



# Surgical treatment of massive bleeding of a right aberrant subclavian artery after oesophageal stent removal



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## ABSTRACT

We report a case of a 9-year-old female who required surgical treatment and resuscitation after severe trans-oesophageal bleeding of a right aberrant subclavian artery (RASA). Bleeding of this RASA was caused by a mechanical irritation due to an oesophageal stent. The stent was placed weeks before to dilate the oesophagus after accidental ingestion of a caustic agent. Although conservative management of benign oesophageal stenosis in children is highly recommended, there are still some major complications to be considered. To avoid erosion of aberrant subclavian artery vascular rings and slings, as described in several case reports, these vessels should be excluded by computed tomography (CT) or magnetic resonance imaging (MRI) scans before placement of oesophageal stents. This case suggests that management of caustic ingestion in children is still a major challenge in paediatric surgical departments.

## 1. Introduction

The most often cause of oesophageal stenosis in children remains caustic ingestion [1]. Significant evidence suggests that endoscopic management of these erosive strictures exhibits the best prognosis [1]. Conservative options, such as balloon dilatation, bougies and stenting, have improved the outcome of patients with benign oesophageal strictures in the past. However, these procedures are complex, and several complications should be considered. In this case, we report on severe bleeding of a right aberrant subclavian artery (RASA) after oesophageal stent removal.

## 2. Case report

An 9-year old girl was admitted to the emergency department of a nearby hospital after accidental ingestion of a caustic agent (barbecue cleaner). The patient was haemodynamically stable, and supportive care was admitted. In the endoscopy, an oesophageal and gastric alkali burn grade 2 was diagnosed (Fig. 1).

To prevent a stricture formation, oesophageal dilatation was performed with Savary bougies at intervals of 7–10 days during the next month. A perforation of the oesophagus resulting in mediastinal emphysema was successfully managed with drainage and antibiotics. Due to failed dilation therapy, an oesophageal stent was placed after 7 months. Antibiotic prophylaxis and cortisone were administered for ten days. Endoscopic control of the stent position was performed one month

later. During the next endoscopy, the position of the stent was corrected with no complications.

One month later, the now 9-year-old girl presented with acute upper gastrointestinal bleeding in the emergency-department of a nearby hospital. After primary stabilization, a pharyngeal blood clot was aspirated. The patient was transferred to the intensive care unit of our hospital where endotracheal intubation was indicated, and blood transfusion was administered due to relevant blood loss. The stent was removed endoscopically, and the bleeding intensified. The patient experienced cardiorespiratory arrest. During the continuous resuscitation, laparotomy and gastrotomy were performed, but the origin of the bleeding could not be identified. After sternotomy, open cardiac massage was performed, and haemodynamic stabilization was achieved. Further exploration and extirpation of the oesophagus revealed the eroded RASA as the bleeding source. Reconstruction of the artery was performed, in the course the RASA stenosed again. Bleeding stopped after ligation of the RASA, and a suction tube was inserted into the upper oesophageal stump.

Postoperative neurologic symptoms, such as eye deviation to the right for some seconds and weakness of the left hand, served as indications for cranial computed tomography (CT) and cranial magnetic resonance imaging (MRI). Several cerebral infarctions were detected. CT scan also revealed that RASA was interrupted (Fig. 2), thus explaining the retrograde filling of the arteria vertebralis dexter and arteria mammaria dexter in terms of a subclavian steal syndrome. This retrograde filling probably caused insufficient perfusion of the cortical

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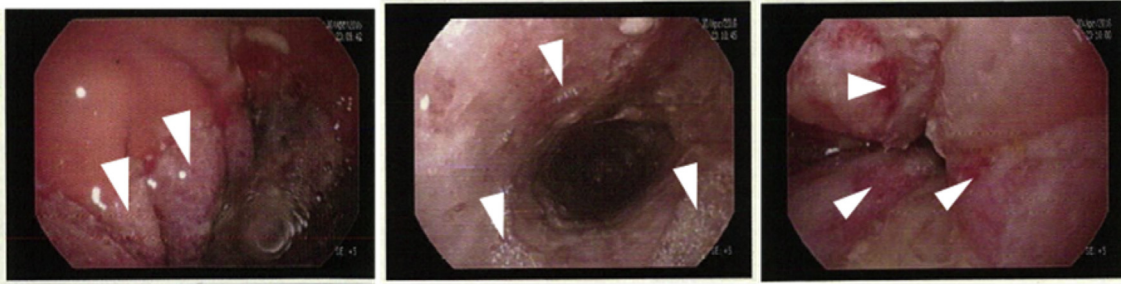


Fig. 1. Endoscopy: alkali burn grade II of the oesophagus and the gastric body.



Fig. 2. CT axial plane at the level of the aneurysmal artery lusoria course in the superior mediastinum.

terminal vessels. Surgical intervention was not performed given that the force of the left hand and neurological symptoms improved.

Two months after haemodynamic shock, gastric pull-through was performed with no complications. On the eighth day after operation, transanastomal feeding was initiated and well tolerated. With the exception of weakness of the left arm and hoarseness, no further neurological symptoms were present. Oral feeding was introduced and well tolerated with minimal signs of dumping. The hoarseness was examined by an otorhinolaryngologist, and hypomobility of the vocal chords was diagnosed. Logo- and ergotherapy ameliorated this condition. The weakness of the left hand was improving. The young girl was dismissed in a good general state of health after an 81-day stay in the hospital.

### 3. Discussion

Accidental ingestion of caustic agents occurs rarely in young children in Western countries. Subsequent oesophageal strictures are the most frequent cause of oesophageal stenosis in children [1]. The acute and long-term management of benign oesophageal strictures is complex and represents a serious problem in paediatric departments.

For the primary management of benign oesophageal stenosis in children, dilatation with balloon-dilators or bougies is recommended according to the European Society of Gastrointestinal Endoscopy guidelines [2]. Typical complications of the balloon dilators include restenosis, balloon leakage, sputum retention and displacement [3]. Semi-rigid dilators require high operator expertise to avoid oesophageal

perforation [1]. Decision for temporarily endoscopic stenting in the case of refractory long segment stenosis, such as that noted in this case, remains a subject of controversial debate. There is a weak recommendation and moderate evidence given the low therapeutic effort and the high incidence of complications. In adults, the overall clinical success rate of stenting is only 40.5%. The adverse event rate is 20.6% with no significant difference between the different types of stents. In addition, strictures longer than 2 cm, such as that in our case, exhibit significantly reduced success rates of stenting [2]. Complications, such as migration of the stent [4,5], oesophageal perforation [6] and death after massive bleeding, have been observed [2].

According to the small diameter and length of the oesophagus of children, commercially available stents are relatively useless in the management of oesophageal stenosis. Custom-made stents have been used in the past and in this case [7]. If the decision for temporarily endoscopic stenting is made, fully covered self-expandable metallic stents or self-expandable plastic stents should be used given the difficulty of the removal of uncovered metal stents according to guidelines [2].

Although the complication rate is high, temporary stenting was performed in this case, and a dramatic development occurred. In this case, massive thoracic bleeding was induced by erosion of an aberrant right subclavian artery via stent migration. Similar cases have been previously reported in the literature [7,8].

An aberrant right subclavian artery can be identified in 0.1–3.0% in the population and is the most common vascular anomaly of the aortic arch [9,10]. Barium oesophagogram can provide an indication, but a definitive diagnosis must be confirmed with CT or MRI [11]. As previously described [1,7], we also recommend to exclude these vascular rings or slings with CT or MRI before oesophageal stents are implanted to avoid severe complications similar to our report. If vascular anomalies are diagnosed oesophageal stenting is contraindicated, and primary oesophageal replacement should be considered.

### 4. Conclusions

Oesophageal stenting should not be used for oesophageal stenosis after caustic ingestion in children with arteria lusoria or other vascular abnormalities. We highly recommend evaluating vascular rings and slings before oesophageal stents are placed. Barium contrast studies can indicate dorsal oesophageal stenosis based on aberrant vessels of the aortic arch, but CT or MRI is necessary to exclude these abnormalities before stenting. Although the prognosis of the young patient was poor from the beginning, several complications could be managed, and the present result is auspicious.

### 5. Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

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## Authorship

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

## Conflict of interest

The following authors have no financial disclosures: (M.R., J.H., D. v.S.).

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