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Case report

Sympathetic neuropathy and dysphagia following doxycycline sclerotherapy

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ABSTRACT

This case report demonstrates neurologic sequela following treatment with doxycycline sclerotherapy. A six-week-old child presented with respiratory distress from a macrocystic lymphatic malformation, extending from the skull base to the anterior mediastinum. Following doxycycline sclerotherapy, the airway symptoms resolved; however, the child developed silent aspiration and Horner's syndrome. Two months following treatment the patient resumed oral diet and at one year post-intervention there has been no recurrence of symptoms, with only mild ptosis remaining.

While neuropathies following doxycycline sclerotherapy have been described, aspiration has never been documented. This case demonstrates a single patient's clinical course and resolution of their neuropathies.

1. Introduction

Lymphatic malformations (LM) are common, benign head and neck tumors seen in children [1–4]. They are the result of errors in embryonic development leading to abnormal lymphatic capillaries that become cystic [1–5]. These cystic spaces can enlarge, especially from infection or an inflammatory response, and compress surrounding structures, including the airway and esophagus [1–3,5].

There exist several different interventions for LM including sclerotherapy and surgical excision [2–6]. Multiple studies have shown the effectiveness and excellent safety profile of doxycycline as a sclerosant used in sclerotherapy [1–7]. Potential complications from doxycycline sclerotherapy typically include pain, fever, infection, and skin irritation [1,3–7]. Neurologic complications, including Horner's syndrome (unilateral miosis, ptosis, with or without anhidrosis), have proven to be rare with only a limited number of documented cases in larger studies [1,5,7,8]. There have been no documented cases of aspiration following doxycycline sclerotherapy. Furthermore, none have detailed the individual presentation and imaging of LM, course of doxycycline sclerotherapy treatment, resulting complications of Horner's syndrome and aspiration, and resolution.

We are reporting the case of a six-week-old child who underwent doxycycline sclerotherapy for a macrocystic LM compressing on the airway, resulting in complications of Horner's syndrome and silent aspiration.

2. Case report

A six-week-old female infant presented to the Children's Hospital of Wisconsin from an outside hospital (OSH) with a four-day history of respiratory distress. The patient was born full term via cesarean section due to maternal tachycardia. No other complications during pregnancy or following birth. Over the previous four days, the patient experienced respiratory distress, cough, rhinorrhea, sneeze, and decreased enteral intake without aspiration symptoms. On the fourth day, the patient was admitted to the pediatric intensive care unit (PICU) of an OSH due to continued respiratory distress, including tracheal tugging and intercostal retractions. The patient received Decadron, nebulized epinephrine, and was placed on continuous positive airway pressure (CPAP). She continued to have signs of respiratory distress, which necessitated intubation after transfer to the Children's Hospital of Wisconsin. Workup for infection, including lumbar puncture, blood culture, and urine culture, was negative. Following failure of extubation trials, flexible and rigid bronchoscopies were performed and showed no evidence of tracheal disease, but demonstrated a retropharyngeal soft tissue mass. A computed tomography (CT) scan with contrast of the neck further demonstrated a large, predominantly right-sided, sharply demarcated and homogenous lesion that extended inferiorly to the thyroid gland. A magnetic resonance imaging (MRI) with contrast of the neck was performed to characterize the cystic nature of the mass and established a diagnosis of macrocystic LM due to

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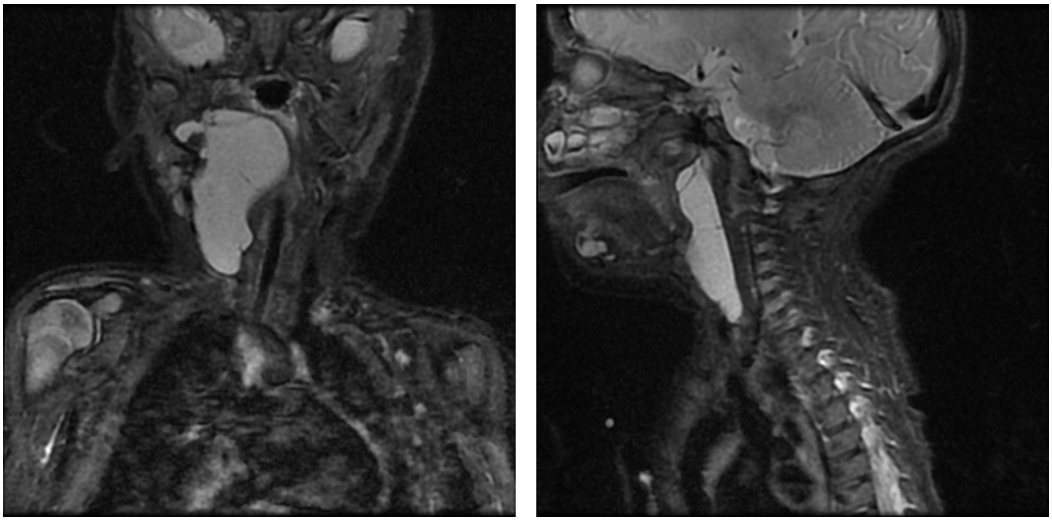

Fig. 1. Pre-treatment MRI of the neck.

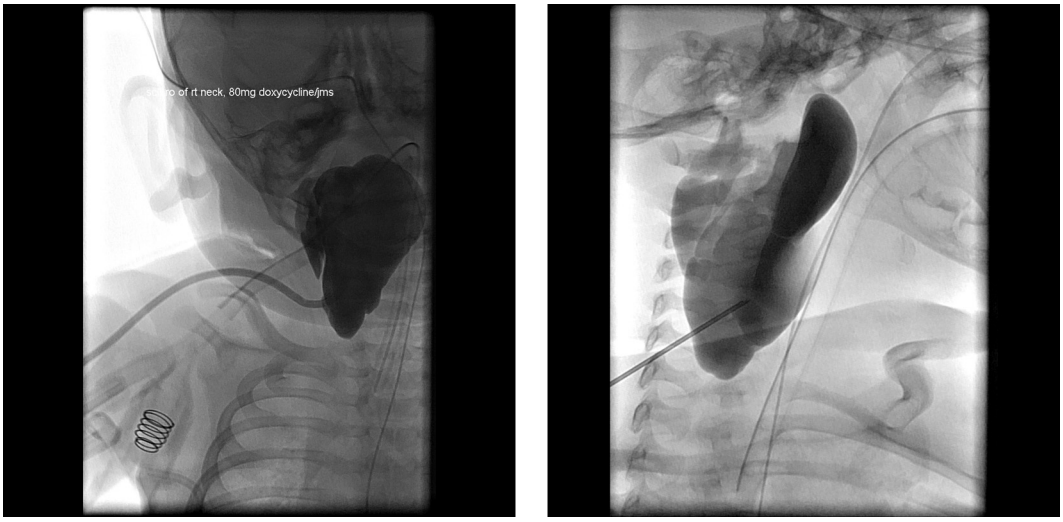

Fig. 2. Doxycycline sclerotherapy under fluoroscopy.

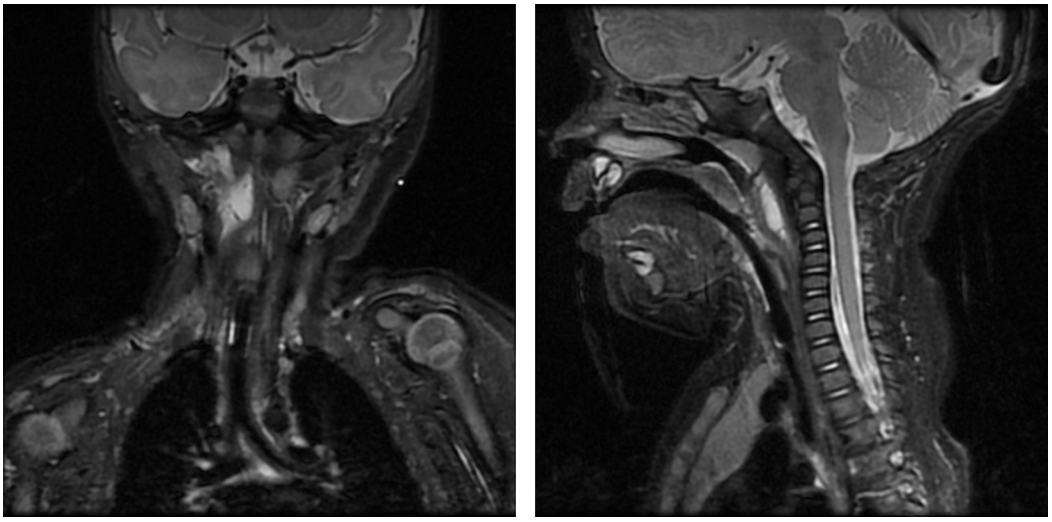

Fig. 3. Post-treatment MRI of the neck.