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CASE REPORT

MR Lymphangiography for Focal Disruption of the Thoracic
Duct in Chylothorax of an Infant: a Case Report
and Literature Review[△]

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Abstract Chylothorax is a rare cause of pleural effusion in children, and it is usually difficult to identify the location of chyle leakage due to the small size of the thoracic duct in children. Herein we report an infant case with chylothorax whose leakage of the thoracic duct was successfully located by magnetic resonance lymphangiography (MRL) using pre-contrast MR cholangiopancreatography (MRCP) and gadodiamide-enhanced spectral presaturation inversion recovery (SPIR) T₁-weighted imaging, which demonstrate the imaging method is easy and effective for detecting the focal disruption of the thoracic duct in children with chylothorax and younger than 8 months old.

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CHYLOTHORAX is a condition of chyle accumulation in the pleural space caused by thoracic duct injury. It is a rare cause of pleural effusion in infants. Chylothorax is diagnosed by a triglyceride level exceeding 110 mg/dL or detection of chylomicrons in the pleural fluid. Identifying the site of chyle

leakage is important for guidance of medical therapy, but it is usually difficult in children due to the small size of the thoracic duct.

Pedal or intranodal lymphangiography, which involves injection of ethiodized oil into the pedal interstitial lymphatics or inguinal lymph nodes, can detect chyle leakage in various conditions such as chylothorax and lymphatic fistulae. Interstitial lymphangiography, which requires an incision to expose the pedal lymphatics, is time-consuming and difficult to perform in children due to the small size of

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their pedal lymphatic vessels.¹ Intranodal lymphangiography requires general anesthesia to ensure complete immobility for intranodal injection and following CT scan to delineate the lymphatic vessels. Its application in children is limited for the slow anesthetic drug injection rate in children and radiation exposure. Single-photon emission computed tomography/CT (SPECT/CT)¹ and near-infrared fluorescence (NIRF) lymphatic imaging² are also useful for detecting focal thoracic duct damage, but they are time-consuming, only provide limited resolution, and require special equipment that is not widely available.

Nonenhanced magnetic resonance lymphangiography (MRL) is readily available in most hospitals. It provides high spatial resolution and soft tissue contrast without radiation exposure. However, it cannot effectively distinguish lymphatic tissue from nonlymphatic tissue.³ Here we report the application of gadodiamide as a contrast agent for MRL and discuss the potential of gadodiamide-enhanced MRL for detecting the focal disruption of the thoracic duct in children with chylothorax.

CASE DESCRIPTION

This study was approved by our Institutional Review Board. Written informed consent was obtained from the patient's parents. A 6-month-old male infant was hospitalized for cough, wheeze and phlegm for 3 days. He had no history of fever, night sweats, anorexia, trauma, or cardiac or lymphatic problems. He exhibited shortness of breath and depression of the suprasternal fossa, supraclavicular fossa and intercostal space during inspiration. Physical examination revealed no rhonchi and moist crackles in both lung, and no abnormalities of the cardiac, nervous, or abdominal systems. Chest film and CT showed pleural effusion in the right thoracic cavity. Thoracentesis was performed, and 80 mL milky exudative fluid was aspirated. Laboratory evaluation detected chylomicrons in the aspirated fluid, and the diagnosis of chylothorax was confirmed. Nasal dietary therapy (5% hydrolysate of ultrafiltrated whey-dominated milk protein, 30 mL/3 h, Alfare Nestlé Health Science) and intravenous partial parenteral nutrition were administered subsequently, omitting fat from the diet. After thoracentesis, the pleural effusion was sustained for 1 month with free liquid deepness measured by ultrasound fluctuating between 0.9 cm and 3.9 cm; therefore, MRL was performed.

Thoracic magnetic resonance imaging (MRI) experiments were performed using a clinical 3.0 T MRI scanner (Achieva, Philips Medical Systems) with a 16-channel phased-array coil. Undiluted gadodiamid solution (1.0 mL, 0.2 mmol/kg,

Omniscan, GE Healthcare, Ireland) was injected subcutaneously into the dorsum of both feet at multiple locations within 1 minute. The injection sites were massaged slightly for approximately 30 seconds after injection. To alleviate pain-induced anxiety, 0.2 mg diazepam (40 µg/kg) was administered intravenously. During MRI, the patient was freely breathing in the headfirst supine position, and sedated with 10% chloral hydras (3 mL/kg) administered rectally.

Axial T1-weighted (T1W) turbo field echo in-phase (TFE IP) images were acquired before and 20, 23, 27, 31 min after gadodiamid injection, with repetition time (TR)/echo time (TE)=10ms/2.3ms, flip angle (FA)=15°, acquisition matrix (MTX)=178×178, reconstruction MTX=324×432, field of view (FOV)=267mm×355 mm, section thickness=5 mm, and acquisition time=1 min 59 s. Pre-contrast MRL was performed using a MR cholangiopancreatography (MRCP) sequence with TR/TE=1926 ms/740 ms, FA=90°, acquisition MTX=205×205, reconstruction MTX=512×512, FOV=300 mm×300 mm, section thickness=2 mm, and acquisition time=2 min 21 s. Post-contrast MRL was performed using a 3D gradient-recalled-echo (GRE) sequence at 5, 16, 22, 26, and 30 min after gadodiamid injection, with TR/TE=4.6 ms/1.6 ms, FA=27°, acquisition MTX=412×412, reconstruction MTX=560×560, FOV=280mm×280 mm, section thickness=1.2 mm, and acquisition time=50 s; and a spectral presaturation inversion recovery (SPIR) T1W imaging sequence at 6 min after gadodiamid injection with TR/TE=606 ms/20 ms, FA=90°, acquisition MTX=241×241, reconstruction MTX=256×256, FOV=150mm×156 mm, section thickness=2.5 mm, and acquisition time=8 min 36 s. MRCP and 3D GRE images were reconstructed with multiple planar reconstruction (MPR) and coronal maximum intensity projection (MIP).

The thoracic duct was not shown on pre- and post-contrast T1W TFE IP images or on the 3D GRE MIP image (data not shown). On the MRCP MPR image (Fig. 1A), the thoracic duct at the 9th and 10th thoracic vertebral levels measured approximately 4 mm in diameter and showed a strong hyperintense signal. The remaining thoracic duct measured less than 1 mm in diameter and showed a faint hyperintense signal. The MRCP MIP image only revealed the dilated part of the thoracic duct (data not shown). The 3D GRE MPR image displayed the full length of the thoracic duct, showing a faint hyperintense signal (Fig. 1B). On the SPIR T1W images at 9th and 10th thoracic vertebral levels (Fig. 2), a hyperintense strip in the pleural effusion was found connecting to the thoracic duct, suggesting thoracic duct damage. The conservative therapy was continued, and the pleural effusion persisted, with free liquid deepness

measured by ultrasound fluctuating between 2.8 cm and 4.0 cm. However, the patient's clinical symptoms diminished significantly. Two months after thoracentesis, the parents checked the patient out of the hospital without consulting the doctor in charge.

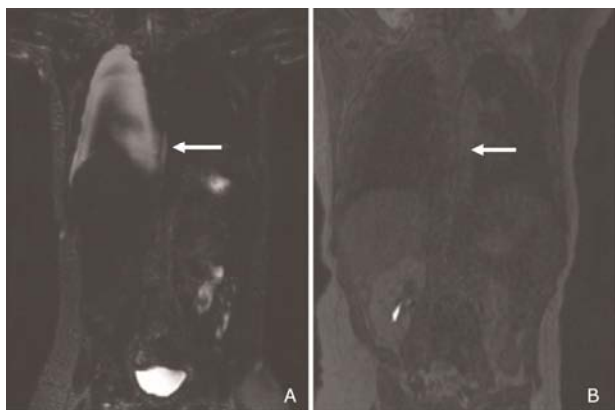


Figure 1. Multiple planar reconstruction (MPR) images showing the thoracic duct.

A. Reconstructed coronal image of the magnetic resonance cholangiopancreatography (MRCP). The thoracic duct (arrow) presented hyperintense signal with approximate diameter of 4 mm at the levels of the 9th and 10th thoracic vertebrae, whereas the remaining thoracic duct was slender, presented a faint hyperintense signal with a diameter less than 1 mm.

B. Reconstructed coronal image of the 3D gradient recalled echo sequence (GRE). Part of the thoracic duct (arrow) presented a mildly hyperintense signal.

DISCUSSION

Unlike nonenhanced MRL, contrast-enhanced MRL can specifically detect lymphatic vessels, but lymphographic MRI contrast agents including Gadofluorine 8, Gadofluorine M, and Gadofluorine P have not been available for clinical use yet.⁴ Gadodiamide is a safe and efficacious gadolinium-based MRI contrast agent for specific visualization of blood vessels that is approved by the Food and Drug Administration (FDA). In this report, we demonstrated the feasibility of gadodiamide-enhanced MRL for detecting focal thoracic duct damage in an infant younger than 8 months old.

Gadodiamide-enhanced SPIR T1W images of the patient showed signs of chyle leakage from the enlarged thoracic duct that measured 4 mm in diameter on pre-contrast MRCP MPR images. According to a study by Yu *et al*⁵, the range of diameter and mean maximum transverse diameter of the thoracic duct were 1.6–6.9 mm and 3.7 ± 0.4 mm, respectively in healthy controls ($n=100$). There has been no previous report on the thoracic duct diameter in children to our best knowledge. We retrospectively reviewed the MRCP data from 72 children aged 2–50 months (mean 21 months) who attended our hospital between January 2015 and June 2015, the range of diameters of thoracic duct were 1.2–2.6 mm, and the mean maximum transverse diameter was 2.02 ± 0.32 mm. Therefore, the maximum diameter of the thoracic duct was expected to be smaller than 3 mm in children, suggesting that the enlarged thoracic duct in this case was abnormal. Furthermore, there was a strip of hyperintense signal observed, which was connected with the enlarged thoracic duct. These

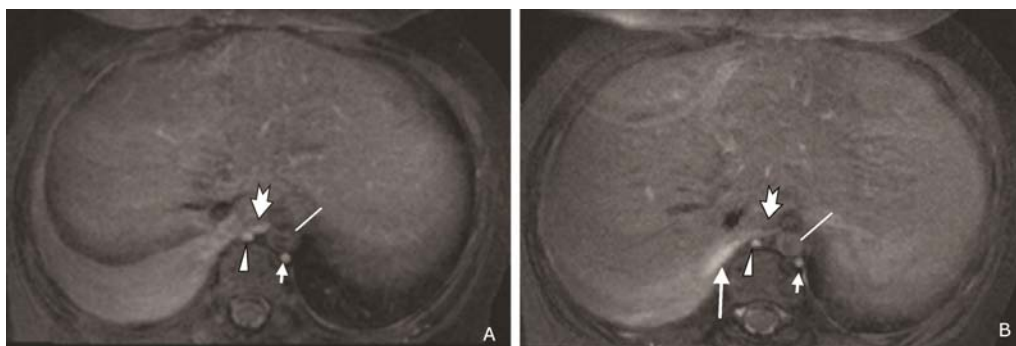


Figure 2. Post-contrast spectral presaturation inversion recovery (SPIR) T1-weighted axial images at levels from the 9th (A) to the 10th (B) thoracic vertebra.

A. The azygos vein (arrow head) and the hemiazygos vein (short arrow) were presented clearly beside the thoracic vertebrae. The thoracic duct (dovetail arrow) located between the azygos vein (arrow head) and the aorta (straight line), presenting slightly high signal intensity, with a larger diameter than that of azygos vein.

B. A strip of hyperintense signal (long arrow) extended from the thoracic duct (dovetail arrow) to the right pleural effusion at the level of the 10th thoracic vertebra, demonstrating the site of chyle leakage.

evidences strongly suggested focal damage of the thoracic duct, which was similar to the findings reported by Deso *et al* based on axial post-lymphangiography CT imaging.⁶ The SPIR T1W imaging sequence is a fast spin echo sequence that offers high resolution. However, it is time consuming, taking more than 5 minutes, and can be affected by serious artifacts at the level of the heart, which could interfere with the visualization of the thoracic duct.

The T1-weighted 3D fast low-angle shot sequence (equivalent to the 3D GRE sequence), which takes less than 10 seconds, has been the primary sequence used in interstitial MRL in recent years.^{7,8} This sequence is also compatible with the MIP technique for depicting the morphological features of the thoracic duct. In our case, the thoracic duct showed a faint signal on the 3D GRE MPR image but was not identifiable on the 3D GRE MIP image. This was likely due to the small size of the thoracic duct in infants, and also could be caused by interference of a high signal of blood vessel, because gadodiamide is a small molecule that can diffuse into the blood circulation soon after interstitial injection.

In conclusion, this report demonstrates that MRL using non-contrast MRCP and gadodiamide-enhanced SPIR T1W imaging can be an easy and effective method for detecting a focal disruption of the thoracic duct in children with chylothorax.

Statement of conflict of interests

All authors have no conflict of interests to disclose.

REFERENCES

1. Yang J, Codreanu I, Zhuang H. Minimal lymphatic leakage in an infant with chylothorax detected by lymphoscintigraphy SPECT/CT. *Pediatrics* 2014; 134: e606-10. doi: 10.1542/peds.2013-2689.
2. Tan IC, Balaguru D, Rasmussen JC, Guillod R, Bricker JT, Douglas WI, et al. Investigational lymphatic imaging at the bedside in a pediatric postoperative chylothorax patient. *Pediatr Cardiol* 2014; 35:1295-300. doi: 10.1007/s00246-014-0946-y.
3. Dori Y, Zviman MM, Itkin M. Dynamic contrast-enhanced MR lymphangiography: feasibility study in swine. *Radiology* 2014; 273:410-6. doi: 10.1148/radiol.14132616.
4. Kiryu S, Inoue Y, Sheng F, Watanabe M, Yoshikawa K, Shimada M, et al. Interstitial MR lymphography in mice: comparative study with gadofluorine 8, gadofluorine M, and gadofluorine P. *Magn Reson Med Sci* 2012; 11:99-107. doi: 10.2463/mrms.11.99.
5. Yu DX, Ma XX, Zhang XM, Wang Q, Li CF. Morphological features and clinical feasibility of thoracic duct: detection with nonenhanced magnetic resonance imaging at 3.0 T. *J Magn Reson Imaging* 2010; 32:94-100. doi: 10.1002/jmri.22128.
6. Deso S, Ludwig B, Kabutay NK, Kim D, Guermazi A. Lymphangiography in the diagnosis and localization of various chyle leaks. *Cardiovasc Intervent Radiol* 2012; 35:117-26. doi: 10.1007/s00270-010-0066-x.
7. Ruehm SG, Schroeder T, Debatin JF. Interstitial MR lymphography with gadoterate meglumine: initial experience in humans. *Radiology* 2001; 220:816-21. doi: 10.1148/radiol.2203010090
8. Turkbey B, Kobayashi H, Hoyt RF Jr, Choyke PL, Nakajima T, Griffiths GL, et al. Magnetic resonance lymphography of the thoracic duct after interstitial injection of gadofosveset trisodium: a pilot dosing study in a porcine model. *Lymphat Res Biol* 2014; 12: 32-6. doi: 10.1089/lrb.2013.0029.