Thoracic duct stenting as a treatment for chylothorax and chylous ascites in patient with Yellow Nail Syndrome

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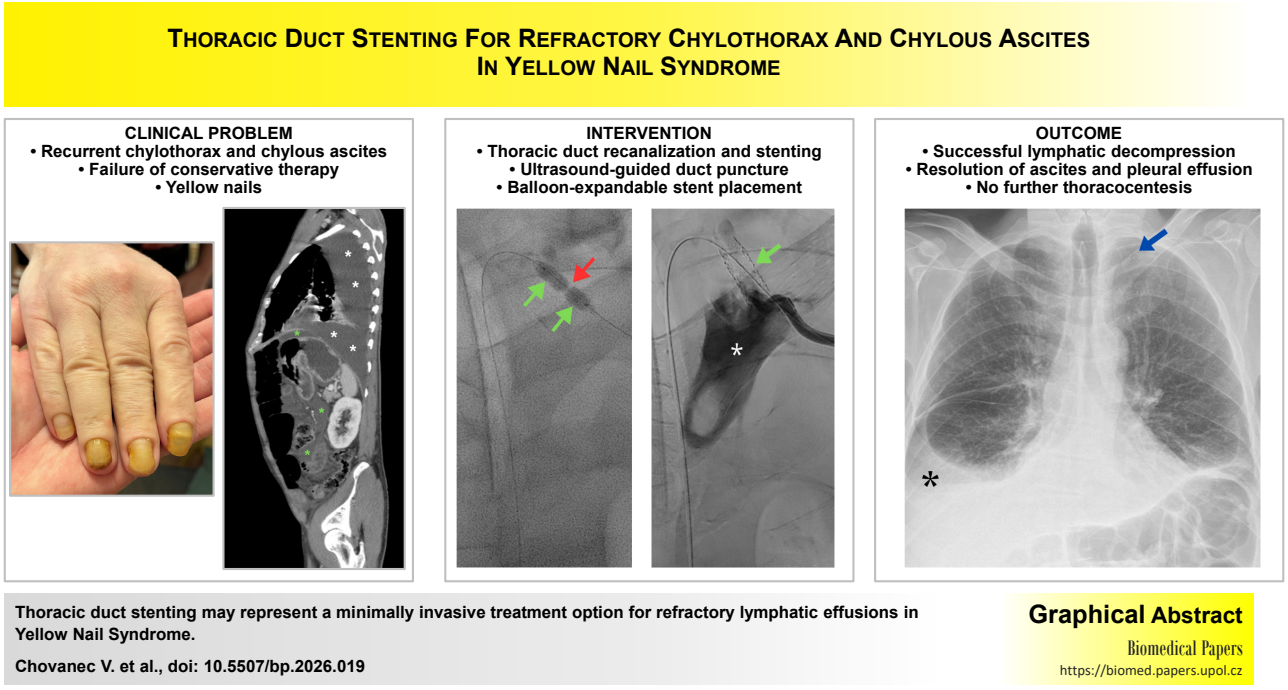
Purpose. To report the approach, technical success, safety, and short-term outcomes of thoracic duct stenting for treating chylothorax and chylous ascites in a patient with Yellow Nail Syndrome.
Case report. A 57-year-old male with Yellow Nail Syndrome, characterized by yellow thickened nails, lymphedema, and respiratory abnormalities, required repeated thoracocenteses for chylothorax. A thoracic duct intervention involving percutaneous transluminal angioplasty and balloon-expandable stent implantation via retrograde access was performed. Procedure characteristics, clinical success, complications, and follow-up were recorded.
Conclusions. Thoracic duct stenting was a safe and technically successful procedure. After the procedure, the patient was relieved of symptoms. This technique could be considered for patients with recurrent chylothorax and chylous ascites, including those with Yellow Nail Syndrome. Further research is required to validate these findings.



Key words: Yellow Nail Syndrome, ductus thoracicus stenting, chylous ascites, chylothorax, thoracic duct decompression

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BACKGROUND

Yellow Nail Syndrome (YNS) is a rare condition characterized by a triad of symptoms: yellow, thickened nails, lymphedema, and respiratory abnormalities, including pleural effusions and chronic bronchitis. The syndrome is believed to arise from lymphatic dysfunction, which can result in complications such as chylothorax and chylous ascites. Traditional management of these complications has relied on dietary interven-

tions, drainage procedures, and, very rarely in surgical approaches.

Yellow Nail Syndrome (YNS) was first described in humans by Peter Samman and William White in 1964 [1]. In their landmark paper, they documented a series of patients with:

1. **Yellow Nail Changes:** Thickened, yellow, slow-growing nails with onycholysis and nail plate discoloration ranging from pale yellow to dark greenish.
2. **Lymphedema:** Often bilateral, affecting the lower limbs, due to impaired lymphatic drainage.
3. **Respiratory Manifestations:** Chronic cough, pleural chylous effusions, bronchiectasis, and recurrent sinusitis[1-6].

YNS affects adults over 50 years old in most cases, though paediatric and familial forms have been reported. The estimated prevalence is less than 1 in 1,000,000. It may appear as an isolated condition or in association with autoimmune diseases, malignancies, or lymphatic abnormalities. The cause of YNS remains uncertain. The primary hypothesis suggests lymphatic dysfunction as the underlying mechanism. Lymphangiographic studies reveal abnormalities such as hypoplastic or dilated lymphatic vessels in patients with severe lymphedema. Diagnosis is clinical and relies on identifying at least two components of the triad. Nail changes are often the first sign, but the triad components may manifest sequentially, complicating diagnosis. Diagnostic imaging such as chest CT can confirm bronchiectasis, pleural effusions, while lymphoscintigraphy assesses lymphatic function[2]. Spontaneous resolution of nail changes in yellow nail syndrome occurs in up to 30% of cases but is less common for systemic symptoms. Prognosis depends on the severity of associated conditions such as malignancy or chronic respiratory disease[2].

The pathophysiology of chylothorax and chylous ascites involves impaired lymphatic drainage, which leads to the leakage of lymphatic fluid into the pleural or peritoneal space. This dysfunction may be caused by lymphatic obstruction due to malignancies, inflammatory diseases or structural congenital abnormalities of the thoracic duct[2-4].

Other, more common reasons for chylothorax include disruption of the thoracic duct due to traumatic or iatrogenic injury. Negative intrapleural pressure during respiration promotes the continuous drainage of chyle into the pleural space, leading to respiratory distress and nutritional deficiencies from the loss of fats, proteins, and lymphocytes[3-5].

CASE REPORT AND PROCEDURE CHARACTERISTICS

A 57-year-old man with congenital moderate intellectual disability and behavioral problems experienced episodic swelling of the lower limbs and scrotum of unknown origin for 10 months. He was admitted to another local hospital due to significant shortness of breath. Yellow discoloration of the nails on his hands and feet was observed (Fig. 1 and 2). A chest X-ray and ultrasound examination revealed significant bilateral pleural effusion and small ascites (Fig. 3). A therapeutic puncture of the pleural effusion and a diagnostic puncture of the ascites were performed, revealing bilateral chylothorax and lymphatic ascites. Conservative treatment, including chest drainage, total parenteral nutrition, and administration of a somatostatin derivative, did not reduce the chylothorax and ascites over the next 27 days. Malnutrition and mineral depletion gradually worsened. The patient was transferred

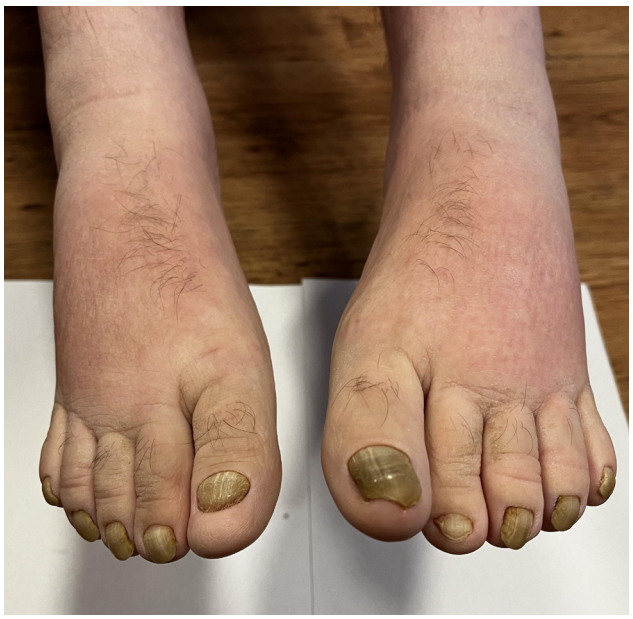

Fig. 1. Yellow thickened nail discoloration of the hands in a patient with Yellow Nail Syndrome prior to treatment.

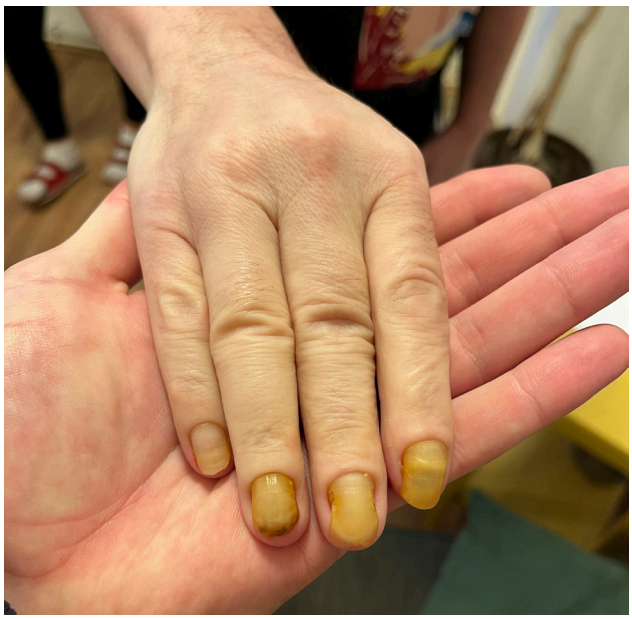

Fig. 2. Yellow thickened nail discoloration of the feet in a patient with Yellow Nail Syndrome prior to treatment.

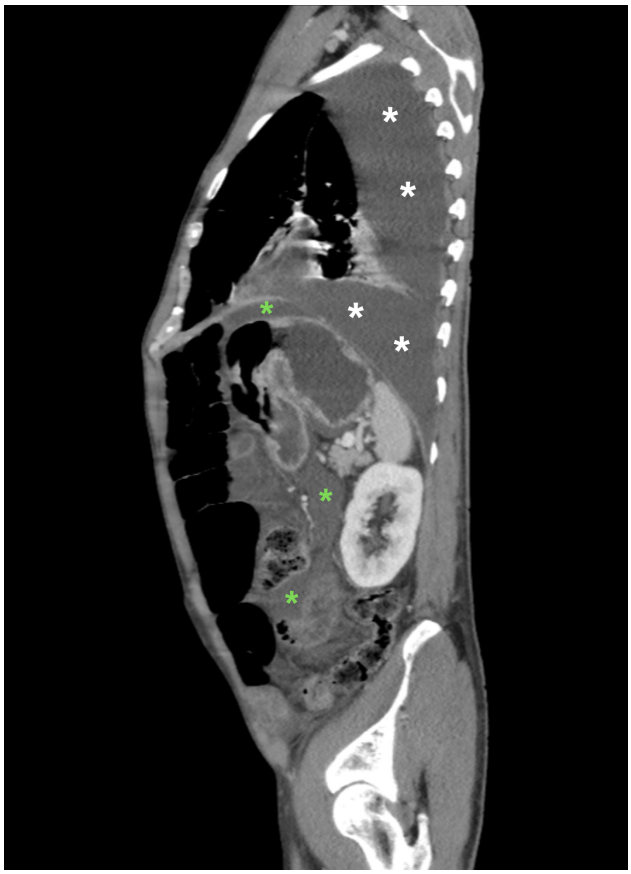

Fig. 3. Chest X-ray demonstrating significant bilateral pleural effusion and small ascites at initial presentation.

to a university hospital for intranodal lymphography with an oil contrast agent. Under ultrasound guidance, 22 G needles were inserted into the inguinal nodes bilaterally, and 22 mL of Lipiodol (Guerbet, Villepinte, France) was injected, in accordance with our institutional intranodal lymphangiography protocol. Lymphography showed slow filling of the lymph nodes and thoracic lymph duct, but no leakage into the pleural or peritoneal cavity (Fig. 4 A, B). The next day, a native CT scan of the trunk also showed no evidence of oil leakage outside the lymphatic system (Fig. 5). Subsequently, the chylothorax and ascites persisted, necessitating repeated thoracocentesis of 1000–1500 mL every 5–7 days.

The patient was re-admitted after 90 days for the therapeutic embolization of the thoracic duct. Due to the risk of intubation, transabdominal access to the cisterna chyli was not chosen; instead, retrograde transvenous access to the thoracic duct was attempted under intravenous sedation. The left brachial vein was punctured using a micropuncture set under ultrasound guidance, and a 6 F sheath was inserted. The attempt to catheterize the thoracic duct using RIM, RC catheters, and wires with V 10 control, Terumo, and Progreat 2.4 microcatheter with GT microguidewire was unsuccessful. The next step was to puncture the cervical segment of the thoracic duct under ultrasound guidance. An ultrasound examination with a linear probe in a sterile cover showed a tubular structure without flow detection in Doppler mode. A 21 G needle was then used to puncture this structure, located at the left venous angle, at the junction of the left internal jugular vein and the left subclavian vein. After applying iodine contrast media, it was confirmed that this was the thoracic duct. The V18 control wire was inserted through the opening of the lymphatic duct into the venous system due to failed retrograde catheterization of the tho-

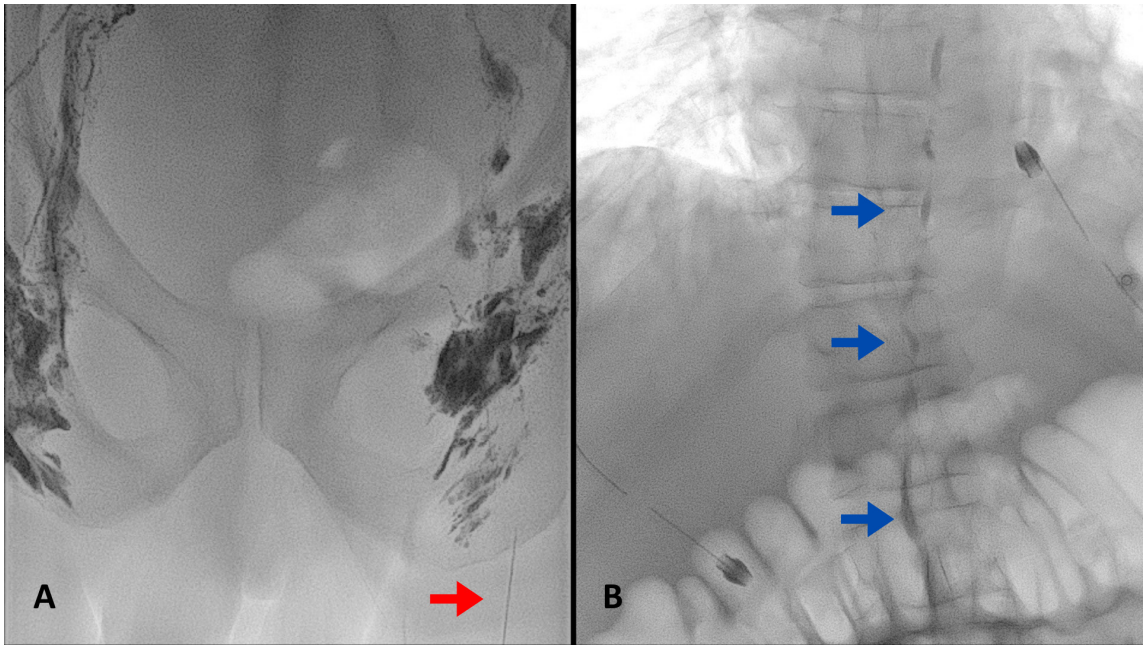

Fig. 4. Intranodal lymphangiography following bilateral inguinal node puncture. (A) Slow filling of lymph nodes and thoracic lymph duct with Lipiodol. (B) No evidence of contrast leakage into the pleural or peritoneal cavity.

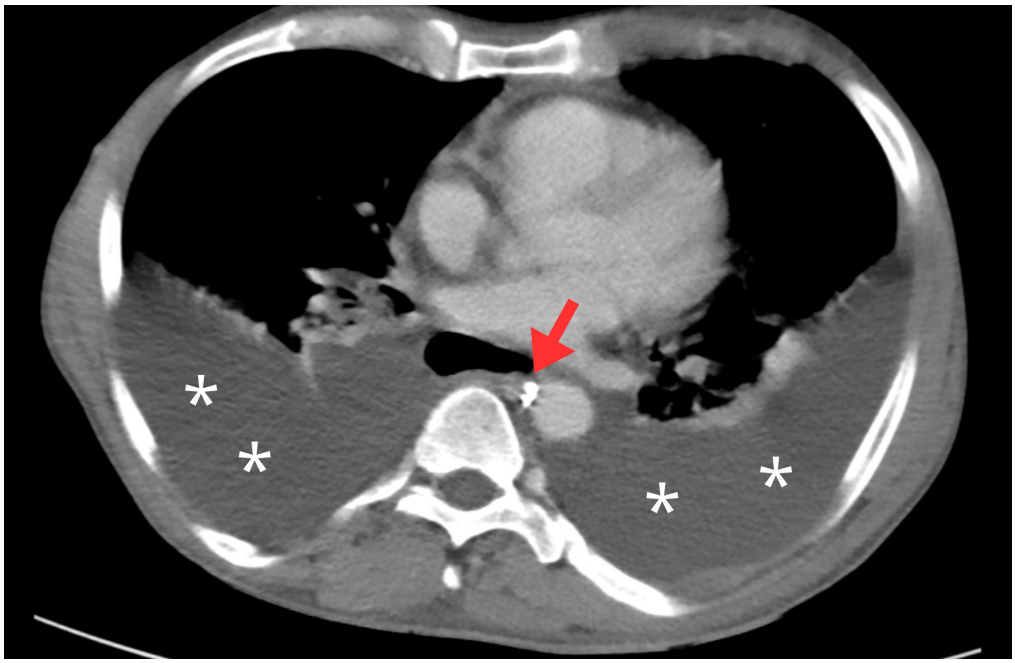

Fig. 5. Native CT scan of the trunk performed the day after lymphangiography showing no Lipiodol leakage outside the lymphatic system.

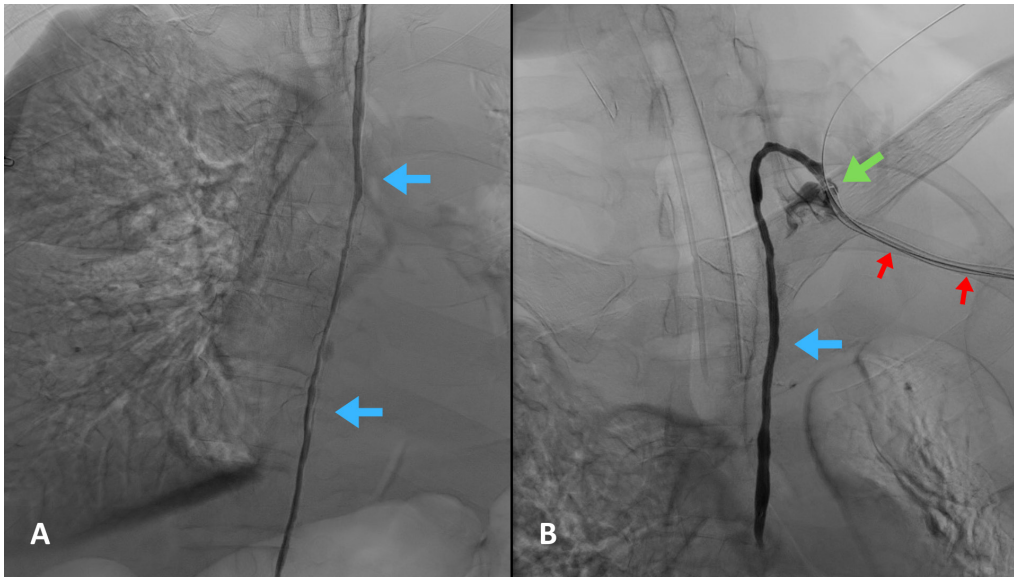

Fig. 6. Retrograde transvenous ductography via cervical puncture of the thoracic duct. **(A)** No evidence of contrast media extravasation, confirming absence of active leak. **(B)** Catheter position within the thoracic duct.

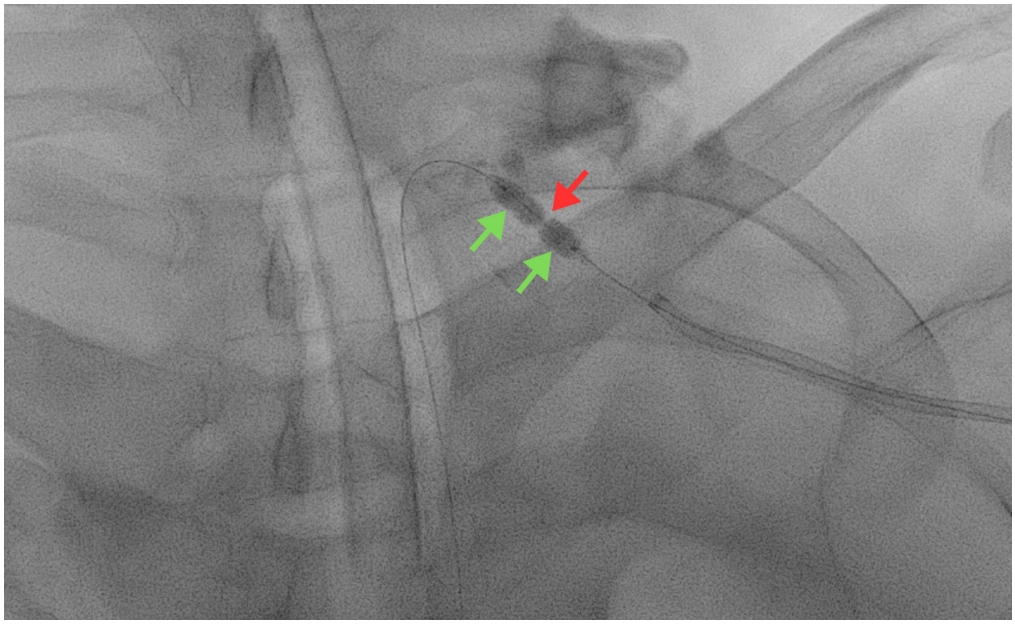

Fig. 7. Balloon dilation of the thoracic duct at its ostium showing a waist on the balloon, confirming ostial stenosis.

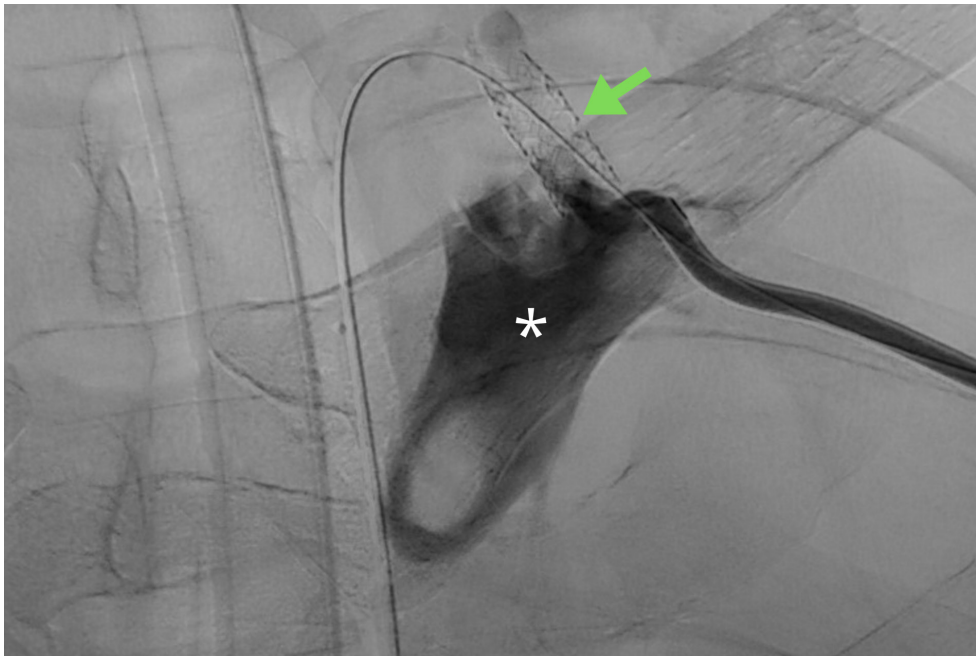

Fig. 8. Fluoroscopic image after deployment of a balloon-expandable stent into the ostial stenosis of the thoracic duct.

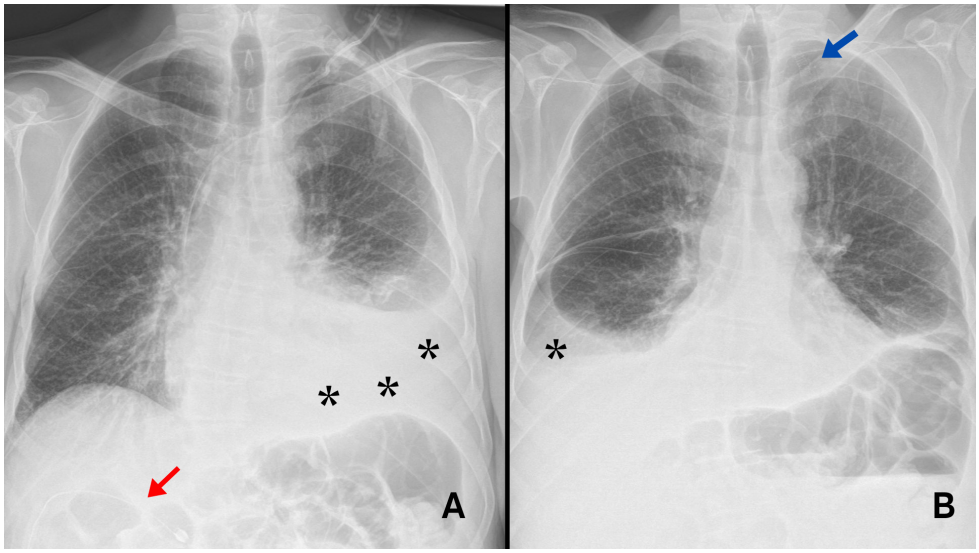

Fig. 9. Chest X-ray imaging prior the procedure **(A)** and at six months follow-up **(B).**

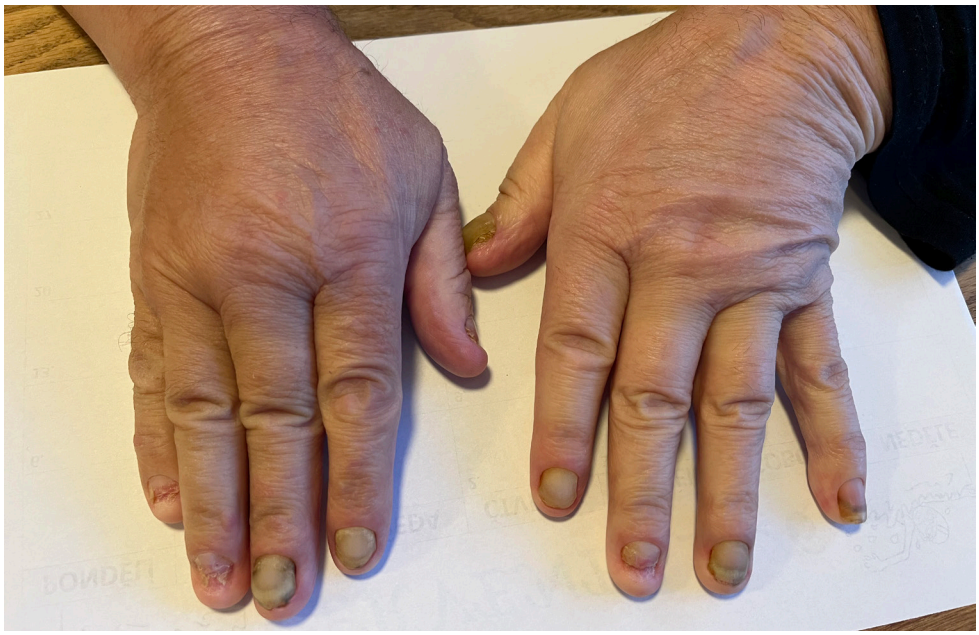

Fig. 10. Resolution of yellow nail discoloration of the hands.

racic duct. The guidewire in the venous system was captured and withdrawn into the sheath in the brachial vein. Subsequently, the DAV catheter was inserted through the thoracic duct. The thoracic duct was then catheterized with a coaxially inserted microcatheter. Ductography was performed with no evidence of contrast media extravasation, so its embolization was abandoned (Fig. 6 A,B). Due to the slow filling of the thoracic lymphatic duct during intranodal lymphography, dilation of the lymphatic duct into the subclavian vein was performed with a 5x40 mm balloon Mustang (Boston Scientific, Marlborough, USA), which was introduced over the V18 control guidewire, and a waist was seen on the balloon at the ostium of the thoracic duct, indicating ostial stenosis as the cause of lymphatic obstruction and the indication for stenting (Fig. 7). After percutaneous transluminal angioplasty (PTA) a 4.5x16 mm balloon-expandable stent Express Vascular LD (Boston Scientific, Marlborough, USA) was inserted into the ostial stenosis of the thoracic duct (Figure 8). The patient was subsequently discharged 4 days later.

The patient is currently being monitored six months after stent placement. During this time, there was a gradual absorption of pleural effusion on the left and ascites (Fig. 9A,B). No further thoracocentesis or paracentesis was required during the six-month follow-up period. A small fluidothorax on the right persisted on the chest radiograph but was regressing. Nutritional status has normalized. Figure 10 shows resolution of yellow nail discoloration.

LITERATURE REVIEW AND DISCUSSION

There are currently no established guidelines for managing Yellow Nail Syndrome, and no specific treatment exists. Symptom management is prioritized and tailored to the individual manifestations experienced by each person[2,5]. A conservative approach is preferred, including drainage of effusions and nutritional and pharmacological adjustments, before considering more invasive options. Conservative therapy to Yellow nail syndrome is summarized in Table 1.

Surgical treatment

When conservative approaches to managing chylothorax are ineffective, surgical options may be considered. The most commonly described surgical technique is thoracic duct ligation. For refractory chylothoraxes, pleuroperitoneal shunts (PPS) and pleuro-venous shunts (PVS) are options worth considering. A PPS redirects chylous fluid into the peritoneal cavity for systemic absorption; however, it is important to avoid using it in individuals who also have chylous ascites, which our patient did. A PVS, on the other hand, directs chyle towards the subclavian or jugular veins. PPS and PVS can be placed under the skin or externally, and they can be activated manually. Although shunts are less invasive compared to video-assisted thoracoscopic surgery (VATS) for thoracic duct ligation, they carry a significant risk of complications such as displacement, skin erosion, infection, blockage, and pneumoperitoneum[20,21]. These methods were unsuitable for our patient due to the presence of chylous ascites and the inability to manually activate the PPS or PVS because of intellectual disability.

Thoracic duct lymphovenous anastomosis (TD-LVA) is a new specialized microsurgical technique used for cen-

Table 1. Summary of conservative treatment approaches to Yellow nail syndrome.

Manifestation	Treatment	Details	Ref.
Nail Changes	Vitamin E (oral/topical)	1,000–1,200 IU/day; more effective when combined with fluconazole	2,6
	Itraconazole + Clarithromycin	Stimulate nail growth and reduce discoloration	7,8
Lymphedema	Complete Decongestive Therapy	Compression bandaging, manual lymphatic drainage, exercises, skin care	
	Elastic Compression Garments	Used for long-term limb volume maintenance	2
Respiratory Issues	**Pleural Effusions**		
	Thoracentesis	For initial symptomatic relief	9
	Pleurodesis, Surgery	Talc pleurodesis, decortication, pleural-peritoneal shunt for recurrent effusions	10–13
	Prednisolone	Used in some cases to control pleural effusion	14
	Bronchiectasis		
	Antibiotics & Vaccinations	Azithromycin; influenza and pneumococcal vaccines	9
Chylous Effusions	Dietary Modifications	Low-fat diet rich in medium-chain triglycerides	15,16
	Octreotide	Somatostatin analog that can reduce chylous effusions	17–19

tral lymphatic reconstruction. This procedure involves creating a new connection between the thoracic duct and a nearby vein. TD-LVA can be performed at various anatomical levels depending on the underlying condition, most commonly connecting to the internal jugular or subclavian vein in the neck, the azygos vein in the thorax, and the ovarian or phrenic vein in the abdomen. Treatment success and long-term outcome remains unclear[22,23].

Thoracic duct embolization

Thoracic duct embolization (TDE) is a non-surgical alternative to surgical methods, aimed at closing the chyle leak at its source. The first report of percutaneous treatment for chyle leaks using embolization was published by Cope in 1998 (ref.[24]). This method has since been adopted by many authors[25]. The procedure begins with pedal or intra-nodal lymphangiography to visualize the cisterna chyli or other enlarged lymphatic vessels. A catheter is then inserted into the thoracic duct, and thoracic ductography is performed to locate the leak. Once the leak is confirmed, embolization coils are deployed to repair the defect. The success rate for TDE is estimated at 60%, with non-traumatic cases showing an improved success rate of nearly 74% (ref.[20,26-28]). This intervention is not recommended for patients with untreated bleeding disorders or abdominal abnormalities, such as an abdominal aortic aneurysm, as accessing the retroperitoneum may lead to serious complications. Documented complications include fat embolism, bile duct leakage, persistent swelling in the legs or abdomen, and chronic diarrhea. A systematic review and meta-analysis by Kim et al., reports an overall complication rate for TDE of 2.4% (ref.[25]). TDE provides a safe, minimally invasive alternative to surgery for managing chylothorax, with a moderate success rate. However, we believe that TDE could worsen our patient's clinical status, as thoracic duct occlusion may increase chylothorax production. When stenosis of the ductus thoracicus is present, stenting has been shown to be the appropriate intervention. In cases where ductography reveals no active extravasation but slow lymphatic flow or mechanical obstruction is identified, thoracic duct stenting represents a rational alternative to embolization. Embolization in the absence of a demonstrable leak may worsen lymphatic hypertension and paradoxically increase chylous output. In contrast, stenting aims to relieve the underlying obstruction and restore physiological lymphatic drainage without occluding the duct.

Thoracic duct stenting

Thoracic duct stenting involves the placement of a stent to relieve obstruction or compression within the thoracic duct. The first reported cases of thoracic duct stenting were reported in 2016. In a case series, thoracic duct reconstruction using a stent-graft was successfully performed in two patients with chylothorax. Both procedures were technically and clinically successful without procedure-related complications[29].

Several subsequent reports and small case series have explored a thoracic duct stenting as a novel intervention for various conditions, including chylothorax, refractory ascites or chyluria[29-32]. To date, no randomized control trials or meta-analyses have been published regarding thoracic duct stenting. This is likely due to the rarity of the procedure and limited number of reported cases. Most of the available data are confined to isolated case reports or very small series. Based on the available literature, it can be hypothesized that at least 13 patients have undergone thoracic duct stenting, considering the nine patients from the 2022 pilot study on refractory ascites treatment, the four patients from the case reports[30,32]. However, this number is likely underestimated, as additional cases may exist in other unpublished studies or reports. The exact total remains uncertain due to the limited number of published cases

Although thoracic duct stenting has been successfully employed for other causes of chylothorax and chylous ascites, thoracic duct stenting in a patient with yellow nail syndrome has not been published yet. We believe this is the first case we are reporting. We summarized key finding of this study in the Table 2.

CONCLUSION

Thoracic duct stenting represents a novel and promising intervention for managing chylothorax and chylous ascites in patients with Yellow Nail Syndrome. While early results of our case report are encouraging, continued in-

Table 2. Summary of key findings regarding thoracic duct stenting.

Category	Details
Outcomes	**Chylothorax Resolution:** Effective in reducing pleural effusions and improving respiratory symptoms in YNS-related cases. **Chylous Ascites Management**: Stenting helped redirect lymphatic flow, resolving fluid accumulation in the abdomen.
Advantages	**Minimally Invasive:** Less invasive than surgical options, with quicker recovery. **Symptomatic Relief:** Durable improvement in symptoms by treating lymphatic dysfunction. **Reduced Procedure Frequency:** Fewer needs for thoracentesis or paracentesis post-stenting.
Limitations and Challenges	**Technical Expertise:** Required specialized skills and equipment. **Complications:** Risks include infection, stent migration, and lymphatic leaks. **Limited Evidence:** Rarity of YNS restricts large-scale study data.

vestigation and clinical experience are crucial to refine this approach and expand its application in this rare and challenging condition. Further research is needed to establish standardized protocols for thoracic duct stenting. Comparative studies are essential to evaluate its efficacy relative to other treatment modalities. Advances in imaging techniques and stent materials may enhance procedural safety and outcomes.

ABBREVIATIONS

YNS, Yellow nail syndrome; PTA, Percutaneous transluminal angioplasty; IU, International units; PPS, Pleuroperitoneal shunts; PVS, Pleuro-venous shunts; TDE, Thoracic duct embolization; TD-LVA, Thoracic duct lymphovenous anastomosis.

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Author contributions: CH performed thoracic duct stenting. PV analysed and interpreted the patient's data regarding interventions. PR, JR, FC, IH provided concept and design of the study. PR, JR, FC, IH critically revised and substantially edited the manuscript. All authors read and approved the final manuscript.

Conflict of interest statement: The authors declare that they have no competing interests.

Ethics approval and consent to participate: Written informed consent was obtained. All methods were carried out in accordance with institutional guidelines and regulations. Patient (legal guardian) have provided acceptance and consent for publishing their data. All personal information has been made anonymous.

Availability of data and material: The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Declaration of generative AI and AI-assisted technologies: No AI tools were used in the preparation, writing, or analysis of this manuscript. All content, data, and conclusions are the sole work of the authors.

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