Radiology Case Reports xxx (2017) 1–4

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: http://Elsevier.com/locate/radcr

Case Report

Emergent sclerotherapy of a newborn with expanding lymphatic malformation causing respiratory distress

Cameron C. Sheehan BS[a], *Winslo Idicula MD*[b], *Andrew Raabe MD*[c], *Kris Jatana MD*[d], *Tendy Chiang MD*[d], *Charles A. Elmaraghy MD*[d,*]

[a] *The Ohio State University College of Medicine, Columbus, OH, USA*
[b] *Department of Otolaryngology-Head and Neck Surgery, Wexner Medical Center at the Ohio State University, Columbus, OH, USA*
[c] *Department of Radiology, Nationwide Children's Hospital, Columbus, OH, USA*
[d] *Department of Pediatric Otolaryngology, Nationwide Children's Hospital, 700 Children's Drive, Columbus, OH, USA*

ARTICLE INFO

Article history:
Received 18 October 2016
Received in revised form
28 December 2016
Accepted 2 January 2017
Available online xxx

Keywords:
Lymphatic malformation
Sclerotherapy
Stridor
Respiratory distress

ABSTRACT

The present report describes a case of acute airway obstruction in a newborn caused by an expanding hemorrhagic macrocystic lymphatic malformation (LM), which was successfully treated with emergent decompression and interventional radiology-guided sclerotherapy. The use of sclerotherapy for macrocystic LMs has been well described for various indications. The urgent interventional treatment obviated the need for a tracheostomy. This case describes the rapid diagnosis and use of sclerotherapy in a large expanding macrocystic LM.

Introduction

Lymphatic malformations (LMs) are benign abnormalities that typically present in the head and neck region as painless neck masses. Fifty percent of these lesions are noted to be at birth and 80%-90% of these lesions are diagnosed before the age of 2 years [1]. Although the etiology of LM is still unclear, recent studies have shown intrinsic and extrinsic factors to play a role, including inappropriate expression of lymphatic specific molecules and increased number of interferon producing cells [1–3]. LMs account for only 5% of all benign pediatric lesions and frequently involve the head and neck region. However, because of their variable clinical course, fluctuation in size, and mass effect on nearby structures, they may pose life threatening risks to this population [4].

We present a unique case of a rapidly enlarging macrocystic LM in a 17-day-old newborn that was thought to be due to hemorrhage. The patient underwent emergent airway

Acknowledgments: This project is supported by intramural funding from the Department of Otolaryngology and the Center for Surgical Outcomes Research at Nationwide Children's Hospital.
Competing Interests: The authors have declared that no competing interests exist.
* *Corresponding author.*
E-mail address: charles.elmaraghy@nationwidechildrens.org (C.A. Elmaraghy).
http://dx.doi.org/10.1016/j.radcr.2017.01.003

evaluation and intervention with resultant intubation and sclerotherapy. Intraoperative evaluation revealed a 5.3 × 4.2 × 3.3 cm cyst in posterior pharynx and deep neck deviating the trachea anteriorly. This case demonstrates intracystic hemorrhage in a newborn with an LM and describes successful decompression and sclerotherapy treatment obviating the use of a tracheostomy.

Methods

Our institution does not require approval for retrospective studies such as this. This retrospective case report includes clinical history, intraoperative findings, radiographic findings, and a review of the current literature.

Case presentation

A 17-day-old normal full-term female presented to the emergency room of a tertiary pediatric hospital with a 1-day history of acute onset respiratory distress and enlarging neck mass over the course of an hour. She had no significant medical history, and there was no reported history of trauma, foreign body aspiration, or previous intubations. On arrival, she demonstrated increased work of breathing, gasping with marked retractions, and cyanosis. She was placed on 100% nonrebreather mask and given racemic epinephrine. Jaw thrust with positioning improved oxygen saturations to the 90s. Urgent otolaryngology evaluation revealed an enlarging neck mass with impending airway obstruction, which prompted the decision to secure the airway in the operating room.

She underwent direct laryngoscopy, which revealed massive anterior retropharyngeal displacement effacing the laryngeal surface and obstruction of the airway. Her airway was secured with a 3-0 uncuffed endotracheal tube, and patient was then transferred to the computed tomography (CT) scanner to investigate developmental (thyroglossal duct cysts, branchial cleft cysts, etc), inflammatory and/or infectious, and neoplastic causes.

Contrast CT scan of the neck demonstrated a large bilobed cystic lesion measuring 5.3 cm × 4.2 cm × 3.3 cm displacing the trachea anteriorly with evidence of increased attenuation consistent with internal hemorrhage (Figs. 1 and 2). Her findings were consistent with a large macrocystic LM with acute hemorrhage; she was then transferred to intervention radiology to undergo sclerotherapy.

Using fluoroscopic guidance and ultrasound, a 5-French catheter was placed and subsequently aspirated 13 milliliters of bloody fluid. There was noted to be significant decompression of the cyst with resolution of mass effect. The cavity was irrigated and chemical thrombolysis was performed on the remaining clot, which was completely aspirated. Sclerotherapy was then performed with 5 milliliters of alcohol with subsequent aspiration of 6 milliliters of bloody fluid (Fig. 3). The drain was secured in place and patient was taken back to the operating room for further evaluation of her airway.

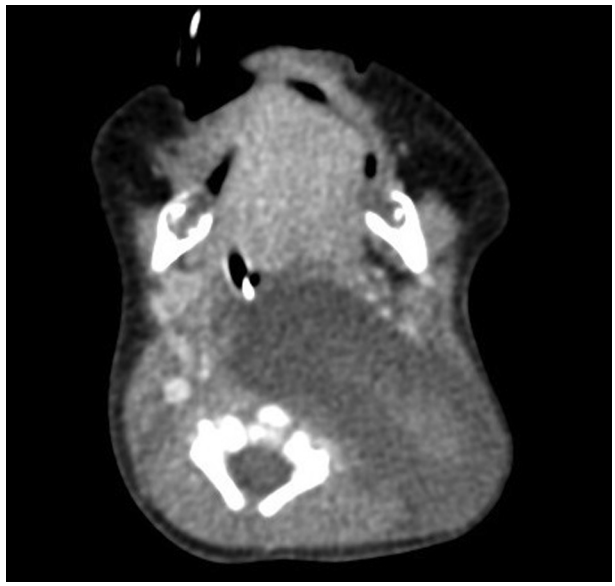

Fig. 1 – Computed tomography (CT) scan axial view demonstrating macrocystic malformation.

Repeat direct laryngoscopy revealed resolution of mass effect with slight retropharyngeal fullness and an uncompromised view of the larynx. The 3-0 uncuffed tube was replaced with a 3-0 cuffed tube without difficulty. Bronchoscopy showed no evidence of tracheomalacia. After completion of the procedure, she was placed on prophylactic cefazolin and transferred to the Pediatric Intensive Care Unit for further monitoring.

The patient continued to improve through her hospital course. The 5-French drain was removed on postoperative day three (POD 3). She was subsequently weaned from the ventilator and after bedside direct laryngoscopy revealed

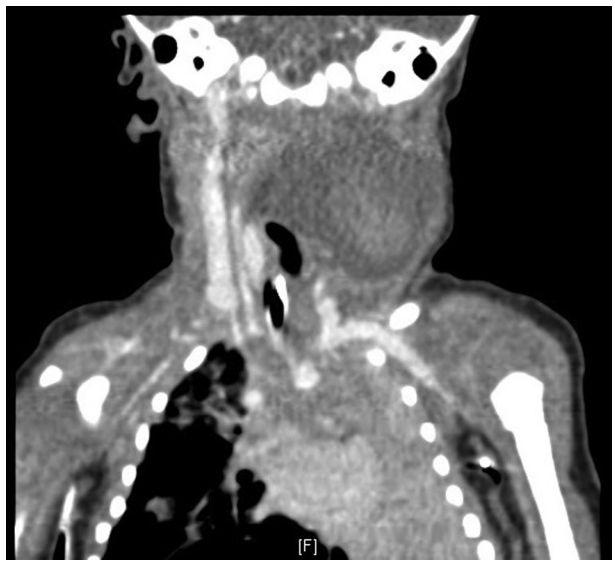

Fig. 2 – CT scan coronal view demonstrating compression of the airway.