Direct contrast-enhanced magnetic resonance lymphangiography in the diagnosis of persistent occult chylous effusion leak after thoracic duct embolization

Sharon C. Kiang, MD,[a,b] Khwaja A. Ahmed, MD,[c] Samual Barnes, PhD,[c] Ahmed M. Abou-Zamzam Jr, MD,[a] *and* Roger T. Tomihama, MD,[c] *Loma Linda, Calif*

ABSTRACT

Thoracic duct embolization (TDE) is currently the technique of choice for chylothorax refractory to medical management. Diagnosis and treatment of persistent lymphatic leakage after an initially successful TDE are complicated by inadequate imaging to localize the nidus of the lymphatic leak. Traditional imaging modalities including nuclear medicine lymphoscintigraphy, magnetic resonance lymphangiography, and fluoroscopic lymphangiography lack spatial resolution and dynamic physiologic real-time evaluation. We report a contemporary imaging technique using direct contrast-enhanced magnetic resonance lymphangiography to diagnose an occult chylous leak after TDE. (J Vasc Surg: Venous and Lym Dis 2019;7:251-7.)

Keywords: Chylothorax; Lymphangiography; Thoracic duct embolization; Direct contrast magnetic resonance lymphangiography

Chylothorax is an uncommon pleural effusion resulting from the leakage of lymphatic fluid from the thoracic duct (TD) and its supplying tributaries. Conventional management of chylothorax consists of nutrition control (nonfat diet or total parenteral nutrition), surgery (TD ligation, pleurodesis), and, recently, percutaneous therapy (TD embolization [TDE]).[1-4] TDE has a high success rate and minimal complications.[5] However, technical success of TDE does not always lead to clinical success.[6-10]

Traditional lymphatic imaging modalities were nuclear medicine lymphoscintigraphy, unenhanced or intravenous contrast-enhanced magnetic resonance (MR) lymphangiography, and conventional fluoroscopic lymphangiography,[11-16] all of which lack detailed spatial resolution or dynamic physiologic real-time evaluation. Direct contrast-enhanced MR lymphangiography (DCMRL) and direct contrast-enhanced intralymphatic cone beam computed tomography (CT) are new techniques that allow direct enhancement of the lymphatic channels with cross-sectional imaging, making them robust diagnostic tools to evaluate lymphatic disease.[17,18]

From the Division of Vascular Surgery, Department of Surgery,[a] and Section of Vascular and Interventional Radiology, Department of Radiology,[c] Loma Linda University School of Medicine; and the Division of Vascular Surgery, Department of Surgery, VA Loma Linda Healthcare System.[b]

Author conflict of interest: none.

Correspondence: Roger T. Tomihama, MD, Assistant Professor, Vascular and Interventional Radiology, Department of Radiology, Loma Linda University Medical Center, 11234 Anderson St, Ste MC-2605E, Loma Linda, CA 92354 (e-mail: rtomihama@llu.edu).

The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2213-333X

https://doi.org/10.1016/j.jvsv.2018.09.013

This is a successful case report of occult chylous leak detected by DCMRL not seen with conventional lymphangiography. Informed consent was obtained from the patient for this report.

CASE REPORT

A 45-year-old man with a past medical history of hypertension, pulmonary embolism, chronic obstructive pulmonary disease, congestive heart failure, and myocardial infarction presented with persistent pleural effusions. Thoracentesis revealed elevated triglyceride levels (639 mg/dL; normal, <100 mg/dL) and presence of chylomicrons, confirming the diagnosis of a chylous effusion. With failure of conservative therapy and with no history of traumatic TD injury, the diagnosis of an idiopathic chylous effusion was made. After discussion between cardiothoracic surgery, medical intensive care unit, and interventional radiology services, the decision was made for percutaneous TDE.

TD lymphangiography and embolization were performed by standard technique. Under ultrasound guidance, the inguinal lymph nodes were punctured with a 25-gauge needle, and 5 to 10 mL of lipiodol was slowly infused (Fig 1, *A*). Intermittent fluoroscopy was used to track the lipiodol contrast agent into the upper retroperitoneal lymphatics. Under direct "gunsight" fluoroscopy guidance (Fig 1, *B*), the cisterna chyli was punctured with a 21-gauge × 15-cm Chiba needle (Cook Medical, Bloomington, Ind) and cannulated with a 0.018-inch microwire (V-18 ControlWire; Boston Scientific, Natick, Mass). The Chiba needle was exchanged over the microwire for a microcatheter (Rapid Transit; Johnson & Johnson, Miami Lakes, Fla). TD injection on conventional digital subtraction angiography and cone beam CT demonstrated no clearly visualized leak (Fig 2). A prophylactic TDE was performed using multiple 3-mm microcoils (Nester microcoils; Boston Scientific) and *n*-butyl cyanoacrylate glue (Trufill; Cordis, Johnson & Johnson, Warren, NJ; Fig 3).

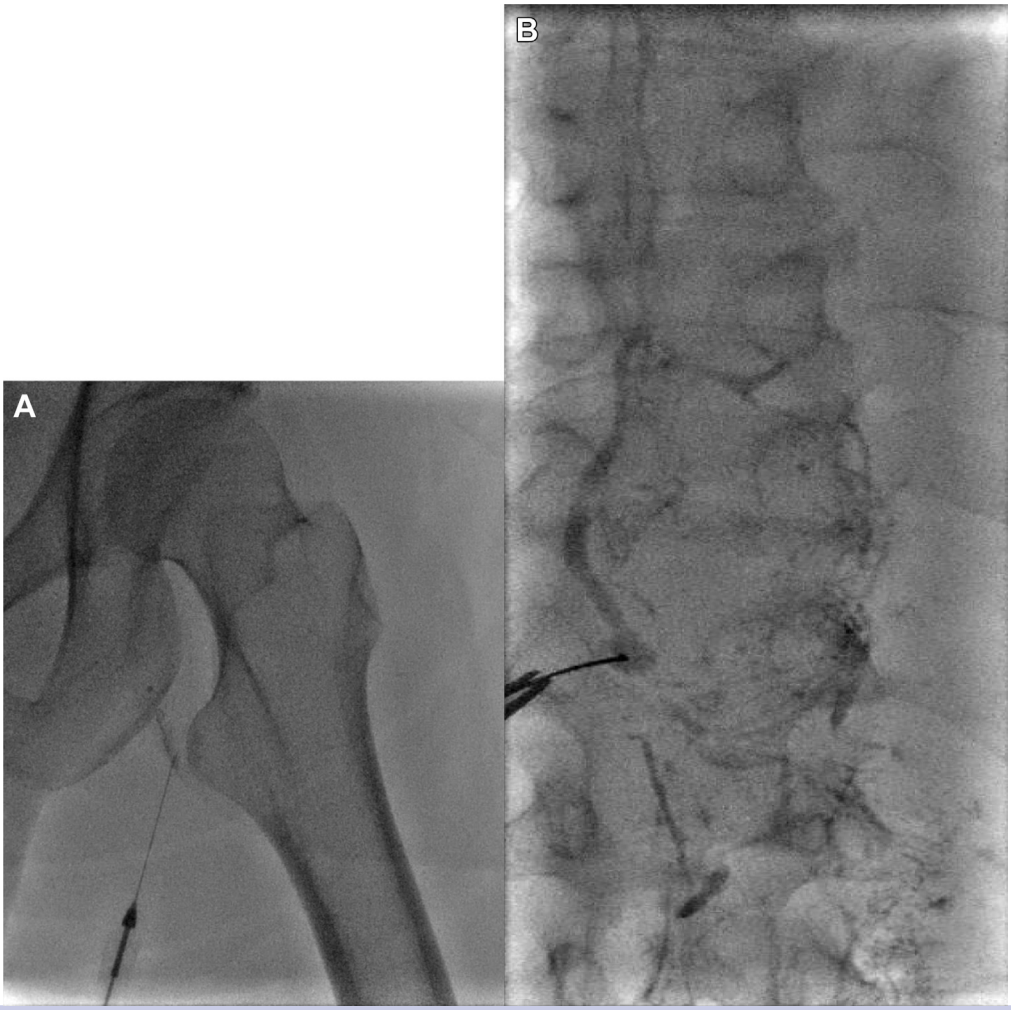

Fig 1. The thoracic duct (TD) was accessed by standard technique with lipiodol lymphangiography. **A,** Ultrasound guidance of left inguinal lymph node puncture was performed with a 25-gauge needle and injection of lipiodol. **B,** Fluoroscopy-guided cisterna chyli puncture with a 21-gauge Chiba needle.

After the procedure, the patient failed to improve with the TDE. We proceeded with DCMRL to evaluate for an occult lymphatic leak. The DCMRL examination was performed on a 1.5T Aera scanner (Siemens, Erlangen, Germany). Beginning in the fluoroscopy suite, the inguinal lymph nodes were punctured under ultrasound guidance with a 25-gauge needle, and 5 to 10 mL of iodinated contrast material was slowly infused. Intermittent fluoroscopy confirmed iodinated contrast material tracking into the lower retroperitoneal lymphatics, confirming proper cannulation of lymphatic channels and not venous cannulation or extravasation into the inguinal soft tissues. The access needle in the groin was secured, and the patient was transferred to the MR imaging suite. A gadolinium contrast agent (gadobenate dimeglumine) was injected into the lymphatic system while dynamic MR images were being acquired (Fig 4). Similar to previously described technique, three-dimensional T1-weighted MR imaging was repeated dynamically for 17 minutes after injection to observe transit of the gadolinium contrast agent through the lymphatic system.[18-20] High-resolution static three-dimensional T1-weighted MR images were then acquired at 30 and 40 minutes after injection. The DCMRL revealed a previously unseen occult chylous fluid leak from the right retroperitoneum into the right pleural space (Fig 5). These leaks were not visible at 17 minutes after injection and only clearly visible at 30 minutes after injection.

Because of the location and size, this occult leak was not amenable to further percutaneous intervention, especially because the TD had already been embolized. Technical limitations to percutaneous treatment of small, deep, retroperitoneal lymphatic channels include their small size and lack of structure, which cannot support microcatheter-based therapy. Subsequently, the patient was medically managed for his persistent chylous effusions.

DISCUSSION

We report a successful case using DCMRL to diagnosis a persistent occult chylous leak after failure of both conventional lymphangiography and prophylactic TDE.

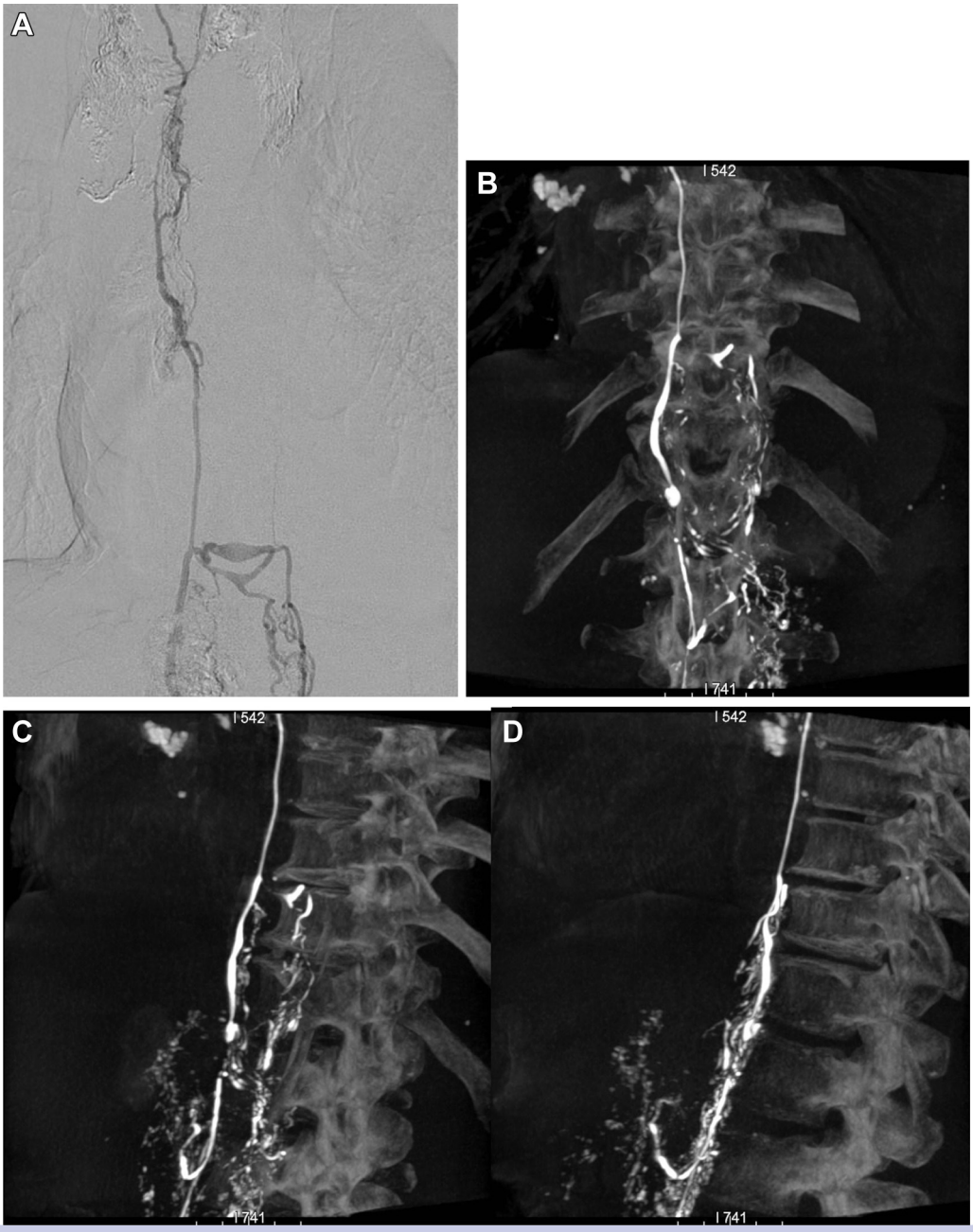

Fig 2. Opacification of the thoracic duct (TD) revealed no visualized leak to the pleural spaces. **A,** Conventional digital subtraction angiography lymphangiogram of the chest and upper abdomen demonstrates no clearly visualized leak. **B-D,** Anteroposterior, left anterior oblique, and lateral projections from a cone beam computed tomography (CT) lymphangiogram of the chest also confirm no clearly visualized leak.

Clinical outcomes of TDE have been mixed. Previous studies demonstrate clinical success of TDE or TD disruption ranging from 45% to 74%.[6-10] The mixed clinical success result may be due to decreased sensitivity of conventional lymphangiography compared with DCMRL for detecting small occult lymphatic leaks.

Previously, imaging for the lymphatic system was limited to nuclear medicine lymphoscintigraphy, unenhanced or intravenous contrast-enhanced MR lymphangiography, or conventional fluoroscopic lymphangiography, each with its advantages and disadvantages.[11-16] Nuclear medicine lymphoscintigraphy and conventional fluoroscopic lymphangiography are powerful because they are dynamic studies that allow real-time continuous evaluation of slow-flowing lymphatic channels. However, these two modalities

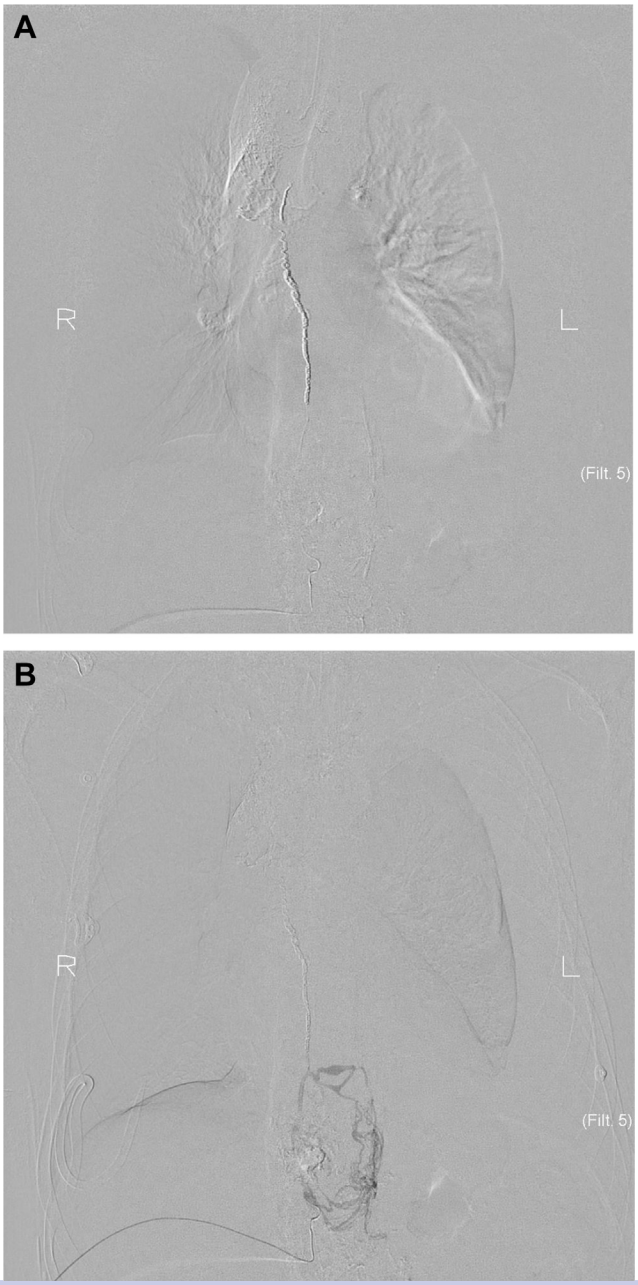

Fig 3. Embolization of the thoracic duct (TD) was performed using multiple platinum-fibered coils and *n*-butyl cyanoacrylate glue. **A,** Embolization of the TD with multiple coils. **B,** After embolization injection, no opacification of the TD or any visualized leak is demonstrated.

do not provide cross-sectional organ and soft tissue detail. Unenhanced or intravenous contrast-enhanced MR lymphangiography visualizes organ and soft tissue contrast detail but is limited because it is a nondynamic study, has poor visualization of the lymphatics, and seldom demonstrates an active leak.[21-25]

The main challenge to obtaining meaningful dynamic cross-sectional images of the lymphatic system is the difficulty in introducing a contrast agent into the lymphatic vessels. DCMRL and intralymphatic contrast-enhanced cone beam CT are new techniques that allow direct enhancement of the lymphatic channels with cross-sectional imaging and are powerful diagnostic tools to evaluate lymphatic disease.[17,18] Potential applications include assessment of fluoroscopically occult chylous leaks, assessment of the severity and extent of lymphatic obstruction, visualization of complex collateral pathways, detection of lymphangiectasia and lymphatic reflux, and guidance for MR interventions that involve the TD.[18]

Intrinsic properties of gadolinium may be one reason that DCMRL was successful. Viscosity of gadolinium in saline preparation is more similar to lymphatic fluid compared with lipiodol. This is evident in the transit time of the intranodal injection. Intranodal gadolinium takes up to only 15 to 30 minutes to opacify the retroperitoneal lymphatics, whereas intranodal lipiodol can take up to 1 to 2 hours.

Lipiodol also has some baseline embolic properties. In conventional lipiodol lymphangiography, there may be temporary embolism of very small lymphatic channels, and thus small occult leaks may not be seen on real-time lipiodol lymphangiography. Matsumoto et al[26] reported that lymphatic leakage was stopped after lymphangiography alone without the need for intervention in eight of nine patients (89%).

Lastly, because DCMRL was performed after the TD procedure, it is possible that the cause of the persistent chylous effusion was an iatrogenic injury from the TDE procedure or that the lymphatic came off a parallel channel. Although possible, it is unlikely because a post-TDE injection of contrast material demonstrated no new leak (Fig 3, *B*), and the remainder of the open TD, adjacent connected lymphatic channels, and percutaneous track were sealed with *n*-butyl cyanoacrylate glue at the conclusion of the procedure.

CONCLUSIONS

We report the use of DCMRL in diagnosis of a persistent occult chylous leak after TDE. The literature reports clinical success rates of TDE ranging from 45% to 74%.[6-10] As demonstrated by this case, the mixed clinical success result may be due to the decreased sensitivity of conventional lymphangiography and TDE compared with DCMRL for detecting small occult lymphatic leaks. The application of this technique should be validated with clinical trials.

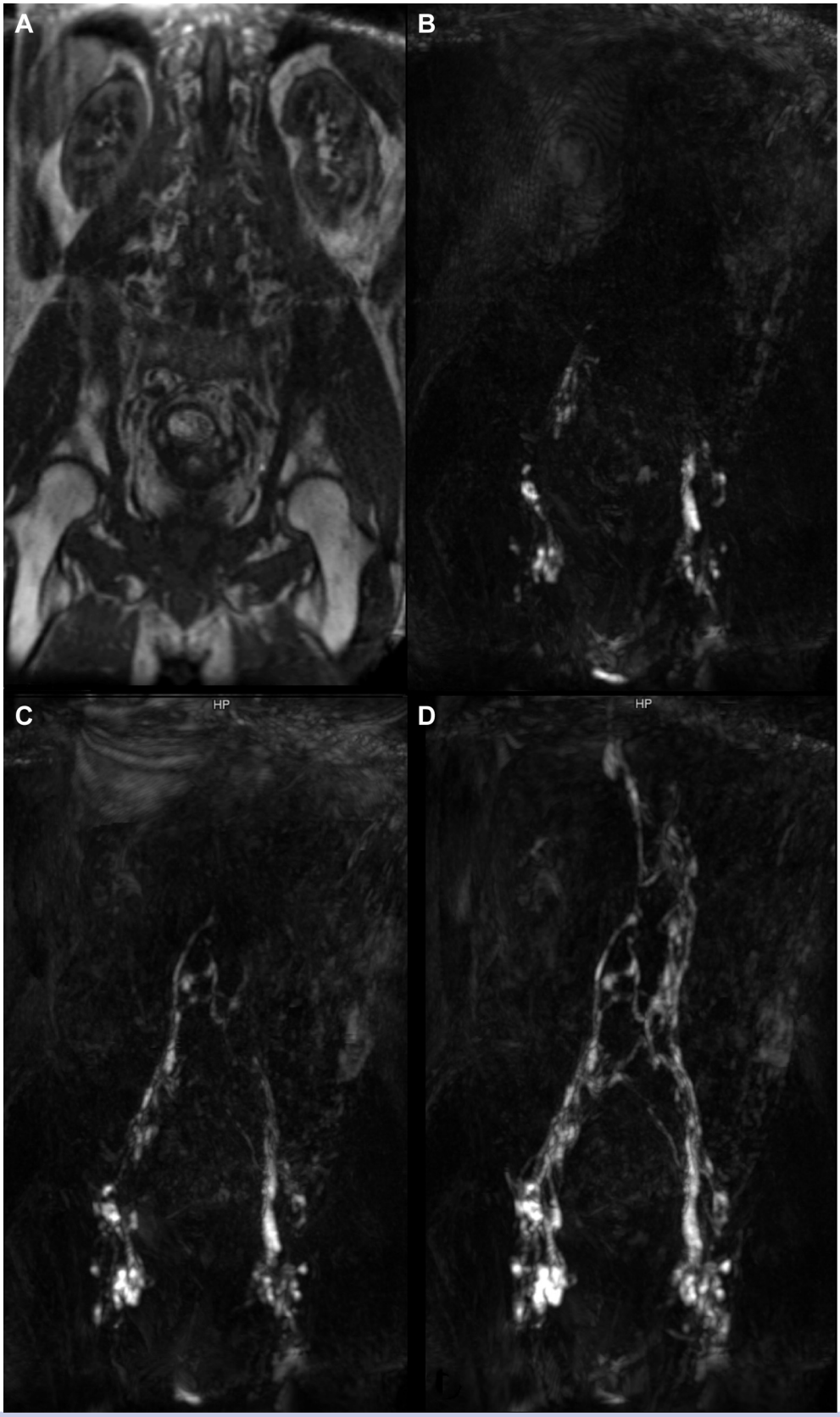

Fig 4. Direct contrast-enhanced magnetic resonance lymphangiography (DCMRL) was performed by again accessing the inguinal lymph nodes and then injecting gadolinium into the lymphatic system. **A,** Baseline coronal T1 image of the abdomen and pelvis. **B-D,** Coronal fat-suppressed T1 intranodal images after gadolinium injection demonstrate sequential opacification of the retroperitoneal lymphatics.

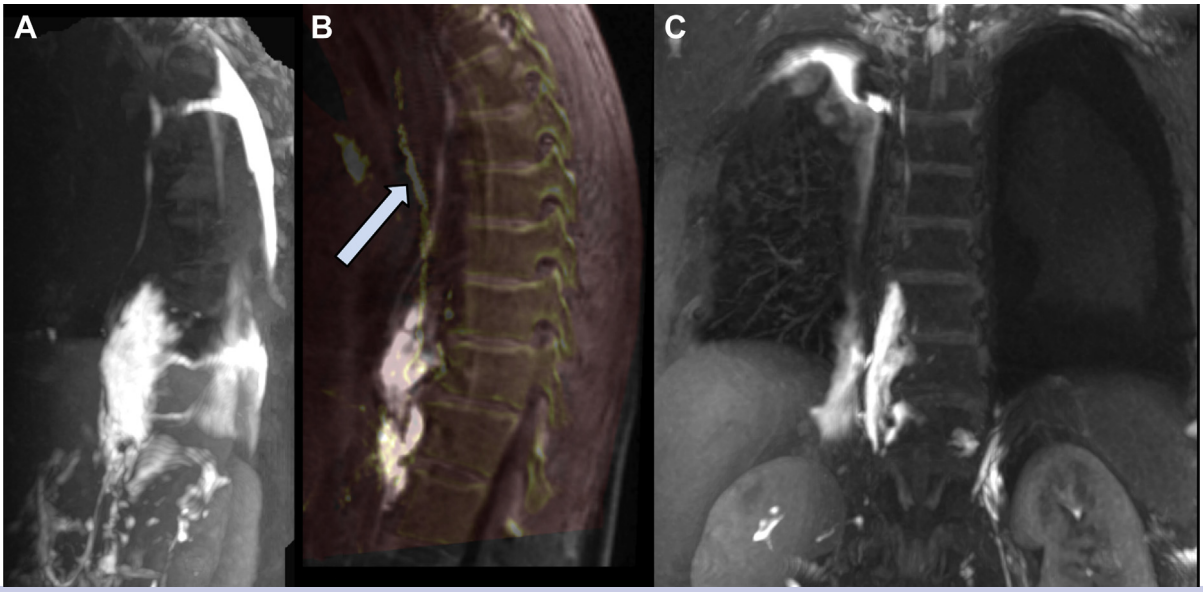

Fig 5. Three-dimensional reconstruction magnetic resonance (MR) lymphangiogram demonstrating the lymphatic leak in the right upper pleural space. **A** and **B,** Sagittal three-dimensional maximum intensity projection image of chest lymphatics demonstrating the leak. **C,** Coronal three-dimensional maximum intensity projection image of chest lymphatics demonstrating the leak. An occult chylous fluid leak from the right retroperitoneum into the right pleural space that was not seen on conventional fluoroscopic lymphangiography is identified. The embolization coils in the occluded thoracic duct (TD) are delineated by the *arrow.*

REFERENCES

1. Itkin M, Kucharczuk J. Thoracic duct embolization (TDE) for nontraumatic chylous effusion: experience in 31 patients. Chest 2010;138:654A.
2. Marts BC, Naunheim KS, Fiore AC, Pennington DG. Conservative versus surgical management of chylothorax. Am J Surg 1992;164:532-4; discussion: 534-5.
3. Christodoulou M, Ris HB, Pezzetta E. Video-assisted right supradiaphragmatic thoracic duct ligation for nontraumatic recurrent chylothorax. Eur J Cardiothorac Surg 2006;29:810-4.
4. Thompson KJ, Kernstine KH, Grannis FW Jr, Mojica P, Falabella A. Treatment of chylothorax by robotic thoracic duct ligation. Ann Thorac Surg 2008;85:334-6.
5. Chen E, Itkin M. Thoracic duct embolization for chylous leaks. Semin Intervent Radiol 2011;28:63-74.
6. Cope C, Kaiser LR. Management of unremitting chylothorax by percutaneous embolization and blockage of retroperitoneal lymphatic vessels in 42 patients. J Vasc Interv Radiol 2002;13:1139-48.
7. Boffa DJ, Sands MJ, Rice TW, Murthy SC, Mason DP, Geisinger MA, et al. A critical evaluation of a percutaneous diagnostic and treatment strategy for chylothorax after thoracic surgery. Eur J Cardiothorac Surg 2008;33:435-9.
8. Itkin M, Kucharczuk JC, Kwak A, Trerotola SO, Kaiser LR. Nonoperative thoracic duct embolization for traumatic thoracic duct leak: experience in 109 patients. J Thorac Cardiovasc Surg 2010;139:584-9; discussion: 589-90.
9. Pamarthi V, Stecker MS, Schenker MP, Baum RA, Killoran TP, Suzuki Han A, et al. Thoracic duct embolization and disruption for treatment of chylous effusions: experience with 105 patients. J Vasc Interv Radiol 2014;25:1398-404.
10. Yannes M, Shin D, McCluskey K, Varma R, Santos E. Comparative analysis of intranodal lymphangiography with percutaneous intervention for postsurgical chylous effusions. J Vasc Interv Radiol 2017;28:704-11.
11. Clément O, Luciani A. Imaging the lymphatic system: possibilities and clinical applications. Eur Radiol 2004;14:1498-507.
12. Notohamiprodjo M, Weiss M, Baumeister RG, Sommer WH, Helck A, Crispin A, et al. MR lymphangiography at 3.0 T: correlation with lymphoscintigraphy. Radiology 2012;264:78-87.
13. Arrivé L, Azizi L, Lewin M, Hoeffel C, Monnier-Cholley L, Lacombe C, et al. MR lymphography of abdominal and retroperitoneal lymphatic vessels. AJR Am J Roentgenol 2007;189:1051-8.
14. Lu Q, Xu J, Liu N. Chronic lower extremity lymphedema: a comparative study of high-resolution interstitial MR lymphangiography and heavily T2-weighted MRI. Eur J Radiol 2010;73:365-73.
15. Cox SJ, Kinmonth JB. Lymphography of the thoracic duct. A new technique for improving visualization. J Cardiovasc Surg 1975;16:120-2.
16. Dori Y, Keller MS, Fogel MA, Rome JJ, Whitehead KK, Harris MA, et al. MRI of lymphatic abnormalities after functional single-ventricle palliation surgery. AJR Am J Roentgenol 2014;203:426-31.
17. Ierardi AM, Pappalardo V, Liu X, Wu CW, Anuwong A, Kim HY, et al. Usefulness of CBCT and guidance software for percutaneous embolization of a lymphatic leakage after thyroidectomy for cancer. Gland Surg 2016;5:633-8.
18. Krishnamurthy R, Hernandez A, Kavuk S, Annam A, Pimpalwar S. Imaging the central conducting lymphatics: initial experience with dynamic MR lymphangiography. Radiology 2015;274:871-8.
19. Dori Y, Keller MS, Rome JJ, Gillespie MJ, Glatz AC, Dodds K, et al. Percutaneous lymphatic embolization of abnormal pulmonary lymphatic flow as treatment of plastic bronchitis

in patients with congenital heart disease. Circulation 2016;133:1160-70.
20. Dori Y. Novel lymphatic imaging techniques. Tech Vasc Interv Radiol 2016;19:255-61.
21. Okuda I, Udagawa H, Hirata K, Nakajima Y. Depiction of the thoracic duct by magnetic resonance imaging: comparison between magnetic resonance imaging and the anatomical literature. Jpn J Radiol 2011;29:39-45.
22. Okuda I, Udagawa H, Takahashi J, Yamase H, Kohno T, Nakajima Y. Magnetic resonance-thoracic ductography: imaging aid for thoracic surgery and thoracic duct depiction based on embryological considerations. Gen Thorac Cardiovasc Surg 2009;57:640-6.
23. Takahashi H, Kuboyama S, Abe H, Aoki T, Miyazaki M, Nakata H. Clinical feasibility of noncontrast-enhanced magnetic resonance lymphography of the thoracic duct. Chest 2003;124:2136-42.
24. Itkin M, Krishnamurthy G, Naim MY, Bird GL, Keller MS. Percutaneous thoracic duct embolization as a treatment for intrathoracic chyle leaks in infants. Pediatrics 2011;128:e237-41.
25. Laor T, Hoffer FA, Burrows PE, Kozakewich HP. MR lymphangiography in infants, children, and young adults. AJR Am J Roentgenol 1998;171:1111-7.
26. Matsumoto T, Yamagami T, Kato T, Hirota T, Yoshimatsu R, Masunami T, et al. The effectiveness of lymphangiography as a treatment method for various chyle leakages. Br J Radiol 2009;82:286-90.

Submitted Jul 25, 2018; accepted Sep 11, 2018.