

CASE REPORT

INTERMEDIATE

CLINICAL CASE

# Esophageal Lesion Reveals an Aortic Pseudoaneurysm in the Setting of *Actinomyces odontolyticus* Bacteremia



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## ABSTRACT

A toddler presented with hematemesis a few weeks after ingesting a penny. Workup revealed an esophageal lesion communicating with an aortic pseudoaneurysm in the setting of *Actinomyces odontolyticus* bacteremia. *A. odontolytica* is an oropharyngeal bacteria known to cause fistulas when introduced into tissue planes. (**Level of Difficulty: Intermediate.**) (J Am Coll Cardiol Case Rep 2023;15:101867) Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## HISTORY OF PRESENTATION

A previously healthy 19-month-old female patient presented after an episode of hematemesis. One month prior, she swallowed a penny and passed it several days later. Two weeks prior to presentation, she had a week of nonbloody, nonbilious emesis that resolved. Three days prior, she had an episode of melena followed by decreased appetite limited to

only liquid intake. Subsequently, she had an episode of hematemesis prompting mother to take her to the local emergency department.

On arrival, she was pale, listless, and febrile to 101.9 °F, with labs demonstrating a hemoglobin of 12.5 g/dL (reference 11.3-14.1 g/dL). Chest and abdominal x-ray films were normal. She received ondansetron and a normal saline bolus and was transferred to a tertiary pediatric hospital.

On arrival to the pediatric emergency department, her vital signs were blood pressure 128/76 mm Hg, heart rate 140 beats/min, respiratory rate 44 breaths/min, oxygen saturation 100%, and temperature 97.5 °F. She was alert, interactive, and not in acute distress. Physical exam was remarkable for conjunctival pallor and dried blood at the nares and lateral commissures with no active bleeding. She had a regular rhythm and no murmurs. Lungs were clear to auscultation bilaterally; she was not in respiratory distress. Abdomen was soft, nontender, and

## LEARNING OBJECTIVES

- To describe life-threatening sequela from a corrosive foreign body injury in the esophagus.
- To understand the pathophysiology of aortic pseudoaneurysm after *Actinomyces* infection.
- To describe the intravascular approach for aortic pseudoaneurysm.

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**ABBREVIATIONS  
AND ACRONYMS**

CT = computed tomography

nondistended with active bowel sounds. Extremities were warm and well perfused.

She was admitted to the gastroenterology service for further workup of hematemesis.

**PAST MEDICAL HISTORY**

She had no significant medical or surgical history.

**DIFFERENTIAL DIAGNOSIS**

Differential for hematemesis and melena includes mucosal injury by a foreign body, Mallory-Weiss tear, arteriovenous malformation, gastritis, peptic ulcer disease, variceal bleeding, and intussusception.

**INVESTIGATIONS**

Lab work was notable for anemia (hemoglobin 6.7 g/dL) and mild coagulopathy (prothrombin time 18.2 seconds [reference 10.8-15.8 seconds], international normalized ratio 1.5 [reference 0.8-1.2], partial thromboplastin time 33.7 seconds [reference 23.1-36.3 seconds]). She underwent an endoscopy, which revealed a vascular, erythematous exophytic lesion along the wall of the midesophagus measuring 3 × 1 cm (Figure 1), with numerous intraepithelial eosinophils seen on histopathology. The stomach and duodenum were normal. A chest computerized tomography (CT) demonstrated an aortic pseudoaneurysm measuring 18 × 12 × 12 mm arising from the rightward aspect of the aortic isthmus that extended rightward, with mass effect on her esophagus, and surrounding inflammation (Figure 2). Two days later,

a cardiac CT demonstrated interval increasing size (1-2 mm) of the pseudoaneurysm.

Given her fever on admission and concerns for inflammation surrounding the pseudoaneurysm, she underwent an infectious evaluation: *Helicobacter pylori* stool antigen, *Bartonella* immunoglobulin G/immunoglobulin M, syphilis screen, T spot, and QuantiFERON gold were negative. Blood cultures grew *Actinomyces odontolyticus*.

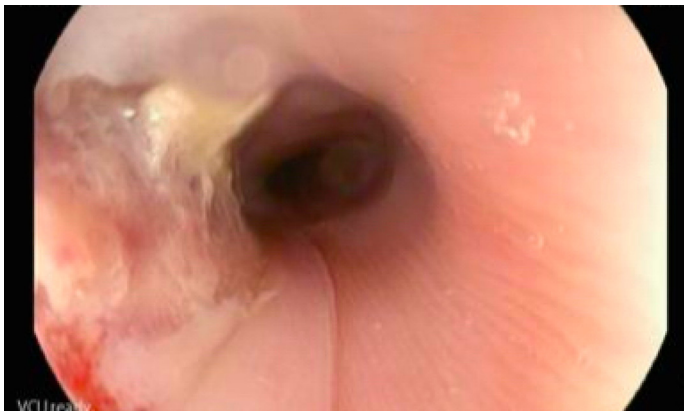
**MANAGEMENT**

On admission, she received 1 unit of packed red blood cells, was made nothing by mouth, and started on an intravenous proton pump inhibitor. She received intravenous vitamin K for 3 days to correct her coagulopathy. Following the discovery of her aortic pseudoaneurysm, she was transferred to the cardiac intensive care unit for closer monitoring given the risk of hemorrhage.

After multidisciplinary review with cardiology, gastroenterology, interventional cardiology, cardiovascular surgery, and cardiac intensive care unit teams, the decision was made to pursue emergent placement of a covered stent to exclude the pseudoaneurysm. On hospital day 4, she underwent carotid artery access via right common carotid artery cutdown by cardiovascular surgery with proximal descending aorta covered stent placement (9 × 29 mm Gore Viabahn; W.L. Gore and Associates) by interventional cardiology (Figure 3). She tolerated the procedure well, without complications. She was empirically started on vancomycin and ampicillin-sulbactam. On hospital day 8, blood cultures that were drawn same day as stent placement grew *A. odontolytica*. Antibiotic therapy was narrowed to high-dose penicillin (the drug of choice for *Actinomyces*) for 2 weeks, followed by ceftriaxone for 4 weeks. Repeat blood cultures were negative. Prior to discharge, an echocardiogram demonstrated a patent aortic stent, possible patent foramen ovale with left-to-right flow, and normal biventricular size and function.

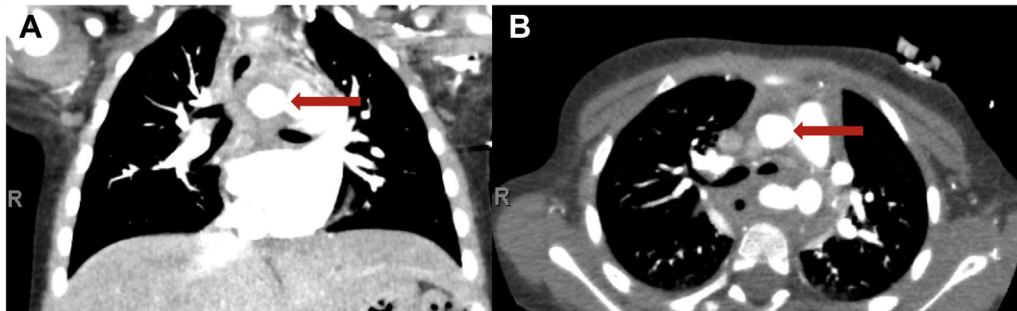
**DISCUSSION**

Our case indicates that communications with the aorta should be considered when a patient presents with hematemesis as it is, albeit a very rare but potentially lethal cause. Yang et al<sup>1</sup> described an adult patient who presented with chest pain and hematemesis 12 days after eating fish. This patient was found to have an esophageal

**FIGURE 1** Upper Endoscopy Image

A 3 × 1 cm vascular, nonbleeding exophytic lesion on the wall of the midesophagus.

**FIGURE 2** Computed Tomography of Chest With Intravenous Contrast



(A) Coronal slice demonstrating an 18 × 12 × 12 mm pseudoaneurysm (arrow). (B) The axial view again demonstrates the pseudoaneurysm (arrow) from the rightward aspect of the aortic isthmus with rightward displacement of the esophagus.

perforation due to the fish bone migrating into the thoracic aorta.

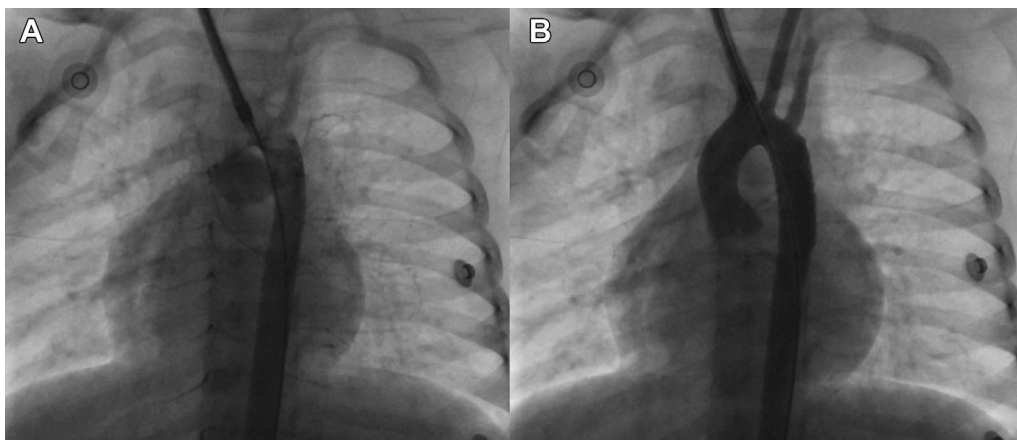
Mycotic aortic aneurysms are rare in children; however, Marques da Silva et al<sup>2</sup> described bacteria found in 53 tissue samples from aortic aneurysm walls, collected from 49 patients during reconstructive surgery. All of the bacteria were gram-positive cocci or rods, and 71% were anaerobic bacteria including *A. viscosus* and *A. naeslundii*. It was not clear if the bacteria were causes of the pseudoaneurysms or secondary colonizers.

Thoracic actinomycosis is most commonly due to aspiration of oropharyngeal secretions. However, it can rarely be caused by nonpenetrating trauma

disrupting the esophagus. When *Actinomyces* is introduced into tissue, it can create sinus tracts across tissue planes.<sup>3</sup> An adult patient presented with cardiac tamponade due to pleuropericardial disease in the setting of *A. odontolyticus* infection that was likely seeded to the lungs during gastric surgery.<sup>4</sup> Our patient swallowed the penny, likely disrupting the lining of the esophagus and introducing *Actinomyces*, with subsequent development of a communication between the esophagus and the aorta.

There are no specific guidelines for use of covered stents in pseudoaneurysms in children. This case involved a multidisciplinary discussion regarding the placement of a covered stent in a patient with active

**FIGURE 3** Anteroposterior Fluoroscopic Images From Covered Stent Placement



(A) Fluoroscopic image demonstrating contrast filling the pseudoaneurysm arising from the proximal descending aorta (arrow). (B) Fluoroscopic image demonstrating the placement of a covered stent with contrast no longer filling the pseudoaneurysm.

bleeding. Surgical repair was felt to not be the best option due to infected tissue and potential difficulty with bleeding control. Decisions were limited by the patient's size, as she will eventually outgrow the stent and need subsequent procedures. However, given her presentation with a herald bleed, we felt that endovascular treatment would be life-saving for her. The gradient across the stent will be monitored noninvasively with echocardiography as well as upper extremity-to-lower extremity blood pressure gradients. If she developed a gradient >20 mm Hg she would require a balloon expansion of the stent. If she doesn't develop a gradient, she would undergo diagnostic cardiac catheterization electively in a few years. Thalhammer et al<sup>5</sup> described 26 endovascular stents that were used to repair 16 pseudoaneurysms, 9 arteriovenous fistulas, and 1 combined lesion in large arteries. Patients in this cohort tolerated it well, with only 4 thrombosed stents at 1-year follow-up.

#### FOLLOW-UP

During admission, an aortopathy panel was sent to evaluate for a genetic cause of her pseudoaneurysm. The panel was remarkable for a variant of unknown significance in the *ELN* gene (c.592C>T, p.Pro198Ser). In the cardiology clinic, she was doing very well without residual symptoms with repeat cardiac CT showing resolution of the pseudoaneurysm. To receive ceftriaxone at home, she was discharged with a central line that became dislodged, requiring that antibiotic therapy be changed to penicillin. Given that

she was on long-term penicillin, she was monitored for neutropenia, which she developed, so penicillin was held until her cell counts recovered. She completed 9 total months of antibiotic therapy, consistent with the recommendation for invasive *Actinomyces* infections.

#### CONCLUSIONS

To our knowledge, this is the youngest reported patient to date in whom a foreign body ingestion induced bacterial seeding and subsequent aorto-esophageal communication. The case highlights the potential for life-threatening sequela from blunt and nonsignificantly corrosive foreign body ingestion into the esophagus. Even young patients can be successfully managed with an endovascular covered stent with prolonged antibiotic therapy against pathogens under such circumstances. The case underscores the importance for the timely removal of esophageal foreign bodies with appropriate subsequent follow-up.

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The authors have reported that they have no relationships relevant to the contents of this paper to disclose.

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**KEY WORDS** aorta, cardiovascular disease, left-sided catheterization