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Thrombectomy in Lemierre's Syndrome: A Systematic Literature Review

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Abstract

Introduction:

Lemierre's syndrome (LS) is a rare but potentially life-threatening condition characterized by septic thrombophlebitis of the internal jugular vein, typically following an oropharyngeal infection. First described in 1936, the syndrome was once considered a "forgotten disease" due to its rarity in the post-antibiotic era. However, recent decades have witnessed a modest resurgence in reported cases, attributed in part to increased clinical awareness, improved diagnostic techniques, and evolving microbial resistance patterns. Antibiotics and anticoagulation remain mainstay of treatment with a high rate of treatment response. However, there is a small subset of patients that fail standard treatment and instead clinically deteriorate necessitating prolonged antibiotic therapy. This cohort of patients typically has unexpected critical care unit admissions and prolonged hospital stays. Historically, these patients would undergo vessel ligation. Thrombectomy has been proposed as an alternative to surgical vessel ligation for treatment resistant LS.

Materials and Methods:

Using PubMed/MEDLINE and Scopus databases, we performed a comprehensive literature search on April 16, 2025, to identify studies relating to thrombectomy in Lemierre's Syndrome.

Results:

Four articles were found that fit inclusion criteria. Although limited by quantity, the studies found demonstrated that patients with treatment resistant disease usually had some clinical improvement after thrombectomy and on average had shorter intensive care unit and hospital stays.

Conclusion:

Thrombectomy use for treatment resistant LS is not well described in the literature. However, while not ideal or appropriate for every patient, there could exist a potential role for thrombectomy in patient with LS symptomology resistant to antibiotics and anticoagulation.

***Keywords*:** Lemierre's Syndrome; Lemierre's Disease; Thrombectomy; Septic thrombophlebitis; Percutaneous mechanical thrombectomy; Thrombolytic therapies.

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Introduction

Lemierre's Syndrome (LS) is a rare disease characterized by thrombophlebitis of the head and neck's vasculature, usually following an infection of the oropharynx (1,2). Sources of infection include most commonly the tonsils and other palatine/peritonsillar tissues; however, the middle ear, teeth, sinuses, and lungs have also been implicated as primary sources in the literature (1-3).

Lemierre's disease was once a devastating disease with mortality rates reported as high as 90%. The incidence is estimated to range from 0.8-3.6 persons per 1 million patients (1,4). Antibiotic therapy is the mainstay of treatment. Systematic anticoagulation therapy remains controversial but is commonly used in treatment depending on clot burden. With the advent of antibiotics, mortality has decreased from a rate as high as 90% to 0-18% (1,3,5,6).

With the usage of antibiotics and anticoagulation, the rates of surgical ligation and other invasive interventions have decreased. LS is most associated with *Fusobacterium necrophorum*, a Gram-negative anaerobe, although a wide spectrum of pathogens has been implicated. Clinical presentation often includes sore throat, fever, neck swelling, and signs of septicemia, frequently complicated by pulmonary emboli and metastatic infections. Given its nonspecific early symptoms and the rapid progression to systemic involvement, prompt diagnosis and aggressive management are critical.

Although prognosis has improved and most cases resolve with antibiotic and anticoagulation treatment there is a small subset of patients that have persistent symptoms of disease despite standard treatment.

These symptoms can range from asymptomatic pulmonary embolism (PE) to symptomatic PEs leading to hypoxia and respiratory compromise. Other embolic phenomena such as transient ischemic attacks (TIAs) and strokes can lead to unplanned ICU stays, extended hospital stays, and increased duration of antibiotic and anticoagulation therapies.

Select case reports have also demonstrated increased clot burden in the internal jugular (IJ) vein leading to airway edema, tracheal compression, and total airway compromise requiring emergency and sometimes prolonged intubation. In these select cases, thrombectomy may represent a viable, minimally invasive option for symptomatic improvement before major surgical intervention. This systematic review aims to synthesize the existing literature on Lemierre's syndrome, with the goal of determining the utility of thrombectomy for improving patient outcomes. We hypothesize that thrombectomy would improve symptomology of LS when disease and clot burden is high and causes unexpected clinical complications.

An intuitional review board (IRB) approval and or exemption was not sought as this study as it analyzed previously published reports. Likewise, ethical approval was not sought as this investigation did not involve any direct patient identifiers as patients were already deidentified and previous reports were HIPAA compliant.

Materials and Methods

Search Strategy and Information Sources

A concise review was conducted using a systematic approach. We performed a comprehensive literature search on April 16, 2025, to identify studies relating to thrombectomy in Lemierre's Syndrome.

The search was conducted using the PubMed/ MEDLINE and Scopus databases for all studies which was then uploaded to Covidence for sorting and review by authors.

The search strategy included a combination of Medical Subject Headings (MeSH), keywords/ concepts and free-text terms including Lemierre's Syndrome, septic thrombophlebitis, suppurative thrombophlebitis, thrombectomy, thrombectomy, thrombophlebitis, thrombolytic therapy, and fibrinolytic agents. Additionally, reference lists of included studies and relevant reviews were screened to identify additional articles.

Eligibility Criteria

Inclusion criteria were as follows: (1) children and adults with confirmed Lemierre's syndrome. (2) thrombectomy or thrombolytics were used as adjuncts to treatment with antibiotics; (3) no limit on year of publication and (4) no limit on language of publication. Exclusion criteria were as follows: (1) no full-text available, and (2) antibiotic monotherapy.

Selection Process

Six authors independently screened the titles and abstracts of all retrieved articles for relevance. Full texts of potentially eligible

studies were obtained and reviewed against the inclusion and exclusion criteria.

Discrepancies in study selection were resolved through discussions among authors with majority voting, and the principal investigating author having the deciding vote if consensus was not reached.

Data Items and Collection Process

A standardized data extraction form was used to collect information from each included study. Extracted data included study characteristics, thrombectomy technique, clinical course, and patient outcome.

Results

Our search returned a total of 761 studies on SCOPUS and PubMed/MEDLINE databases. After initial abstract screening, only 13 articles met full inclusion criteria for full text review. After full text review, 4 articles met the proper criterion and were included in this review. An overview of this process is represented in the PRISMA study flowchart found in (Figure 1 and Table 1).

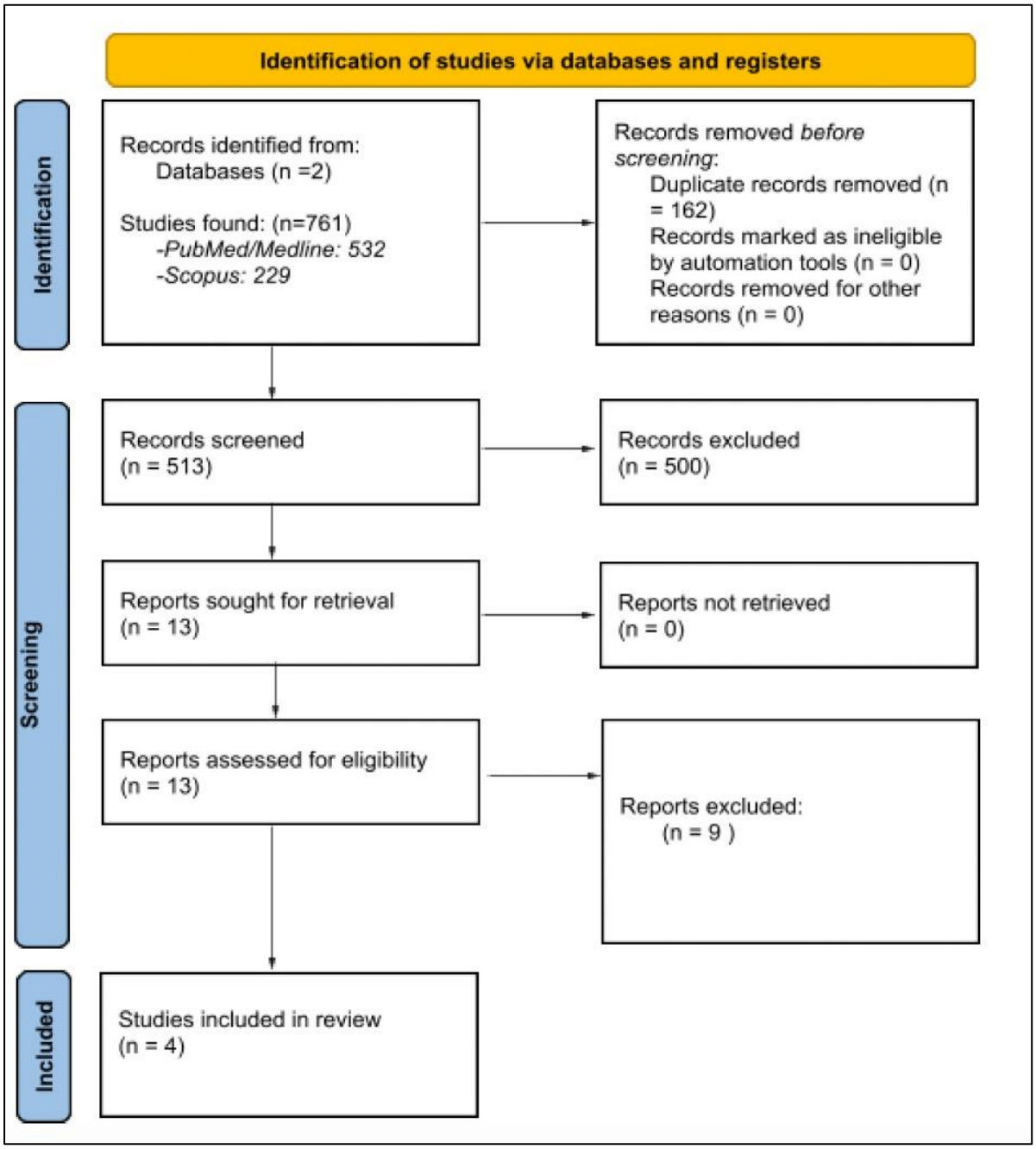

Fig 1. PRISMA flow chart for search criteria

Table 1. Study characteristics and outcome

Case	Patient Demographics	Thrombectomy Type	Outcome
Cuddy et al.	25yo M - lower lip and chin swelling. - CT demonstrated a left common facial vein thrombus	Open thrombectomy	- Clinical course improved, and he eventually extubated and weaned off pressors. - Discharged on day 20 of hospitalization.
B.K.A et al.	37yo M - 2-week history of o worsening headaches, limited neck range of motion o fevers o upper extremity weakness o inability to swallow o inability to void - CT angiography: o abscesses of the epidural, paravertebral and retropharyngeal spaces o occlusive thrombus in the left sigmoid sinus associated with left IJV extension.	IJV mechanical thrombectomy with microcatheter placement with administration of tissue plasminogen activator (tPA).	- Following thrombectomy, the patient was restarted on anticoagulation and taken back for another hematoma evacuation. - Hospital course was complicated by complications from the cervical abscess and subsequent hematomas.
Min et al.	52yo W - 8-day duration of left painful neck swelling - left arm pain - CT scan revealed a thrombus in the left IJV, subclavian vein and brachiocephalic vein.	Percutaneous aspiration thrombectomy	- Failed to remove the clot due to tight adherence to the vein and only a small amount was aspirated. - Antibiotic treatment was maintained for 6 weeks (2 weeks IV, 4 weeks of oral) - Anticoagulation was maintained for 3 months.
Davis et al.	55yo W - neck pain - retropharyngeal abscess - bilateral IJV thromboses demonstrated on CT scan	Percutaneous aspiration thrombectomy	- Extubation on day 8. - Downgraded from critical care management on day 10.

Cuddy et al. described a case wherein a 25-year-old male presented with an atypical presentation of Lemierre disease treated with debridement and open thrombectomy. The patent initially presented with lower lip and chin swelling. Contrast-enhanced computed tomography (CT) scan of the face demonstrated a left common facial vein thrombus (7). He was initially treated with intravenous (IV) antibiotics but clinically decompensated. His clinical course was complicated by persistent tachycardia, septic emboli to the lung in addition to chin/lip edema that rapidly progressed to a necrotizing infection. He was taken to the operating room for debridement after which his symptoms did not improve, and he became septic with hypoxic respiratory failure requiring mechanical ventilation and vasopressor support. Eventually, despite no previously established role, he was taken for open thrombectomy after which his clinical course improved, and he was later discharged on day 20 of hospitalization.

B.K.A et al. reported a case of a 37-year-old man who presented with a 2-week history of worsening headaches, limited neck range of motion, fevers, upper extremity weakness, inability to swallow and an inability to void (8). CT angiography demonstrated abscesses of the epidural, paravertebral and retropharyngeal spaces in addition to a large occlusive thrombus in the left sigmoid sinus associated with left IJV extension. Treatment began with antibiotics

and systemic anticoagulation however given significant neurologic decline, systemic anticoagulation was discontinued, and the patient was taken to the operating room for emergent exploration by neurosurgery. Blood cultures and deep neck operative cultures both grew S. intermedius. This finding plus the presence of the IJV thrombus led to the diagnosis of Lemierre's syndrome. Repeat neck exploration was performed in the setting of a large posterior, cervical epidural hematoma which caused altered mental status and new onset bilateral lower extremity weakness.

After hematoma evacuation, a repeat CT venogram demonstrated multiple sites of intracerebral hemorrhage and thrombus propagation from the left sigmoid sinus and left IJV to the transverse sinus. Given his recent epidural hematoma evacuation, systemic anticoagulation became contraindicated, and the patient underwent emergent diagnostic cerebral angiogram with mechanical thrombectomy of the left sigmoid sinus, the left transverse sinus, and the left IJV (8).

Following thrombectomy, the patient was restarted on anticoagulation and was then taken back for another hematoma evacuation. Although the overall hospital course was complicated by complications from the cervical abscess and subsequent hematomas, thrombectomy provided moderate symptom relief.

Min et al. described a 52-year-old woman who presented with an 8-day duration of left painful neck swelling and left arm pain (9). A neck CT scan revealed a thrombus in the left IJV, subclavian vein and brachiocephalic vein which help establish the diagnosis of Lemierre syndrome. Percutaneous aspiration thrombectomy failed to remove the clot due to tight adherence to the vein and only a small amount was aspirated. Systemic antibiotic and anticoagulation therapies were initiated. Antibiotic treatment was maintained for 6 weeks (2 weeks IV therapy and 4 weeks of oral therapy) while anticoagulation was maintained for 3 months before full recanalization of the thrombosed vein was visualized on repeat imaging.

Davis et al. described a 55-year-old woman who presented with neck pain and was found to have a retropharyngeal abscess associated with bilateral IJV thromboses demonstrated on CT scan (2). The hospital course was complicated by fever, tachycardia, sepsis, and patchy opacities in all lung fields. Other complications included acute stroke associated with mental status changes, dysphonia, and facial droop due to embolic phenomena. She also developed airway edema, stridor, and increased work of breathing leading to hypercapnic respiratory failure. She was initially trialed on high flow nasal cannula followed by bilevel positive airway pressure (BiPAP) but subsequently required intubation/mechanical ventilation due to worsening respiratory function. Despite continuous IV antibiotics and anticoagulation, the patient continued to clinically deteriorate. Repeat imaging demonstrated airway compression caused by increased clot burden, venous congestion and edema, and a secondary concern for worsening septic emboli to her lungs. At this point, the decision was made to proceed with thrombectomy. The patient underwent thrombectomy on hospital day 6 wherein a significant amount of clots were aspirated. Following thrombectomy, the patient had a rapid recovery with immediate improvement in disease burden and eventual extubation on day 8 and step down from critical care management on day 10.

Discussion

In Lemierre's syndrome, thrombosis can lead to a variety of complications including but not limited to septic emboli resulting in PEs and strokes, airway compression, edema, and eventual respiratory collapse if clot burden is large and located in the IJV (2).

While antibiotic and anticoagulation treatment usually lead to speedy recovery in a majority of LS patients, there is a subset of the population wherein symptoms persist and lead to significant clinical deterioration. Historically, this subset of the population was treated with surgical ligation of the vessel for resolution of lingering symptoms (1,2,5,6,8,10).

Given medical advances, mechanical thrombectomy may present a viable, less invasive treatment pathway for these treatment resistant patients.

A role for thrombectomy in Lemierre's syndrome is not well established in the current literature. Use of thrombectomy has only been described in a handful of case reports; currently, use of thrombectomy is dependent on

each patient's individual clinical scenario and multidisciplinary discussions among clinicians. In the case reports discussed above, patients tended to be slow to respond to standard treatment or had significant clinical deterioration despite treatment leading to further inventions such as percutaneous thrombectomy. Most of these patients had rapid improvement following thrombus aspiration. Davis et al. found that after aspiration thrombectomy, their patient was able to be extubated with immediate symptom relief and downgraded from the critical care unit in less than 48 hours (2). B.KA et al. also reported use of thrombectomy in a patient with recurrent posterior neck hematoma wherein anticoagulation was contraindicated; this patient also had moderate symptom relief following thrombectomy (8).

While beneficial based on these selected case reports, thrombectomy is not without its limitations. Min et al. attempted a percutaneous aspiration thrombectomy but only aspirated a small number of clots due to a tightly adhered thrombus (9).

This case report demonstrated that thrombectomy might not be useful in certain patients depending on clot morphology, vessel adherence pattern, and location. This patient ended up being maintained on prolonged IV antibiotics and 3 months of anticoagulation therapy with a protracted hospital stay. This highlights that thrombectomy can fail and is not successful in every case. However, when compared to the successful thrombectomy cases, this example does highlight that thrombectomy can possibly shorten treatment duration and hospital stay. This reinforces that percutaneous thrombectomy might provide some therapeutic benefits in treatment resistant Lemierre's syndrome. Of course, given the small sample size, it is unknown whether this observational finding can be applied to the general populace.

Previously, these resistant LS patients would be treated with vascular inventions such as vessel ligation as a last resort. Thrombectomy, when compared to vessel ligation is minimally invasive and carries less cardiovascular risk in these critically ill patients however there is still some associated risk. Common risks associated with thrombectomy include bleeding, infection, recurrence of the clot, failure to retrieve or lyse the clot and damage to vascular structures. Mechanical thrombectomy carries the risk of cerebral stroke and vessel dissection while surgical thrombectomy carry the risk of increased bleeding and damage to surrounding organs and structures. Catheter directed thrombectomy carry an increased risk of allergic reactions to the thrombolytic agent injected. Common indications include myocardial infraction, stroke, pulmonary embolus, and complete vessel occlusion. Contraindications include intracranial hemorrhage, uncorrected coagulopathies, uncontrolled hypertension, small vessel occlusion, and a large infarct core with minimal penumbra (11).

Limitations of this study include language bias as we included only studies written in English or that have an English translation readily available which limited the obtained literature and may reflect an incomplete summary of what is truly known on thrombectomy used in LS. Inherent in the methodology of this study design is a publication bias as the tendency is for positive data to be reported and published. The publications that were found after an extensive search were all case reports which limits generalizability as these findings may not be representative of other patients. We also have an inherent selection bias as case reports that are published usually represents some rare phenomena and exist as novel narrative observations which make it impossible to rule out confounding variables.

Conclusion

In conclusion, Lemierre's syndrome tends to respond well to antibiotic and anticoagulation treatment. However, in specific cases such as clinical deterioration or increased disease burden, thrombectomy might yield some therapeutic benefit.

In this review of thrombectomy in LS, we found that patients with treatment resistant disease usually had rapid improvement after thrombectomy and on average had shorter hospital stays. While not ideal or appropriate for every patient, this finding elucidates a possible role for thrombectomy in this unique subset of patients. Unfortunately, due to the rarity of disease and its high response to antibiotics and anticoagulation, it is unlikely that a large meta-analysis could be performed

and be adequately powered to definitively prove or disprove this observational finding from this small sample of case reports.

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