Case report

Non-operative treatment for perforated gastro-duodenal peptic ulcer in Duchenne Muscular Dystrophy: a case report

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Abstract

Background: Clinical characteristics and complications of Duchenne muscular dystrophy caused by skeletal and cardiac muscle degeneration are well known. Gastro-intestinal involvement has also been recognised in these patients. However an acute perforated gastro-duodenal peptic ulcer has not been documented up to now.

Case presentation: A 26-year-old male with Duchenne muscular dystrophy with a clinical and radiographic diagnosis of acute perforated gastro-duodenal peptic ulcer is treated non-operatively with naso-gastric suction and intravenous medication. Gastrointestinal involvement in Duchenne muscular dystrophy and therapeutic considerations in a high risk patient are discussed.

Conclusion: Non-surgical treatment for perforated gastro-duodenal peptic ulcer should be considered in high risk patients, as is the case in patients with Duchenne muscular dystrophy. Patients must be carefully observed and operated on if non-operative treatment is unsuccessful.

Background

Duchenne muscular dystrophy (DMD) is a fatal X-linked recessive disease. It is the most common congenital neuromuscular disorder of childhood [1]. It's clinical course and characteristics caused by skeletal and cardiac muscle degeneration are well known and documented [2]. Progressive muscle weakness inevitably results in the complete inability to walk around the age of 11 [3]. By the age of 18 patients are predisposed to serious pulmonary infections [4]. It leads to death before the age of 30. Gastrointestinal involvement is less well known. Dystrophic changes in smooth muscle of the gastro-intestinal tract have been implicated in causing gastro-intestinal dysfunction [5]. We want to present and discuss a patient with Duchenne muscular dystrophy with a clinical and radiographic diagnosis of acute perforated peptic ulcer. Treatment options for an acute perforated peptic ulcer should be well considered in a patient at pulmonary and cardiac risk caused by DMD.

Case presentation

A 26-year-old male has DMD and is non-ambulatory. He was presented on our emergency department because of severe sudden onset abdominal pain. Upon presentation there was sharp constant pain located in the epigastrium, with nausea and vomiting. Complaints had been present...
Figure 1
Sitting chest radiograph showing free air in the right subphrenium
for several hours. There were no signs and symptoms of gastrointestinal bleeding. On physical examination there was direct tenderness in his abdomen with rebound tenderness and guarding. Bowel sounds were absent. His body temperature was 38.3 °C, there was a tachycardia at a rate of 150/min and blood pressure was 115/60 mmHg. A sitting chest radiograph showed free air in the peritoneum (figure 1).

Previous medical history was without respiratory aid and no signs of nightly hypoxia, but with recurring episodes of pneumonia. His functional vital capacity was 30% of the predicted value. Two years ago he had two episodes of acute gastric bleeding from peptic ulcer vessels; both diagnosed endoscopically and subsequently treated with endoscopic epinephrine injections with bipolar coaptation. Thereafter Helicobacter Pylori (HP) eradication therapy (omeprazole, metronidazole and clarithromycin for 7 days) was given, followed by a continuous use of omeprazol and sucralfate. Furthermore he was known to have a chronically dilated stomach, with recurring episodes of poor gastric passage and vomiting.

Based on his previous medical history and signs and symptoms on current presentation a clinical diagnosis of perforated peptic ulcer was established. The associated perioperative risks of pulmonary dysfunction and cardiac failure in this patient were high. Therefore non-operative treatment for perforated peptic ulcer was started. The patient was treated with continuous naso-gastric suction (20 cm H2O) and intravenous omeprazol continuously and regular doses of intravenous broad-spectrum antibiotics aimed at gram positive, gram negative and anaerobic bacteria (Cefuroxim and Metronidazole) were administered. The patient's symptoms rapidly improved thereafter. After a five-day period the naso-gastric suction tube was removed. The pain and nausea had subsided; there was less direct tenderness in the abdomen and no rebound tenderness or guarding. Bowel sounds were present. Body temperature was normal. Oral intake of fluids and solids was started. Twelve days after admission the patient was dismissed from hospital in good clinical condition.

**Discussion**

DMD is caused by a deletion in the dystrophin gene located on the short arm of the X-chromosome [6]. The resulting lack of dystrophin causes degeneration of striated muscle [7]. Respiratory dysfunction caused by dystrophic changes in respiratory muscle causing pulmonary infection is the most common complication in DMD [8,9]. Dystrophin is also present in smooth muscle. The involvement of smooth muscle in addition to the progressive dystrophic changes in striated muscle may cause clinical dysfunction of the gastro-intestinal tract [10-12]. Both gastric dilatation and intestinal pseudo-obstruction have been described as gastro-intestinal complications in DMD [13,14]. To provide evidence that smooth muscle is functionally impaired in patients with DMD, Barohn et al. compared gastric-emptying time between DMD patients and healthy control subjects. They showed a significant difference in gastric emptying time in patients with DMD. Furthermore, on examination at autopsy degeneration and loss of smooth gastro-intestinal muscle similar to changes in skeletal and cardiac muscle were found in DMD patients [15,16]. To our knowledge however there are no reports on gastro-duodenal (perforated) peptic ulcer as a complication in DMD.

The common treatment for patients with perforated peptic ulcer is surgical repair [17]. In most cases an omental patch repair is performed, followed by treatment with H2 receptor antagonists and if indicated HP eradication therapy [18-20]. Non-operative treatment for perforated peptic ulcer can be successful, as first reported by Taylor in 1957. He compared operative and non-operative treatment for perforated peptic ulcer and reported similar mortality rates in both groups (5%) [21]. Crofts et al. compared the outcome of non-operative treatment with that of emergency surgery in a randomised trial and reported an overall mortality rate of 5% in each group and no significant difference in morbidity [22]. Crofts et al. conclude that an initial period of non-operative treatment is safely allowed except in patients over 70 years old [23]. Kaene et al. reported favourable survival rates (2.4% mortality) in a series of 42 patients treated non-operatively compared with reported surgical mortality [24]. The importance of careful clinical observation by an experienced surgeon is emphasised in reported literature, because non-operative treatment should be promptly abandoned if unsuccessful [21,25,26].

In spite of those reports non-operative treatment has never really been accepted as standard treatment [17,27]. However in selected cases non-operative treatment for perforated peptic ulcer may be considered. Boey et al. identified major medical illness, preoperative shock and longstanding perforation, as perioperative risk factors [28]. Based on these risk factors they correctly predicted the outcome in 93.8% of patients in a series of 259 patients treated operatively for perforated peptic ulcer. Mortality rate increased with the presence of one, two or all three risk factors. Boey et al. conclude that non-operative treatment should be re-evaluated for patients with all three risk factors because of their uniformly dismal outcome after surgery [29]. Patients with DMD are at high risk for perioperative respiratory dysfunction or heart failure [30-32]. Therefore in this case a non-operative approach was well considered and proved to be successful.
Conclusion
This case demonstrates a gastro-duodenal peptic ulcer as a possible complication in DMD. Furthermore it illustrates that in pulmonary and cardiac high risk patients non-operative treatment for perforated peptic ulcer can be successful. If treated non-operatively patients should be carefully observed and operated on if non-operative treatment is unsuccessful.

Competing interests
None declared.

Abbreviations
DMD: Duchenne muscular dystrophy
HP: Helicobacter Pylori

Authors contributions
Brinkman J-M: rewrote the manuscript in 2003.
Van Royen BJ: helped rewriting the manuscript in 2003.
Brinkman J-M and Oddens J: drafted the manuscript in 2001.
Weyer J and Olsman JG: read and approved the manuscript in 2001 and 2003.

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