Case Report

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# Self-bougienage of oesophageal stricture by an 8-year-old child

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## ABSTRACT

Oesophageal corrosive injuries have a high potential to result in stricture formation with the requirement for repeat oesophageal dilation. Especially in children, oesophageal bougienage is performed under general anaesthesia or strong sedation. In developing countries without comprehensive medical care, this service might not be available. We report the case of daily oesophageal self-bougienage performed by an 8-year-old Afghan child as highly effective treatment of recurrent oesophageal stricture formation after caustic substance ingestion.

**Key words:** Children, oesophageal stricture, self-bougienage

## INTRODUCTION

Paediatric patients with caustic substance ingestion require immediate diagnosis and treatment. Upper gastrointestinal endoscopy is the gold-standard procedure to assess the severity of injury. An appropriate treatment regime is based on these findings.[1] Criteria for endoscopy are pH value, ingested volume, presence of oral burn and/or presence of symptoms like dysphagia, haematemesis, respiratory distress or stridor.[2] Early treatment may include acid suppression by proton pump inhibitors, i.v. antibiotics, high-dose corticosteroids and nutritional support.[2] In due course, oesophageal dilation in case of stricture formation might be necessary. Oesophageal narrowing is a catastrophic complication with an incidence of 5% to 47%, depending on the severity of tissue injury[3,4] and the time and type of initial treatment. Stenosis is frequently accompanied by progressive malnutrition with need for gastrostomy tube feeding. In developing countries without a nationwide functioning healthcare system, early admission and immediate treatment of children with oesophageal lesions caused by chemical burn is frequently not provided. This delay results in a high rate of recurrent strictures of more than 70% compared with 30% in patients with early treatment.[5]

We present the case of a severe malnourished Afghan child with recurrent high-grade stenosis of the upper oesophagus due to ingestion of battery acid. He first underwent endoscopically visualised wire-guided oesophageal dilation and later could be taught daily self-bougienage. He fully recovered from malnutrition with compete relief of dysphagia.

## CASE REPORT

An 8-year-old boy from Afghanistan presented with severe malnutrition due to dysphagia caused by a high-grade oesophageal stricture. The stricture resulted from ingestion of battery acid. The boy was referred to us by the non-governmental organization (NGO) “Pease Village International” 5 months after the injury. The medical history was incomplete. In Afghanistan, several oesophageal dilations and a gastrostomy feeding tube insertion had been performed. At the time of admission to our hospital in Germany, the gastrostomy feeding tube was blocked. Enteral nutrition was provided only by a nasogastric feeding tube. His body mass index was 11 (weight: 18 kg, height: 128 cm). Further clinical examination and abdominal ultrasonography revealed no pathological result. The initial blood test showed minimal elevated liver enzymes (alanine transaminase, 2.6 μkatal/l; aspartate transaminase, 2.0 μkatal/l) which became normal within 2 weeks.

The blocked gastrostomy feeding tube was changed and imaging of the stomach and upper part of the small bowel with contrast medium given through the gastrostomy tube was performed. It showed an atypical stomach configuration with narrowing of the corpus and distortion of the antrum caused by the gastrostomy

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and/or scar formation due to the initial acid ingestion. An additional barium swallow showed a proximal oesophageal high-grade stenosis over a length of 7 cm with a 2 mm lumen at the level of the aortic arch and a 1.7-cm poststenotic dilation [Figure 1].

The gastrostomy feeding tube was switched to a skin level feeding device and a dietary management of disease-related malnutrition with 1.5 kcal/ml tube feed was started. On day 9 after admission, the first flexible upper gastrointestinal endoscopy was performed under general anaesthesia. The stenosis was located 15 cm distal to incisor teeth with a point-shaped opening. First, the stricture was cannulated with a guide wire and correct positioning was confirmed by intraoperative X-ray. The endoscope was removed and Savary-Gilliard dilators were introduced over the guide wire in sequentially increasing sizes starting with 5 mm. The advisability of proceeding to a larger bougie was judged by the amount of resistance. In this first bouginage, the oesophageal stricture was dilated up to 9 mm. The procedure was repeated weekly. Parallel to these dilations, oral feeding was started with fluids and soft food. After 5 initial bouginages, the oesophageal narrowing was dilated up to 13 mm and the patient returned to solid food. The gastrostomy button was removed and the patient was discharged to the NGO based in Germany.

Four weeks later, the patient was readmitted with recurrence of dysphagia. Endoscopy showed a recurrent oesophageal stricture. The narrowing was dilated up to 15 mm using Savary-Gilliard bougies. Histology revealed fibrosis and minimal inflammation without dysplasia or malignancy. Within the next months, this procedure was repeated another 18 times. Immediately after oesophageal dilation, the patient was able to tolerate solid food but soon he deteriorated and represented with dysphagia and weight loss.



**Figure 1: Barium swallow at first day of admission showing a high-grade oesophageal stenosis over a length of 7 cm with a 2-mm lumen at the level of the aortic arch**

The resistance to therapy of this oesophageal stenosis and the special circumstances of this case with no chance of a long-term follow-up in a specialised paediatric centre in his home country directed us to the possibility of self-bouginage. This was the first paediatric patient in our institution considered to perform oesophageal dilation by himself and therefore permission by our institutional ethical committee was obtained before we started with the new treatment protocol. Informed consent was given by the person having care and custody of the child after detailed explanation of complications such as perforation and recurrence. We used flexible Rüsch oesophageal bougies size 7 to 11 mm with an olive tip. The procedure was performed under topical pharyngeal anaesthesia with Xylocaine spray. The fasting child was sitting on a stretcher with reclined head. The tip of the bougie was inserted into the pharynx, where the gag reflex opened the upper oesophageal sphincter and allowed insertion of the bougie into the oesophagus. When the resistance of the stricture was encountered, the bougie was carefully forced through it, hold for a few seconds and removed. The procedure was daily performed on the ward. Within 2 months, the dilator size was sequentially increased up to 11 mm. Initially, the bouginage was only performed by the surgeon but step by step, the patient was more involved in the procedure. First, he was bringing the tip of the dilator to his pharynx and after insertion of the bougie by the surgeon, he removed it by himself. Later, the patient was able to perform the whole procedure by himself. He did not even use the Xylocaine spray anymore.

The patient was finally discharged home to his family in Kabul after one year of being in Germany. He is independently performing the oesophageal self-bouginage every day. There is no food restriction, no dysphagia and therefore a satisfying weight increase of 9 kg compared to first admission. In Kabul, the NGO has organised regular physical examinations of the patient by a local doctor. This follow-up should be continued as long as oesophageal dilations are necessary. In case of deterioration, there would be the possibility of readmission to our hospital.

## DISCUSSION

This is the first case report of successful daily self-bouginage in an 8-year-old child with oesophageal narrowing due to caustic substance ingestion. Repeated oesophageal dilation has been shown to be an effective therapy for oesophageal strictures with a success rate of

75%.[6] Up to 36 oesophageal dilations per patient over a time period of 34 months have been reported.[7] In our patient, 23 bouginages using Savary-Gilliard dilators have been performed under general anaesthesia over a period of 12 months. Control endoscopy performed directly after dilation up to 15 mm frequently revealed minor mucosal bleeding. There is evidence that this recurrent mucosal trauma results in increasing scar formation and therefore in recurrent narrowing of the oesophagus.[8] Our idea was to perform a minimal oesophageal bouginage first up to 7 mm on a daily base to prevent mucosal injury and to provide enough lumen for toleration of solid food. Daily general anaesthesia or even daily sedation on the ward to perform the oesophageal dilation was no option. Oesophageal stricture dilation in children without sedation had been successfully performed, but the procedure was done in an outpatient department by healthcare professionals.[9] Again, this was no option for our patient, because continuous long-term dilation programs are impossible in his home country. In addition, repeated healthcare utilisation in developing countries is expensive, might result in loss of work for parents and neglect of other children at home.[10] One reasonable treatment option for our patient was to learn to dilate the oesophagus narrowing by self-bouginage, a procedure which is more than difficult even for adults and seemed to be unimaginable for an 8-year-old child. There are only few studies available investigating and advocating self-bouginage as long-term therapy for oesophageal strictures. The youngest patient mentioned in these studies was 15 years old.[11,12]

In our case, every effort was done to maintain the native oesophageal patency. However, if self-bouginage would have failed, an oesophageal replacement either by colon replacement or gastric pull-up would have been the next treatment option. Oesophageal replacement is associated with multiple postoperative complications like anastomotic stricture, late graft stenosis, redundant intrathoracic colon with stasis and cologastric reflux,[6] and therefore long-term follow-up in a specialised paediatric unit is necessary to identify and to treat these complications.

In conclusion, we have been shown that even in the early childhood, self-bouginage of an oesophageal stricture is possible. It might be an option to provide a high quality of life in developing countries with absence of specialised healthcare.

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