Case Report

OPEN ACCESS

Haemoptysis as a presentation of an infected aortic aneurysm rupture

Elisa Landín-Rey[1], Maria Elena Toubes-Navarro[1], Miguel Domínguez-Robla[2], María Rey-Bascuas[4], Luis Valdés-Cuadrado[1,3,5]

[1]Pneumology Department, Hospital Clínico Universitario de Santiago de Compostela, Spain; [2]Radiology Department, Hospital Clínico Universitario de Santiago de Compostela, Spain; [3]Interdisciplinary Research Group in Pneumology, Instituto de Investigaciones Sanitarias de Santiago (IDIS). Santiago de Compostela, Spain; [4]Department of Angiology and Vascular Surgery, Hospital Clínico Universitario de Santiago de Compostela, Spain; [5]Department of Medicine, Faculty of Medicine, University of Santiago de Compostela, Spain

ABSTRACT

Introduction: Infective thoracic aortic aneurysms are uncommon, especially presenting with haemoptysis.
Case presentation: We report the case of an 81-year-old male who presented with fever and pleuritic chest pain and was initially misdiagnosed with community-acquired pneumonia. A CT scan later revealed a saccular, ruptured thoracic aortic aneurysm. Despite antibiotic therapy, the patient developed haemoptysis, necessitating thoracic endovascular aortic repair (TEVAR). Post-procedure, the patient showed significant clinical improvement and was discharged in stable condition 45 days later.
Conclusions: Infected thoracic aortic aneurysms presenting as haemoptysis are exceptionally rare but life-threatening. Early clinical suspicion (manifested by haemoptysis, fever and thoracic pain) is essential, particularly in patients with risk factors such as immunosuppression or previous infections. This case emphasizes the importance of prompt diagnosis and intervention, along with the use of appropriate imaging techniques to reduce morbidity and mortality associated with this rare yet severe condition.

Key words: Haemoptysis, infected aneurysm, aortobronchial fistula, thoracic endovascular aortic repair (TEVAR).

Introduction

The term "mycotic aortic aneurysm" originally referred to aneurysms associated with bacterial endocarditis [1]. However, it has been progressively replaced by "infected aneurysm" since a pathogen, usually a bacterium, induces the degradation of the aortic wall, leading to the aneurysm development. Thoracic aortic aneurysms account for 1.8% of all aneurysms in the thoracic-abdominal aorta [2-4]. Haemoptysis is a rare presentation generally occurring when an aortobronchial fistula is present, often draining into the left bronchial tree and potentially causing fatal asphyxiation. The predominant pathogenic mechanism involves bacterial inoculation from a primary infection site –identifiable in 86% of cases- affecting a previously compromised artery and likely influenced by prior immunosuppression [4]. In this article, we present a case of infected aneurysm with haemoptysis and perform a systematic review of the literature.

Correspondence: Elisa Landín-Rey. Pneumology Service, Hospital Clínico-Universitario de Santiago. Rúa da Choupana, s/n, 15706 Santiago de Compostela, A Coruña, Spain. elisa.landin.rey.@sergas.es

Authors' contributions: All authors have contributed equally to this manuscript. All authors read and approved the final version of the manuscript and agreed to be accountable for all aspects of the work.

Ethics approval and consent to participate: No ethical committee approval was required for this case report by the Department, because this article does not contain any studies with human participants or animals. Written informed consent was obtained from the patient who was enrolled in accordance with the Helsinki Declaration.

Conflict of interest: Authors declare no conflict of interest.

Case report

An 81-year-old man presented to the emergency department with fever. He had a history of two previous hospital admissions: one four years prior for heart failure with acute pulmonary edema and respiratory acidosis and another the following year for native aortic valve endocarditis secondary to *Enterococcus faecalis*. A diagnosis of chronic lymphocytic leukemia had been previously established. Five days before this event, he had been experiencing sharp, continuous, left sided chest pain aggravated by deep breathing and coughing, accompanied by rigors. On examination, fever -38.5ºC-, a panfocal systolic murmur and crackles at the base of the left hemithorax were noted. A chest x-ray, performed at the Emergency Department, revealed a homogeneous left retrohilar pulmonary consolidation that reached the posterior wall and crossed the fissure (Figure 1A). Blood cultures showed methicillin-sensitive *Staphylococcus aureus* positivity.

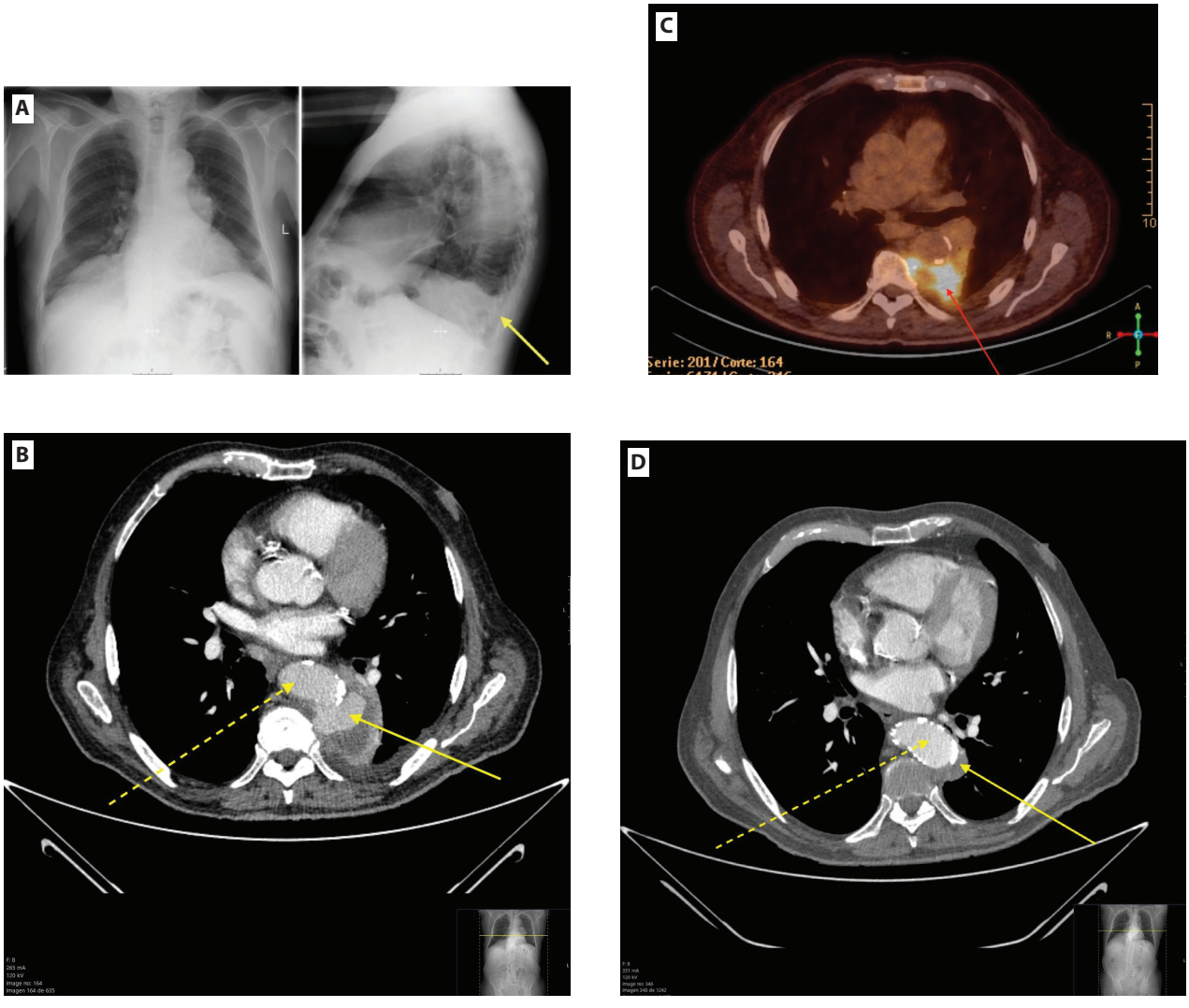

Figure 1. A) Posteroanterior (left) and lateral (right) chest X-ray upon admission showed retrohilar opacity erasing the aortic contour (arrow); B) CT scan of the chest (mediastinal window) demonstrates a posterior descending aortic aneurysm with contained rupture (solid arrow) and 'draped aorta' sign (dashed arrow). Peripheral thrombotic-necrotic material (without enhancement) is noted within the aneurysm sac; C) PET-CT image of the chest showing an hypermetabolic focus around the aortic aneurysm is observed (arrow). D) CT scan of the chest (mediastinal window) showing the aortic stent delineating the normal aortic lumen (dashed arrow). The excluded aneurysm has retracted and is now completely thrombosed and of diminished size (solid arrow).

He was initially diagnosed with community-acquired pneumonia and treated with intravenous levofloxacin - 500 mg every 12 hours for 2 days followed by 500mg every 24 hours for 10 days. Upon receiving the blood culture results, cloxacillin -2 g every 4 hours for 10 days- was added. As clinical progress was suboptimal, with persistent fever and thoracic pain, a thorax CT was performed on the 4th day of admission. CT revealed a ruptured saccular aneurysm of the descending thoracic aorta, contained in the adjacent structures, with a high risk of open rupture and exit to the thorax (Figure 1B). A PET-CT was performed to assess the extent of infection with a higher FDG-PET avidity favourable for a conservative treatment followed by elective surgery, rather than urgent intervention. The thoracic PET-CT demonstrated an hypermetabolic focus (Figure 1C). No definitive signs of endocarditis were seen on transthoracic and transesophageal echocardiograms. A few days later, he presented with haemoptysis and, given the severity of the clinical condition, a conical thoracic endoprosthesis was implanted using a bilateral percutaneous femoral approach. Following the procedure, the patient recovered well. A follow up CT 44 days after admission showed improvement in the aneurysm (Figure 1D). He was subsequently discharged from hospital 45 days later.

Discussion

We conducted a systematic review of the literature from 2009 to 2023 –15 years. We performed this search using 3 scientific databases –Embase, Cochrane and PubMed [keywords: ("Haemoptysis" (Mesh) and "Aneurysm, infected" (Mesh)], along with a manual search in the reference lists of the included articles, for published cases of infected aortic aneurysm (IAA) presenting with haemoptysis due to an aortobronchial fistula (Figure 2). Due to the infrequency of these cases, relevant aspects of the disease - including clinical presentation, evolution and the effectiveness of both medical and surgical treatments- remain relatively unknown.

We retrieved 88 articles, of which 12 cases were included [5-16] [7 men (58.3%) and 5 women; mean age: 69.8±11.7 years (91.7% ≥65 years)] (Table 1). Seventy-six studies were excluded due to non-aortic

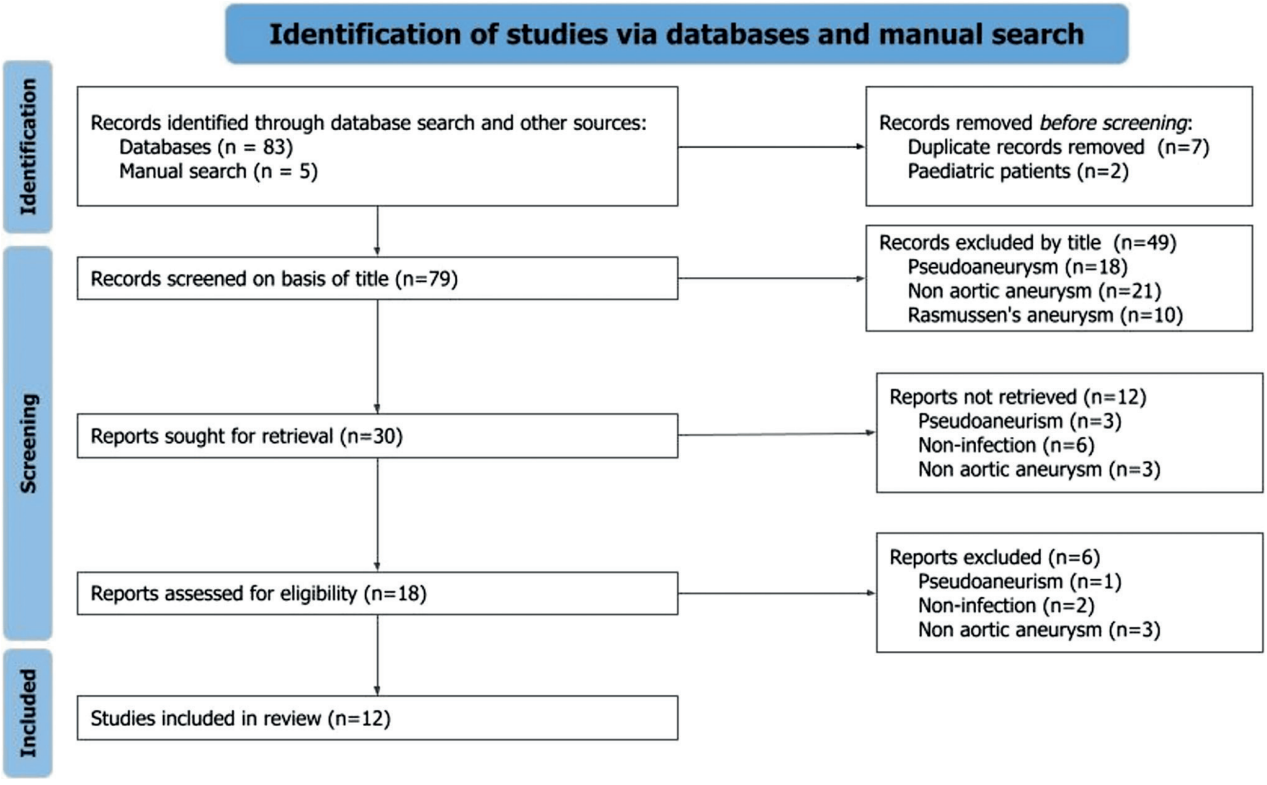

Figure 2. PRISMA flowchart of studies identification and selection process for this review.

Table 1. Main characteristics of the cases described in the literature.

Reference	Sex	Age	RF	Presentation	Localisation aorta	Germ	Aortobronchial fistula	Treatment	Outcome
Inoue, 2009[5]	M	65	Pyogenic spondylitis	Haemoptysis and fever	Thoracic aorta	*Salmonella*	+	Antibiotics, TEVAR and open surgery	Discharged
Daitoku, 2010[6]	F	73	-	Haemoptysis and fever	Aortic arch	-	+	Antibiotics and TEVAR	Discharged
Beiko, 2013[7]	F	67	MDS, BMT	Cardiorespiratory arrest	Aortic arch	*Aspergillus*	NR	Diagnosis at autopsy	Death
Tamura, 2013[8]	M	70	Diabetes mellitus	Haemoptysis, fever, and lower back pain	Thoracic aorta	*Klebsiella pneumoniae*	NR	Antibiotics and TEVAR	Discharged
Heneghan, 2015[9]	M	42	HBP, IV drugs	Haemoptysis, chest pain, dyspnoea	Aortic arch	*Methicillin-resistant Staphylococcus aureus*	NR	IV Antibiotics and TEVAR	Discharged
Hui, 2016[10]	M	85	HBP, Ca bladder	Haemoptysis, syncope	Thoracic aorta	*Mycobacterium bovis*	+	Palliative	Death
Isbir, 2017[11]	M	68	COPD, HBP	Haemoptysis, chest pain	Ascending aorta	*Treponema Pallidum*	+	IV Antibiotics and TEVAR	Discharged
Fernández, 2017[12]	F	71	HBP	Haemoptysis, dyspnoea, fever, chest pain	Thoracic aorta	*Escherichia Coli*	+	IV Antibiotics and TEVAR	Discharged
Novelli, 2018[13]	F	78	HBP, cirrhosis of the liver, diabetes mellitus	Haemoptysis	Aortic arch	*Mycobacterium Tuberculosis*	NR	Antituberculosis	Discharged
Wu, 2019[14]	M	68	HBP, DLP	Recurrent haemoptysis	Thoracic aorta	*Brucella suis*	+	IV Antibiotics and TEVAR	Discharged
Liu, 2022[15]	M	87	-	Haemoptysis, productive cough, and fever	Aortic arch	-	+	TEVAR and Open Surgery No antibiotics registered	Discharged
Tournaye, 2023[16]	F	79	Ischemic heart disease, atrial fibrillation	Haemoptysis, dyspnoea, chest pain	Aortic arch	*Staphylococcus aureus*	NR	Antibiotics and TEVAR	Discharged

AML, acute myeloid leukaemia; BMT, bone marrow transplant; Ca: carcinoma; COPD, chronic obstructive pulmonary disease; DLP, dyslipidaemia; DM, diabetes mellitus; F, female; HBP, high blood pressure; IV, intravenous; M, male; MDS, myelodysplastic syndrome; MRSA, methicillin-resistant *Staphylococcus aureus*; NR, no record; RF: risk factors; TEVAR, thoracic endovascular aortic repair by endoprosthesis.

*Histological finding: assumed to be tuberculosis (negative culture); +, present; -negative.

aneurysm, 27; pseudoaneurysm, 22; Rasmussen's aneurysm, 10; non-infection, 8; duplicate cases, 7; of paediatric age, 2.

In a previous series of 43 patients treated for IAA over a 25-year period –1976 to 2000-, 70% of individuals presented with at least one risk factor -pre-existing arterial damage from iatrogenic intervention, intravenous drug use, immunodeficiency disorders, history of infections at alternative sites, underlying atherosclerosis, or a pre- existing aneurysm [17].

In our case, several risk factors were identified: high blood pressure, diabetes mellitus, previous infections -endocarditis- and immunosuppression secondary to chronic lymphoid leukaemia. In the series reviewed over the last 15 years, 83% (10/12) presented with at least one identifiable risk factor [Table 1].

While most patients with IAA are symptomatic, the nonspecific nature of symptoms often delays diagnosis until advanced stages, when sepsis and/or rupture of the aneurysm has already occurred. In our review, haemoptysis- whether threatening, episodic, recurrent or self-limited- was required for inclusion and an aortobronchial fistula was demonstrated in 7 of 12 patients (58%)- unspecified in the rest. The classical triad of fever, chest pain, and pulsatile mass [17] is rarely present (Table 1). Overall, although haemoptysis presents a broad differential diagnosis, IAA should be considered despite its relatively low prevalence among thoracic aneurysm- not exceeding 2% of cases [3]. Early diagnosis is crucial to mitigate IAA's high morbidity and mortality [2].

According to literature, between 30-40% of ruptured thoracic aortic aneurysms occur in the descending thoracic aorta, with remaining cases involving the aortic arch and ascending aorta [18]. Despite its infectious nature, blood cultures and tissue cultures are not always positive, with positivity rates ranging from 50-85% and 76%, respectively [19]. Pathogens with a higher affinity for the arterial wall are usually gram-positive, especially *Staphylococcus aureus* [17], although gram-negative pathogens- comprising 35% of cases- pose a higher risk of complications [20]. The patient's aneurysm was infected with *Staphylococcus aureus*, consistent with 2/12 (17%) cases in the reviewed series.

There are no trials comparing the efficacy of IAA intervention by open surgery versus thoracic endovascular aortic repair (TEVAR) using a stent. Open surgery is typically selected when the risk of adverse events (e.g., dissection, rupture, sudden death) outweighs surgical risks, and there is appropriate anatomy and access, as it allows a more definitive repair [2]. TEVAR, however, has greater clinical utility and lower aneurysm-related mortality in the follow up period but carries a higher reoperation rate. Both in the patient described, who experienced no complications after a 20-month follow up period, and in the 9 reviewed patients, TEVAR outcomes were favourable, making it a viable option for ruptured IAA when open repair is contraindicated due to comorbidity or complex rupture. Out of the 12 patients reviewed, 2 died: one with a post-mortem diagnosis of IAA and another receiving palliative care for advanced malignancy.

Conclusions

In summary, infected aortic aneurysms are rare but critical conditions, particularly when involving the thoracic aorta. Haemoptysis, although uncommon, may signal an aortobronchial fistula, a potentially fatal complication. Our case highlights the diagnostic and management challenges associated with these aneurysms. Initially misdiagnosed as community-acquired pneumonia, the condition was later identified as an infected aneurysm through advanced imaging.

Managing infected aortic aneurysms is particularly challenging due to subtle and often non-specific symptoms, which can delay diagnosis and treatment. Identifying risk factors, performing appropriate imaging techniques and suspecting this entity, are essential for early clinical diagnosis and timely intervention, which are crucial for improving patient outcomes and preventing potentially fatal complications.

References

1. Osler W. The gulstonian lectures on malignant endocarditis. Br Med J 1885;1:467.
2. Isselbacher EM, Preventza O, Hamilton Black J 3rd, Augoustides JG, Beck AW, Bolen MA, et al. Guideline for the diagnosis and management of aortic disease: a report of the american heart association/american college of cardiology joint committee on clinical practice guidelines. Circulation 2022;146:334-482.
3. Sörelius K, Mani K, Björck M, Sedivy P, Wahlgren CM, Taylor P, et al. Endovascular treatment of mycotic aortic aneurysms. A European multicenter study. Circulation 2014;130:2136-42.
4. Cinà CS, Arena GO, Fiture AO, Clase CM, Doobay B. Ruptured mycotic thoracoabdominal aortic aneurysms: a report of three cases and a systematic review. J Vasc Surg 2001;33:861-7.
5. Inoue H, Iguro Y, Yamamoto H, Ueno M, Higashi A, Tao K, et al. Palliative stent-graft insertion followed by an

allograft replacement for an infected and ruptured aortic aneurysm. Ann Thorac Cardiovasc Surg 2009;15:261-4.
6. Daitoku K, Fukuda I, Taniguchi S, Minakawa, M. Endovascular treatment of an aortobronchial fistula caused by a distal aortic arch mycotic aneurysm: report of a case. Surg Today 2010;40:54-6.
7. Beiko T, Huggins J. A mycotic aneurysm and a pulmonary nodule in immunocompromised patient: where is occam's razor when you need it?. Chest 2013;144 (meeting abstract): no pagination.
8. Tamura K, Yoshitaka H, Totsugawa T, Tsushima Y, Chikazawa G, Ohno T, et al. Bridge use of endovascular repair and delayed open operation for infected aneurysm of aortic arch. Ann Thorac Surg 2013;96:1471-3.
9. Heneghan RE, Singh N, Starnes BW. Successful emergent endovascular repair of a ruptured mycotic thoracic aortic aneurysm. Ann Vasc Surg 2015;29:843.e1-e.6.
10. Hui DS, Stoeckel DA, Kaufman EE, Jacobs DL. Massive hemoptysis from an aortobronchial fistula secondary to BCG-related mycotic thoracic aortic aneurysm. Ann Thorac Surg 2016;101:350-2.
11. Isbir S, Hamidov A, Seven IE, Ak K. Massive hemoptysis related to contained rupture of syphilitic aortic aneurysm into the pulmonary parenchyma. J Thorac Cardiovasc Surg 2017;154: 23-5.
12. Fernandez L, Gutierrez L, Yara JD. A case of massive hemoptysis secondary to aortobronchial fistula caused by mycotic aneurysm in thoracic aorta: a challenging multidisciplinary therapeutic approach. Am J Respir Crit Care Med 2017;195:1931.
13. Novelli M, Cataldi A, Pilato A, Quadri R, Savoldi S. Tuberculous mycotic aneurysm of the aorta: a case report of haemoptysis. Recenti Prog Med 2018;109:398-400.
14. Wu SJ, Huddin JC, Wanger A, Estrera AL, Buja LM. A case of Brucella aortitis associated with development of thoracic aortic aneurysm and aortobronchial fistula. Cardiovasc Pathol 2019;39:5-7.
15. Liu C-H, Huang S-C, Hsu C-T. A fatal masquerade in pneumonia: ruptured thoracic aortic aneurysm. Clin Case Rep 2022;10:e05285.
16. Tournaye E, Hollering P, De Roover D, Dossche K, Vercauteren SRW. Staphylococcus aureus sepsis and hemoptysis as messengers of a rather impractically located mycotic aneurysm. Acta Chirurgica Belgica 2023;123: 430-5.
17. Oderich GS, Panneton JM, Bower TC, Cherry KJ, Rowland CM, Noel AA, et al. Infected aortic aneurysms: aggressive presentation, complicated early outcome, but durable results. J Vasc Surg 2001;34:900-8.
18. Taylor LM, Deitz DM, McConnell DB, Porter JM. Treatment of infected abdominal aortic aneurysms with extra-anatomic bypass, aneurysm excision, and drainage. Am J Surg 1988;155:655-8.
19. Kilic A, Shah AS, Black JH, Whitman GJ, Yuh DD, Cameron DE, et al. Trends in repair of intact and ruptured descending thoracic aortic aneurysms in the United States: a population-based analysis. J Thorac Cardiovasc Surg 2014;147:1855-60.
20. Negishi K, Ono Y, Kurosawa K, Takamatsu H, Nakano A, Hasegawa A, et al. Infective endocarditis complicated by mycotic aneurysm of a coronary artery with a perforated mitral valvular aneurysm. J Am Soc Echocardiogr 2009;22:542-4.
21. Jarrett F, Darling RC, Mundth ED, Austen WG. Experience with infected aneurysms of the abdominal aorta. Arch Surg 1975;110:1281-6.

Received for publication: 8 October 2024 - Accepted for publication: 15 November 2024

Licensee Mattioli 1885, Italy
Multidisciplinary Respiratory Medicine 2025; 20: 1004
doi: 10.5826/mrm.2025.1004