this report, we describe a five-month-old boy with cervical lymphangioma who needed emergency tracheal intubation because of rapid enlargement of the tumor by infection and underwent prophylactic elective tracheostomy for tracheal compression.

Case Report

A five-month-old boy was brought to the emergency department with fever and stridor associated with rapid growth of a neck mass. One month before presentation, a soft mass had been found on the left side of his neck, and cervical lymphangioma was diagnosed by computed tomography and magnetic resonance imaging. He had no respiratory symptoms and was followed as an outpatient. One day before presentation, he developed a fever of 39°C and stridor when crying. On arrival, his oxygen saturation was 99% and stridor was noted with crying, but no effort was observed while breathing. He was admitted to the hospital, and antibiotics were administered. However, the stridor worsened and his oxygen saturation declined to 80% despite oxygen administration; therefore, he was intubated and ventilated. Fentanyl, midazolam, and rocuronium were administered for induction of anesthesia. A 3.5-mm tracheal tube was inserted. Chest and neck computed tomography revealed a large cystic mass that compressed the trachea (Fig. 1). Fine needle

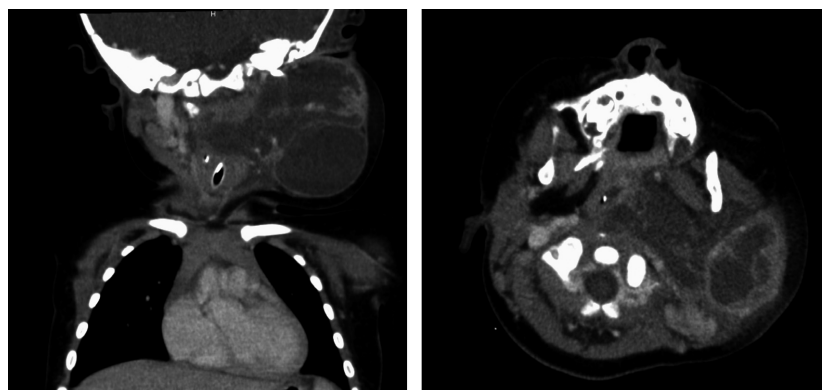


Figure 1. Computed tomography showing the multilocular cystic mass progressing from the left parotid gland to the pharyngeal space and compressing the trachea.

aspiration of the cyst was performed, and 20 mL of serous liquid was removed; however, the cervical swelling did not improve. Intralesional injection of OK-432 was planned, but there was concern that transient swelling due to the local inflammatory response after OK-432 injection would cause airway obstruction and prolonged intubation. Moreover, after OK-432 injection and transient swelling, reintubation seemed likely to be more difficult if accidental extubation occurred. Therefore, tracheostomy was performed by an otolaryngologist, and OK-432 was then injected. After transient swelling and erythema, the mass markedly decreased in size. Three weeks after OK-432 injection, the tracheostomy tube was removed.

Discussion

Lymphangiomas are rare congenital malformations of the lymphatic system that affect 1 in 6000–16,000 live-born neonates. Approximately 75% of lymphangiomas occur in the head and neck region [3]. They are histologically benign and commonly asymptomatic, so affected patients are often followed up as outpatients without any treatment. However, lymphangiomas of the neck can cause life-threatening complications, including airway obstruction [1]. Patients can present to the emergency department with airway obstruction resulting from sudden enlargement of a lymphangioma by hemorrhage or infection. Respiratory distress secondary to a lymphangioma can progress rapidly, and intervention to secure the airway should be considered immediately. In this case, the respiratory distress progressed rapidly, and the patient required emergency tracheal intubation. In such cases of upper airway obstruction, the clinician should consider having the anesthesiologist intubate the patient and avoid using muscle relaxants.

Indications for treatment of lymphangioma are compression of surrounding structures such as the airway, cosmetic problems, and recurrent infection [4]. Various methods are available for treatment of lymphangioma, including surgical excision and sclerotherapy with bleomycin or OK-432 [2]. Complete surgical excision is the definitive approach to lymphangioma, but it is often difficult to achieve because of diffuse lesions and the risk of damage to important organs [4]. Percutaneous sclerotherapy has been proposed as an alternative to surgical resection. Bleomycin has been used, but it must be used with extreme caution because of the risk of pulmonary fibrosis [5]. Several previous reports have suggested that intralesional injection of OK-432 should be a first-line treatment for lymphangioma [2].

Lymphangioma is classified as cystic lymphangioma, cavernous lymphangioma, and lymphangioma simplex [6]. It can appear at any site, but cystic lymphangioma is seen in the cervical region more commonly than is cavernous lymphangioma. OK-432 is reportedly more effective for cystic lymphangioma than cavernous lymphangioma. Cavernous lymphangioma is often intractable to medical treatment [7]. Additionally, sildenafil is reportedly effective for severe lymphangioma. In cases that are intractable to medical treatment, the use of sildenafil might be considered [8]. In the present case, computed tomography showed a multilocular cystic mass progressing from the left parotid gland to the pharyngeal space. Serous liquid was removed by fine needle aspiration; the mass was thus regarded as a cystic lymphangioma, and OK-432 injection was effective.

OK-432 injection produces a local inflammatory reaction that leads to shrinkage of the lesion [2]. OK-432 injection is safe in the majority of children, but complications of OK-432 injection have been reported, including transient pyrexia, swelling, tenderness, severe upper

airway obstruction, and enlargement of the mass [4]. When a lymphangioma seen around the trachea or cervical region is treated with intralesional OK-432, the physician should be aware that such airway complications may occur and prepare for possible airway obstruction.

Airway protection is a major indication for tracheostomy [9, 10]. Complications of tracheostomy include granuloma, infection, and bleeding. Granuloma is induced by contact of the airway wall with the tracheostomy tube and increases in size as the duration of tube placement becomes longer. The timing of and indications for tracheostomy in children are still controversial. The reported average mechanical ventilation period before tracheostomy is 26–32 days [9, 11, 12], which is longer than that in our case. On the other hand, two patients with lymphangioma reportedly died of hypoxic damage related to difficulty with reintubation after endotracheal tube displacement [13]. In our case, reintubation seemed difficult when accidental extubation occurred. Securing the airway was given the highest priority in our patient; therefore, prophylactic elective tracheostomy was performed with a shorter period of mechanical ventilation than in previous reports. In cases of cervical lymphangioma with tracheal compression, when reintubation seems difficult or the duration of intubation may be prolonged, securing the airway is most important and prophylactic and elective tracheostomy should be considered.

Conclusion

Physicians should be aware of the risks of airway obstruction caused by sudden enlargement of cervical lymphangioma. Furthermore, because of transient swelling related to injection of OK-432, an intervention to secure the airway may be needed. In patients with tracheal compression, when reintubation seems difficult or the duration of intubation may be prolonged, prophylactic and elective tracheostomy should be considered.

Informed Consent

Informed consent to report this case was obtained.

Conflict of Interests

None declared.

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