Upper Gastrointestinal Bleeding From Aberrant Right Subclavian Artery-Esophageal Fistula

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Background: The most common aortic arch abnormality is an aberrant right subclavian artery (ARSA). ARSA-esophageal fistula is a rare sequela that can present with a life-threatening upper gastrointestinal (GI) bleed.

Case Report: We report the case of an 88-year-old male who presented with signs of upper GI bleeding. Esophagogastroduodenoscopy demonstrated extrinsic compression of the upper third of the esophagus with ulceration. Imaging studies revealed ARSA posterior to the esophagus with pseudoaneurysm formation. These findings confirmed an upper GI bleed secondary to ARSA-esophageal fistula. The patient underwent prompt embolization of the ARSA pseudoaneurysm, followed a few days later by coil embolization of the ARSA pseudoaneurysm. Despite these interventions, the patient continued to have bleeding with anemia. He and his family opted to avoid any further interventions and instead pursued comfort care. The patient was discharged to hospice and died 3 months later.

Conclusion: ARSA-esophageal fistula is a rare but potentially lethal cause of upper GI bleeding. Initial signs and symptoms can be subtle, but the presence of a GI bleed requires immediate stabilization. Surgical interventions have been shown to have longer-lasting success, but endovascular repair may be an option for patients who are deemed unfit for surgery but still require prompt stabilization. Regardless of the intervention, mortality rates for ARSA-esophageal fistula are high.

Keywords: Aberrant right subclavian artery, aneurysm, esophageal fistula, gastrointestinal hemorrhage

INTRODUCTION
Aberrant right subclavian artery (ARSA), or arteria lusoria, is the most common aortic arch abnormality, with a prevalence of 0.2% to 2.5%.1,2 ARSA is typically an incidental finding that seldom causes any symptoms.3 However, ARSA is prone to aneurysmal development that can remain asymptomatic or cause compressive symptoms, including shortness of breath and dysphagia lusoria secondary to extrinsic compression of the esophagus.3 The recurrent laryngeal nerve is also prone to a degree of constriction from ARSA, causing progressive hoarseness. An even rarer consequence is the development of ARSA-esophageal fistula that can present as a life-threatening upper gastrointestinal (GI) bleed, so prompt diagnosis and management are essential for patient survival.

CASE REPORT
An 88-year-old male presented to the emergency department (ED) with a 1-day history of shortness of breath. He reported one episode of dark black, bloody stools 2 days prior. He also reported dysphagia for 6 months and hoarseness of voice for 1 week. He had an extensive cardiovascular history, including aortic stenosis status post transcatheter aortic valve replacement, thoracic aortic aneurysm (TAA), atrial fibrillation on warfarin, and congestive heart failure. A month and a half prior to his presentation, computed tomography (CT) angiography of the chest to evaluate the TAA demonstrated an ascending TAA and saccular pseudoaneurysm formation from an ARSA, resulting in marked mass effect and displacement of the esophagus (Figure 1). Bilateral carotid subclavian bypass was recommended; however, the patient declined surgery at the time.

On presentation to the ED, he was hemodynamically stable. Laboratory workup revealed hemoglobin of 6.5 g/dL from a baseline of 11.0 g/dL. Prothrombin time was 17.1 seconds, with an international normalized ratio (INR) of 1.8. Following initial resuscitation with 2 units of packed red blood cells and intravenous pantoprazole, esophagogastroduodenoscopy (EGD) was performed and demonstrated extrinsic compression of the upper third of the esophagus, with associated areas of ulceration and visible underlying vasculature (Figure 2). EGD findings were concerning for development of ARSA-esophageal fistula and the potential for subsequent bleeding. Vascular surgery performed urgent embolization of the ARSA using Amplatzer plugs.
Figure 1. Computed tomography angiography of the chest demonstrates an ascending thoracic aortic aneurysm with saccular pseudoaneurysm formation. Arrows point to the aneurysm compressing the esophagus.

Figure 2. Esophagogastroduodenoscopy demonstrates (A) external compression into the lumen of the esophagus and (B) the inferior part of the compression with ulceration (arrows) and possible visible vessel.

Figure 3. Interventional radiology imaging demonstrates (A) precoiled pseudoaneurysm and (B) postcoiled pseudoaneurysm.

The patient initially did well and had no further signs of bleeding. He was discharged 2 days after the procedure with hemoglobin of 7.7 g/dL and INR of 1.2. One day after discharge, he presented again to the ED with worsening fatigue, weakness, and shortness of breath. He had 2 additional episodes of melena and a drop in hemoglobin to 6.5 g/dL. He was readmitted, and repeat CT angiography demonstrated a new bleed within the pseudoaneurysm sac. In conjunction with vascular surgery, interventional radiology performed a coil embolization of the ARSA pseudoaneurysm (Figure 3).

Despite these interventions, the patient continued to have ongoing signs of bleeding with anemia. He required multiple blood transfusions for stabilization, ultimately receiving...
In our review of literature, we found 35 reported cases of ARSA-esophageal fistula,1,11-43 Of the 35 cases, 33 cases are included in our summary table, while 2 cases were excluded due to inability to access the primary source or the source being in a language other than English.13,23,32 Six cases were in the pediatric population, with patient ages ranging from 5 months to 11 years.22,28,30,34,37 Although diagnosis is typically made with CT imaging, endoscopic visualization may be the initial diagnostic test to exclude other causes, particularly in patients who are hemodynamically stable at the time of presentation. The most common etiology for development of ARSA-esophageal fistula appeared to be secondary to compression, friction, or pressure necrosis from recent instrumentation with an endotracheal tube, nasogastric tube, or tracheostomy tube, seen in 14 of the 33 summarized cases.14,15,17,19,21,22,24,27,29,31,38-41 Fistula development from prior placement of esophageal stent was noted in 5 cases.34,36,42,43 Other reported causes for fistula formation included placement of a salivary bypass tube and a possible consequence of gastric pull-up surgery.12,28

Fistula formation attributable to ARSA aneurysm with resultant GI bleeding is rare. Only 10 other cases attributed to this etiology have been reported.11,12,16,18,19,23,25,33,36,42 Only 1 patient with upper GI bleed secondary to ARSA aneurysm survived all bleeding episodes, and the patient required balloon tamponade with surgery to replace the aorta.33 Our patient survived the initial bleeding event, but died 3 months later, as he declined definitive surgical intervention to stop the recurrent bleeding.

Bleeding from ARSA-esophageal fistula is life-threatening, with a high mortality rate at initial presentation, regardless of etiology. Of the 33 summarized cases, thirteen cases survived all episodes of bleeding (39%).21,22,24,27,28,30,31,33,34,36-38,43 The severity of bleeding, in the setting of several comorbid conditions, likely contributed to the high mortality rate.

Opinions conflict regarding acute treatment for bleeding from ARSA-esophageal fistula secondary to aneurysm, with the historic approach being excision with reconstruction of the aorta and esophagus.33 A more recent approach (2000) is placement of an endovascular stent, but success with this approach is questionable because of the rates of infection that may result in the need for further surgery.44 Reported cases in adults involved treatment with surgery alone in 5 cases,11,16,26,30,38 endovascular repair alone in 2 cases,41,42 and a combination of endovascular and surgical intervention in 6 cases.28,29,31,35,36,43 Balloon tamponade was performed in 16 cases,1,14,15,21,22,24,25,27,28,31,33,35-37 with 3 of those 16 performed solely with endovascular repair.1,27,32 Six of the 11 patients who underwent either surgery alone or surgery with endovascular repair survived (55%),28,30,31,36,38,43 whereas only 1 of the 5 cases of endovascular repair with or without balloon tamponade survived (20%).27 Although the higher percentage of survival among patients who had surgery with or without endovascular repair compared to the 5 patients who underwent endovascular repair without surgery might suggest a better outcome with surgery, this discrepancy is likely confounded by patient factors and comorbidities for which surgical intervention is usually deferred, as well as the relatively low number of patient cases reported to have been treated with endovascular repair without surgery.

DISCUSSION

When ARSA is present, the right subclavian artery branches distal to the left subclavian artery and serves as the fourth branch of the left-sided aortic arch (Figure 4). From there, the artery travels posterior to the esophagus in 80% of cases but can also be found between the trachea and esophagus in 15% of cases and anterior to the trachea in 5% of cases.4 ARSA has been reported to have a female predominance as high as 3:1, although other studies suggest no significant sex discrepancy.5 ARSA can be associated with other medical conditions and is seen in up to 25% of patients with esophageal atresia and in 16% to 39% of patients with Down syndrome.6,7

Despite its prevalence in the general population, ARSA is frequently asymptomatic and discovered incidentally on imaging. However, ARSA can present with dysphagia secondary to esophageal compression, a phenomenon known as dysphagia lusoria.8 Depending on the exact location, ARSA can cause some degree of compression of the recurrent laryngeal nerve, resulting in hoarseness, a condition known as Ortner syndrome.9 ARSA can also cause compression of the trachea, resulting in shortness of breath. With advancing age, ARSA is prone to aneurysmal development or sclerosis.3 The late presentation of dysphagia in our elderly patient is not uncommon, as dysphagia is more frequently seen in the elderly compared to younger patients with ARSA and can be attributed to decreased flexibility of the esophagus or increasing compression from the developing aneurysm. Some degree of atherosclerosis contributing to rigidity of the ARSA is also possible.10

Figure 4. Schematic representation of aberrant right subclavian artery. (Image courtesy of Dr Vincent Tatco, Radiopaedia.org, rID: 52193 radiopaedia.org/cases/development-of-aberrant-right-subclavian-artery-illustration)
| Study                  | Age, Sex | Medical History                                                                 | Etiology                                                                 | Treatment                        | Outcome                          |
|------------------------|----------|----------------------------------------------------------------------------------|--------------------------------------------------------------------------|----------------------------------|-----------------------------------|
| Lynn, 1969<sup>11</sup> | 57, M    | Atherosclerosis, hypertension, angina                                             | ARSA aneurysm                                                            | Surgery                          | Fatal                             |
| Reynes et al, 1976<sup>12</sup> | 72, F    | Mediastinal mass                                                                  | ARSA aneurysm                                                            | None                             | Fatal                             |
| Livesay et al, 1982<sup>14</sup> | 25, M    | Motor vehicle accident with traumatic head injury                                 | Endotracheal tube, nasogastric tube, and tracheostomy                    | Balloon tamponade, surgery       | Fatal                             |
| Belkin et al, 1984<sup>15</sup> | 27, M    | Head and neck squamous cell carcinoma                                            | Nasogastric tube                                                         | Balloon tamponade, surgery       | Fatal                             |
| Edwards et al, 1984<sup>16</sup> | 81, M    | Mass in right upper hemithorax (tortuous innominate artery)                       | ARSA aneurysm                                                            | Surgery                          | Fatal                             |
| Gossot et al, 1985<sup>17</sup> | 72, F    | Type I aortic dissection with endoluminal prosthesis, giant cell arteritis       | Endotracheal tube, nasogastric tube, and tracheostomy                    | None                             | Fatal                             |
| Kullnig, 1989<sup>18</sup>   | 66, M    | Hypertension                                                                     | ARSA aneurysm                                                            | None                             | Fatal                             |
| Stone et al, 1990<sup>19</sup> | 72, M    | Smoker, status post cardiopulmonary bypass surgery                               | ARSA aneurysm, tracheostomy                                              | None                             | Fatal                             |
| Hirakata et al, 1991<sup>21</sup> | 55, M    | Status post subtotal esophagectomy for esophageal cancer                          | Nasogastric tube, radiation arteritis, and other surgical trauma         | Balloon tamponade                | Survived                          |
| Miller et al, 1996<sup>22</sup> | 11, F    | Status post craniotomy for intracerebral hemorrhage                              | Endotracheal tube, nasogastric tube                                       | Balloon tamponade, surgery       | Survived                          |
| Singha et al, 1998<sup>23</sup> | 82, M    | N/R                                                                              | ARSA aneurysm                                                            | None                             | Fatal                             |
| Feugier et al, 2002<sup>24</sup> | 24, M    | Polytrauma, burns, alcohol abuse                                                  | Nasogastric tube and tracheostomy                                       | Balloon tamponade, surgery       | Survived                          |
| Lehmann et al, 2006<sup>25</sup> | 78, M    | N/R                                                                              | ARSA aneurysm                                                            | Balloon tamponade                | Fatal                             |
| Millar et al, 2007<sup>26</sup> | 57, M    | Status post esophagectomy with gastric pull-up for esophageal cancer             | Pressure from gastric pull-up vs foreign body                             | Surgery                          | Fatal (survived initial hemorrhage but died 18 days later from additional bleed) |
| Inman et al, 2008<sup>1</sup>   | 63, M    | Supraglottic squamous cell carcinoma                                             | Salivary bypass tube                                                     | Endovascular repair, balloon tamponade | Fatal                             |
| Magagna et al, 2008<sup>27</sup> | 73, F    | Status post laryngectomy and tracheostomy for laryngeal carcinoma                | Tracheostomy                                                             | Endovascular repair, balloon tamponade | Survived                          |
| Fuentes et al, 2010<sup>28</sup> | 3, F     | Esophageal atresia type III                                                      | Esophageal prosthesis                                                    | Endovascular repair, balloon tamponade, surgery | Survived                          |
| Chapman et al, 2010<sup>29</sup> | 34, F    | Motor vehicle accident with trauma                                               | Endotracheal tube, nasogastric tube, and tracheostomy                    | Endovascular repair, balloon tamponade, surgery | Fatal                             |
| Situma et al, 2011<sup>30</sup> | 5 months, F | Esophageal atresia with distal fistula                                           | Status post colonic esophageal grafting                                  | Surgery                          | Survived                          |
| Jain et al, 2012<sup>31</sup>   | 57, F    | Status post cardiopulmonary bypass surgery for scimitar syndrome                 | Endotracheal tube, nasogastric tube, and tracheostomy                    | Endovascular repair, balloon tamponade, surgery | Survived                          |
| Study                  | Age, Sex | Medical History                                                                 | Etiology                                                                 | Treatment                                    | Outcome | Note                                                                 |
|------------------------|----------|---------------------------------------------------------------------------------|---------------------------------------------------------------------------|----------------------------------------------|---------|----------------------------------------------------------------------|
| Pop et al, 2012        | 67, M    | Status post transhiatal esophagectomy for esophageal cancer, status post         | Damage to arterial wall from cervical sepsis and/or staples from gastric | Endovascular repair, balloon tamponade      | Fatal   |                                                                      |
|                        |          | pneumonectomy for lung cancer, neuroendocrine liver cancer                      | tubing                                                                    |                                              |         |                                                                      |
| Takahashi et al, 2013  | 63, M    | N/R                                                                             | Infected ARSA aneurysm                                                   | Balloon tamponade, surgery                  | Survived|                                                                      |
| Lo et al, 2013         | 16 months, N/R | Esophageal atresia with distal fistula                                           | Esophageal and Polyflex airway stent                                    | Endovascular repair, Bougienage tamponade   | Fatal   |                                                                      |
|                        | 18 months, N/R | Esophageal atresia, duodenal atresia                                            | Esophageal stent                                                          | Surgery, hydrostatic dilator                | Survived|                                                                      |
| Morisaki et al, 2014   | 74, F    | Rheumatic arthritis                                                             | ARSA aneurysm                                                             | Endovascular repair, balloon tamponade, surgery | Fatal   |                                                                      |
| Hosn et al, 2014       | 29, F    | Sleeve gastrectomy                                                              | Esophageal stent                                                          | Endovascular repair, balloon tamponade, surgery | Survived|                                                                      |
| Joynt and Grifka, 2015 | 17 months, F | N/R                                                                            | Spontaneous development of fistula                                        | Balloon tamponade, surgery                  | Survived|                                                                      |
| Watanabe et al, 2015   | 55, M    | Intracranial hemorrhage                                                         | Nasogastric tube and tracheostomy                                        | None                                         | Fatal   |                                                                      |
| Oliveira et al, 2016   | 20, M    | Motor vehicle accident with polytrauma                                          | Endotracheal tube and nasogastric tube                                   | Surgery                                      | Survived|                                                                      |
| Kudose et al, 2017     | 20, F    | VATER (vertebral, anal, tracheal, esophageal, and renal) association, primary  | Tracheostomy                                                              | None                                         | Fatal   |                                                                      |
|                        |          | pulmonary hypertension, diabetes mellitus, atrial septal defect, status post 3   |                                                                         |                                              |         |                                                                      |
|                        |          | lung transplants                                                                |                                                                         |                                              |         |                                                                      |
| Shires and Rohrer, 2018| 44, F    | Gastroesophageal reflux disease, hypertension, pneumonia                        | Nasogastric tube, endotracheal tube, and tracheostomy                    | Endovascular repair                          | Fatal   |                                                                      |
| Zheng et al, 2019      | 67, M    | Esophageal and laryngeal cancer, hypertension                                    | Esophageal stent and pseudoaneurysm                                      | Endovascular repair                          | Fatal   |                                                                      |
| Merlo et al, 2020      | 29, F    | Tracheoesophageal fistula, ventriculoperitoneal shunt for hydrocephalus          | Esophageal stent                                                          | Endovascular repair, surgery                | Survived|                                                                      |
| Present case, 2021     | 88, M    | Hypertension, transcatheter aortic valve replacement, thoracic aortic aneurysm  | ARSA aneurysm                                                             | Endovascular repair                          | Fatal (survived initial event, but died 3 months after discharge from recurrent bleeds) |        |                                                                      |

Note: Age is given in years unless otherwise indicated.
ARSA, aberrant right subclavian artery; F, female; M, male; N/R, not reported.
CONCLUSION

ARSA-esophageal fistula is an uncommon and potentially fatal cause of GI bleeding. Patients with known ARSA aneurysm should elicit a high index of suspicion, particularly those who present with concomitant worsening dysphagia. Given the high mortality with active bleeding, endovascular repair is a potential alternative for patients who are not suitable for surgery because of their comorbidities or high surgical risk. However, even with intervention, mortality remains high.

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