Aortoesophageal fistula as a complication of Montgomery salivary bypass tube

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Abstract The authors describe a 2-year-old girl with a right-sided aortic arch who developed a sudden, fatal aortoesophageal fistula after prolonged placement of Montgomery salivary bypass tubes in the distal esophagus.

Aortoesophageal fistula in children may result from congenital vascular rings, ingestion of a foreign body, penetrating trauma, or iatrogenically induced injury, such as a complication of surgical instrumentation (eg, rigid esophagoscopy) [1-10]. In adults, prolonged placement of a nasogastric tube may cause aortoesophageal fistula [11]. To our knowledge, there have been no reports of erosion mediated by a Montgomery salivary bypass tube (MSBT) into the aorta [1,2].

The MSBT is a variably sized, medical-grade silicone tube with a funnel-shaped superior end that facilitates the collection of saliva and food and helps maintain proper esophageal positioning. Common indications for MSBT placement include (1) esophageal fistulas or stenoses, usually because of local tumor growth; (2) tracheoesophageal fistulas; (3) esophageal strictures after extensive surgery and/or radiation therapy, or after ingestion of caustic material; and (4) pharyngocutaneous fistulas after laryngectomy. The diameter of the tube is maximized to assure stable positioning. Although the MSBT was initially designed to sit above the upper esophageal sphincter, some groups have successfully placed the tube entirely in the esophagus [12,13].

1. Case report

The patient was a 2-year-old girl with tetralogy of Fallot, dextrocardia, hypoplastic right lung, and tracheoesophageal fistula with duodenal and esophageal atresia. A surgical repair in the neonatal period was performed by ligation of the tracheoesophageal fistula and primary repair of the esophagus. Her esophagus remained stenotic postoperatively. She was dependent on a gastrostomy feeding tube throughout life and underwent a Nissen fundoplication for gastroesophageal reflux. A partial esophageal resection with re-anastomosis...
was performed 8 months premortem to remove the stenotic area. However, the stenosis recurred, with repeated dilations required as often as every 2 weeks. Further surgical intervention was considered but a more conservative approach was attempted first. She had a soft MSBT placed in the distal esophagus 4 months antemortem, which was obstructed by foreign bodies multiple times and subsequently migrated prograde. Proximal re-stenosis necessitated the placement of a second MSBT 7 weeks antemortem, located end-to-end proximal to the original MSBT. Her most recent intervention was 1 week antemortem for removal of a chicken bone from the proximal MSBT. She presented on the day of demise with 1-week of abdominal pain and having pulled out her gastrostomy feeding tube. Hematemesis was observed and the patient had a 10.1 mg/dL hemoglobin concentration. There was no melena. She underwent esophagoscopy, which revealed blood clots in the esophagus and stomach, but no active source of bleeding. Both MSBTs were in place. Shortly thereafter she experienced a massive upper gastrointestinal tract hemorrhage. She died in the operating room despite aggressive resuscitative measures including thoracotomy. Autopsy revealed deep circumferential erosions of the esophagus at the level of the MSBTs (Fig. 1A). The superior edges of the funnel-shaped tubes were deeply embedded in the esophagus, adjacent to the distal MSBT (Fig. 1B). The aorta, esophagus, and heart were all surrounded by dense postsurgical adhesions.

2. Discussion

Aortoesophageal fistula has many causes; however, the association of a silicone MSBT with a fatal event has not been previously described. The mechanism of injury in this case appears to be pressure necrosis from the MSBT. The presence of dense fibrous tissue fusing the aorta and esophagus together because of multiple prior surgeries may have eliminated the normal ability of the esophagus to expand in response to the MSBT and may have prevented the aorta from being displaced rather than eroded by the MSBT. The right-sided aortic arch also placed the aorta in jeopardy relative to the esophagus. A history of repeated foreign bodies lodged in the stents raises the question of a possible contribution from this source. Esophageal strictures after esophageal atresia repair is typically managed by repeated dilatation. For those who do not respond to dilatation, procedures to resect the stricture or to replace the esophagus may be considered. The risks of reoperation in such patients, particularly those with comorbid conditions such as tetralogy of Fallot, is high, but the risks of aortoesophageal fistula associated with the placement of thin-walled silicone-based tubular stents should be considered as well.

Pressure necrosis from long-term instrumentation as a mechanism for the development of an aortoesophageal fistula has been reported. The most common cause is the presence of a nasogastric tube for prolonged periods. Nasogastric tube erosion into the aorta has been reported in patients with vascular rings [3]. A nasogastric tube caused an aortoesophageal fistula by eroding into the right-sided aorta of a 13-year-old adolescent girl after 8 days [9]. A nasogastric tube has also been implicated in one adult case where no other abnormality was identified [11]. Unlike the nasogastric tube, however, the MSBT has previously been thought to be pliable enough to avoid pressure necrosis injuries. In the case we report here, the medical-grade silicone of the MSBT was sufficient to cause an aortoesophageal fistula. The right-sided aortic arch, combined with extensive postsurgical fibrosis, appears to be contributing factors in this case.

References


