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Development and repair of aorto-esophageal fistula following esophageal button battery impaction: A case report

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ABSTRACT

Background: Complications from esophageal button battery impactions remain a real fear for practicing pediatric gastroenterologists and surgeons. This case describes a child who developed an aorto-esophageal fistula 25 days after initial battery ingestion and survived due to prompt placement of an aortic stent via minimally invasive surgery, avoiding an open procedure.

Case presentation: A 6-year-old female presented acutely with a mid-esophageal button battery impaction witnessed by her parents. Presenting symptoms included chest pain and emesis. Button battery location and size were confirmed on X-ray. She underwent removal with flexible esophagogastroduodenoscopy (EGD) and rigid esophagoscopy. She was admitted to the hospital and received conservative medical management, with serial cross-sectional imaging via chest MRIs to assess the evolution of her injury according to available national guidelines, and was discharged after 12 days of close inpatient monitoring. Despite these measures the patient represented 25 days post-ingestion with hematemesis from a new aorto-esophageal fistula, requiring emergent cardiac catheterization with successful, life-saving aortic stent placement. She remained admitted for an additional 12 days of monitoring as her diet was advanced slowly post-catheterization. Since this second hospitalization she continues to do well, with outpatient follow-up by multiple subspecialists.

Conclusions: This case highlights the continued uncertainty regarding the risk of developing this complication, as well as gaps in the current literature and guidelines for managing these patients following ingestion and esophageal injury. It also details the unique course following development of this complication and its surgical repair.

1. Background

Esophageal button battery (BB) impactions in children continue to be harrowing cases for pediatric gastroenterologists and surgeons. Patients are at risk for rare, serious complications such as formation of a tracheoesophageal fistula (TEF), an esophageal stricture, or a predominantly fatal complication of an aorto-esophageal fistula (AEF). Expert opinion-based guidelines exist for battery removal and immediate post-removal care [1]. However short and long-term clinical management of these injuries remains challenging, as limited literature exists detailing clinical course following battery removal in regards to patient

monitoring, serial imaging, or feeding practices to determine which interventions may predict or even prevent these complications. We present a case of a 6-year-old female with esophageal impaction of a button battery who survived after developing an AEF 25 days following ingestion due to prompt aortic stent placement.

2. Case presentation

A previously healthy 6-year-old female presented to a local emergency department (ED) with chest pain and emesis after a witnessed ingestion of a 21 mm BB. Chest x-ray demonstrated a BB in the middistal esophagus, with the negative pole oriented posteriorly (Fig. 1A

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List of Abbreviations

EGD Esophagogastroduodenoscopy AEF Aorto-esophageal Fistula

BB Button Battery

TEF Tracheo-esophageal Fistula

HD Hospital Day

ED Emergency Department

NG nasogastric

MRI magnetic resonance imaging PICU Pediatric Intensive Care Unit

CVL: central venous line pRBCs Packed red blood cells FFP Fresh Frozen Plasma

NASPGHAN North American Society of Pediatric

Gastroenterology, Hepatology, and Nutrition

and B). Sucralfate and honey were not given and the Poison Center was not contacted by the ED. She was immediately transferred to our tertiary care facility, arriving 4 hours after ingestion. Institutional BB removal protocol was initiated and the patient was emergently taken for endoscopy. Removal was first attempted via flexible endoscopy by pediatric gastroenterology and was unsuccessful due to the severely limited visualization from food, debris and active esophageal bleeding near the proximal posterior edge of the battery. Pediatric surgery was consulted and was able to remove the battery via rigid esophagoscopy

approximately 6 hours after ingestion. On endoscopy the midesophagus appeared erythematous, corroded, and ulcerated (Fig. 2A-F). Of note, acetic acid wash was not performed after rigid endoscopic removal. A nasogastric (NG) tube was placed and the patient was transferred to the Pediatric Intensive Care Unit (PICU). Subsequent chest magnetic resonance imaging (MRI) showed injury at the midesophagus, with inflammatory changes abutting and surrounding the descending aorta (Fig. 3A-C). The patient was started on intravenous ampicillin-sulbactam and NG tube feeds, with nothing by mouth. Two days post-removal she developed fever to 38.9° Celsius, with C-reactive protein elevated to 8.8 mg/dL, transient respiratory distress needing high-flow nasal cannula and atelectasis on chest x-ray. An esophagram was performed to assess for any esophageal leak, which showed a persistent filling defect along the posterior wall of the distal esophagus. A repeat MRI performed 5 days after ingestion showed minimal change, at which point patient was started on clear liquids by mouth (Fig. 3D-F). She was advanced to a soft mechanical diet by hospital day (HD) #10. A third MRI on HD #11 showed mild improvement of edema (Fig. 3, G-I). She was discharged on HD #12 with instruction to continue soft diet at

home and strict return precautions.

Twenty-five days following battery ingestion, the patient developed acute hematemesis at home which the parents described as "a gallon" of emesis within 5 minutes. An emergency medical transport helicopter flew her approximately 50 miles to our hospital. In the ED, she was tachycardic with decreased systolic blood pressure, pallor, and weak distal pulses. No active hematemesis was noted, however, hemoglobin was 5.1g/dL and the patient was "covered in dried blood." Massive transfusion protocol was activated and she underwent emergent cardiac catheterization.

During cardiac catheterization, angiography demonstrated outpouching of the anterior descending aorta consistent with intimal injury, representing an aorto-esophageal fistula (Fig. 4, A-C). A discrete nodular aortic irregularity was noted around T10. At this region, the aorta measured 8.4–8.9 mm in diameter. A 12 mm imes 34 mm 8 zig premounted Covered Cheatham Platinum (CP) stent was advanced through a 12 French (Fr) long sheath in the right femoral artery. The stent was carefully centered to treat the area of intimal injury. It was deployed in the usual fashion (outer balloon inflation pressure 8 atm). Afterwards, angiography demonstrated excellent stent position and sealing of the irregularity in the aortic wall. The stent was larger than the adjacent aortic caliber, suggesting stable stent position. The patient was transferred to the cardiac ICU for further care. She received 37mL/kg packed red blood cells (pRBC's) and 9.5mL/kg fresh frozen plasma (FFP) in the first 24 hours, followed by 10mL/kg pRBC on HD#2. She did not require further blood products after this, and hemoglobin stabilized to 11.5 g/ dL. She was hospitalized for twelve days: received exclusive parenteral nutrition for 6 days, was transitioned to NG feeding for 6 days, then advanced to a soft oral diet once esophagram demonstrated no leak. She received empiric meropenem, vancomycin, and fluconazole during hospitalization and was discharged on omeprazole and prophylactic amoxicillin-clavulanic acid.

It has been three months since hospital discharge. Outpatient esophagram was performed which demonstrated no esophageal leak, and computed tomography angiography (CTA) showed stable stent position with no evidence of persistent fistula (Fig. 5A and B). She is tolerating a regular diet and has stopped her antibiotics and proton-pump inhibitor. The Covered CP stent can be expanded over time as this patient grows. She will be monitored yearly to assess stent and aortic size to determine future interventions such as stent expansion or aortic replacement.

3. Discussion and conclusions

The National Capital Poison Center (NCPC) database reports a total 64 deaths in children following button battery ingestion worldwide since 1977; 61% (39/64) of which were due to documented arterioesophageal fistulae. BB impaction in the mid-esophagus raises concern

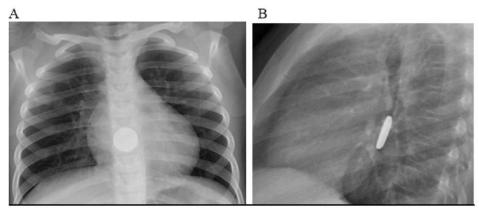


Fig. 1. A-B: Initial Chest X-ray demonstrates approximately 21mm button battery in the mid esophagus (T7-8) with negative pole oriented posterior on lateral view.

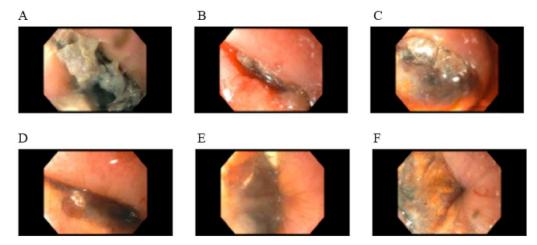


Fig. 2. A-F (color image): Endoscopic images of impacted button battery demonstrating corrosion of the battery and bleeding (A–C) and injury after removal showing erythema and ulceration (D–F). (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

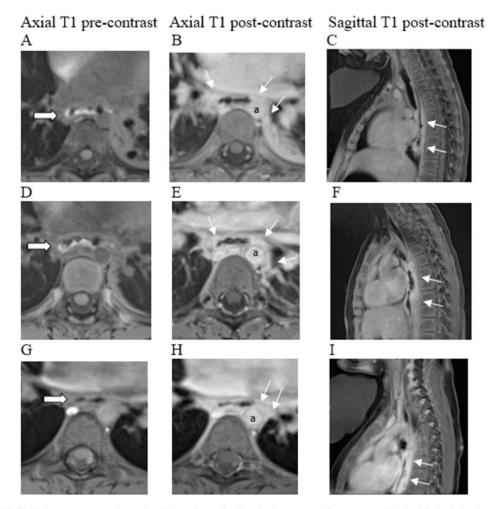


Fig. 3. A-I: Initial MRI (A, B, C) demonstrates esophageal wall irregularity (=>) and enhancement of posterior mediastinal fat (\rightarrow) abutting and surrounding thoracic aorta (a); Second MRI (D, E, F) demonstrates little change; Esophagus (=>) is improved on Third MRI (G, H, I), however enhancement abutting thoracic aorta (a) is minimally changed.

for arterio-esophageal fistulae due to close anatomic proximity. However, NCPC data reports arterio-esophageal fistulae associated with batteries at all levels within the esophagus, including fistulae with the aorta, the subclavian, carotid, and thyroid arteries [2]. Previously documented cases of survival of an AEF following BB ingestion reported

open surgery while this case highlights minimally invasive stent placement [3-6].

Previous data suggests that serious complications are seen more often in children less than 5 years old, batteries greater than 20 mm, and unwitnessed ingestions [7]. Our patient was 6 years old, the battery was

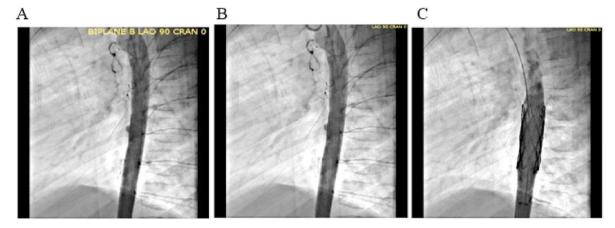


Fig. 4. A-C: Imaging from the catheterization lab showing outpouching of the anterior descending aorta consistent with intimal injury (A,B) and repair with stent placement(C).

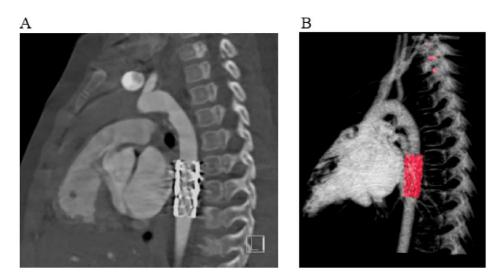


Fig. 5. A-B: Sagittal reformat (A) and 3D rendering (B) from CTA performed 2 months following aortic stenting demonstrates stable position of stent in descending thoracic aorta. No contrast leak seen, however evaluation of prior intimal aortic injury is limited due to metallic streak artifact.

21 mm and her ingestion was witnessed with the family promptly seeking medical care.

This case highlights several key issues in clinical management following esophageal BB impaction. Current North American Society of Pediatric Gastroenterology, Hepatology, and Nutrition (NASPGHAN) guidelines stress prompt removal of esophageal button battery within 2 hours of ingestion [1]. In addition to location within the esophagus, length of time prior to battery removal, and orientation of negative battery pole are likely the most important factors contributing to development of AEF [8]. Our patient's battery removal was significantly delayed due to the patient presenting to an outside hospital necessitating transfer. In a larger retrospective review of esophageal BB ingestion at our institution, this is not uncommon with >85% of patients requiring transfer to tertiary care pediatric hospital for removal [9]. Administration of honey or sucralfate at time of initial recognition is a possible intervention to mitigate degree of mucosal injury prior to battery removal [10]. This was not performed in our case, although literature on these interventions is sparse and it is unclear if they would have significantly altered any outcome.

Following battery removal, NASPGHAN guidelines recommend MRI every 5–7 days until "injury recedes from the aorta," with expected significant risk of AEF suggested to end at 21 days [1]. Unfortunately, these guidelines are not based on outcomes evidence, as they were published before serial MRIs were routinely performed for this injury.

Recent data in the radiology literature suggests that serial MRIs may not predict the development of severe complications [11]. In this case, a total of 3 MRIs were performed at day 1, 5, and 11 following battery removal documenting continued improvement mediastinal injury. Thus, this case further raises questions regarding the utility of serial MRI imaging.

Current practice recommends an esophagram to rule out esophageal perforation before oral feeding [1]. However, early esophagram in the setting of extensive mediastinal edema may provide false reassurance because edema may prevent a perforation from being visualized. Furthermore, fistulas and strictures can develop in children without a perforation as they are the result of continued tissue damage during the prolonged esophageal healing process following injury.

Missing from current guidelines are recommendations on feeding practices following BB ingestion. The healing process may be affected by nutrition type, timing, and mode of delivery. It is possible that allowing soft foods before significant improvement on MRI caused stress which disrupted esophageal healing. Prolonged tube feeds or oral liquid diet may further minimize risk. It is unclear if the benefits of enteral nutrition for gut tissue healing in conditions like Crohn's disease are applicable to these injuries [12].

This case is also unique in the use of life-saving aortic stent placement for AEF. Endovascular stent intervention has replaced open surgical repair in the setting of blunt thoracic injury due to shorter

operative time and improved outcomes [13,14]. Stent placement has similar advantages in cases such as ours, however a multidisciplinary approach to procedure planning is necessary with availability of surgical support for open repair if necessary.

Previously, 21 days was cited as the period of risk for developing an AEF [1]. This patient fistulized 25 days following ingestion, making the case unique in its rarity, length of time to development, and outcome. The patient developed a severe complication despite 12 days of conservative inpatient management and serial MRIs. This case highlights a successful multidisciplinary treatment approach for AEF with prompt aortic stent placement.

This case also underscores the need for further research and advocacy regarding BB ingestions. While this is a chronic concern in pediatric gastroenterology, the prolonged periods of homebound children like ours during COVID-19 has made it even more pertinent now as early data shows an increase in BB poison center calls in 2020 [15].

This report describes development of an AEF outside the previously documented risk period, which developed despite close monitoring and adherence to national guidelines. Several gaps in the literature and guidelines especially in regards to safe feeding in this population were revealed. Importantly, this case also resulted in a positive outcome due to multidisciplinary efforts in recognizing and treating a life-threatening complication of BBI with minimally invasive surgery.

Ethics approval and consent to participate

The need for approval for case reports is waived at our institution.

Consent for publication

The patient's mother has provided written consent to submit this case for publication and a copy is available upon request.

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Authors' contributions

Dr. Sinclair composed the bulk of the introduction and discussion drafted the initial manuscript, and reviewed and revised the manuscript.

Drs Stevens and McElhanon drafted the case report, composed parts of the discussion, and reviewed and revised the manuscript.

Drs Santore and Chahine provided surgical perspective of the hospital course and discussion and reviewed and revised the manuscript.

Dr. Riedesel provided radiological images and descriptions and

reviewed and revised the manuscript.

All authors attest that they meet the current ICMJE criteria for Authorship.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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