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Neonatal oesophageal perforation: The role for non-operative management

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## ABSTRACT

**Aim:** Isolated oesophageal perforation in neonates is a rare but potentially life-threatening condition. Although management has historically been operative, conservative management (antibiotics, bowel rest, parenteral nutrition) is now more routinely used. The aim of this study was to evaluate the management of this condition in two large neonatal surgical centres.

**Methods:** A retrospective cohort study was conducted for neonates admitted to the Hospital for Sick Children (Toronto, Canada) or The Royal Children's Hospital (Melbourne, Australia) with a diagnosis of oesophageal perforation, from 2006-2016. Patients with oesophageal atresia or tracheo-oesophageal fistula were excluded. Data were collected from chart review regarding demographics, clinical course, management and outcomes.

**Results:** Eleven neonates with oesophageal perforation were identified over a 10-year period at the two centres. Median gestational age at birth was 25.3 weeks (interquartile range {IQR} 24.2 – 28.8) and the majority (7/11, 64%) of neonates were extremely low birth weight. Diagnosis was made on day 1 of life for 9/11 (81%) neonates and was secondary to nasogastric tube insertion in 10/11 (91%) neonates. Only 4 (36%) neonates had symptomatic complications. All neonates were managed with bowel rest and intravenous antibiotics for a median of 7 (IQR 7 – 10) days; 2 patients required operative intervention. Three neonates (27%) developed chronic lung disease and two (19%) died prior to discharge.

**Conclusions:** Oesophageal perforation is severe complication secondary to instrumentation of the upper gastrointestinal tract in neonates. Prompt and accurate diagnosis is crucial. Non-operative management is effective for the majority, though morbidity is common.

## KEY WORDS

- Neonate
- Oesophageal perforation

## WHAT IS ALREADY KNOWN ON THIS TOPIC

- Oesophageal perforation is a known complication of oropharyngeal instrumentation in neonates

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## WHAT THIS PAPER ADDS

- Presentation of neonatal oesophageal perforation may be asymptomatic
- Conservative management is appropriate for the majority of neonates with isolated oesophageal perforation

## INTRODUCTION

Neonatal oesophageal perforation is a rare occurrence in the absence of oesophageal atresia or other anatomical abnormalities.<sup>1,2</sup> While spontaneous perforation has been described, it is more frequently secondary to instrumentation of the upper gastrointestinal tract or upper airway, during nasogastric tube insertion or endotracheal intubation.<sup>1</sup> These procedures may be technically challenging, especially in very low birth weight neonates, and may result in serious complications. Furthermore, the diagnosis of oesophageal perforation may be delayed or unclear. Differential diagnoses include oesophageal atresia, pharyngeal pseudo-diverticulum or an extrinsic mass. As these conditions have different management and prognosis, accurate diagnosis is crucial in order to enable appropriate treatment and avoid unnecessary procedures.

Mortality in neonates with isolated oesophageal perforation has been reported to be as high as 30%; however, this could be attributed to a population bias, as neonates with extremely low birth weight and/or gastrointestinal malformations are more frequently affected.<sup>2</sup> In recent years, the management of isolated neonatal oesophageal perforation has shifted from surgical repair to a non-operative approach, including bowel rest, total parenteral nutrition (TPN) and antibiotic therapy.<sup>1</sup> The aim of the present study was to evaluate the management of this rare condition in two large neonatal surgical centres.

## MATERIALS AND METHODS

**Study design:** This was a retrospective cohort study of neonates admitted to the Hospital for Sick Children (Toronto, Canada) and The Royal Children's Hospital (Melbourne, Australia) from March 2006 to March 2016.

**Inclusion criteria:** All neonatal patients (<44 weeks corrected gestational age) with a diagnosis of oesophageal perforation during the study period were eligible for inclusion.

**Exclusion criteria:** Patients were excluded if they had a diagnosis of oesophageal atresia and/or tracheo-oesophageal fistula.

**Data collection:** Information regarding demographics, clinical presentation, comorbidities, investigation and treatment was collected from retrospective chart review.

**Statistical analysis:** Descriptive statistics were used to analyse perinatal and postnatal characteristics. Data were expressed as median (IQR).

## RESULTS

Eleven neonates with an isolated oesophageal perforation were identified at the Hospital for Sick Children and The Royal Children's Hospital during the study period. The median gestational age at birth was 25.3 weeks (IQR 24.2 – 28.8) (Table 1). The median birth weight was 780 grams (IQR 676 – 1180), with the majority (7/11, 64%) of neonates being extremely low birth weight (ELBW). Only two patients were term ( $\geq 37$  weeks) gestation.

The diagnosis of oesophageal perforation occurred during the first day of life in 9/11 (81%) patients. The median age at presentation was 4 hours. Nine neonates were between 1 and 6 hours old at the time of presentation, one neonate was 7 days old (25 weeks' corrected gestational age) and another was 3 months old (37 weeks' corrected gestational age). Oesophageal perforation was thought to be secondary to enteric instrumentation in all neonates; 10/11 (91%) neonates had documented difficulty with nasogastric tube insertion. Of note, 10/11 (91%) neonates had been intubated prior to the diagnosis of oesophageal perforation, though none had a documented difficult intubation.

The clinical presentation of the oesophageal perforation varied between the neonates. Four neonates had symptomatic complications: three with respiratory distress secondary to pneumothorax or pneumomediastinum and one with cardiovascular instability secondary to mediastinitis. The remaining patients were asymptomatic, but had the diagnosis suspected following chest x-ray (Figure 1).

Definitive diagnosis was made with a contrast study in the majority of patients (7/11, 64%). Management was conservative in 9/11 (81%), including bowel rest, TPN and broad-spectrum intravenous antibiotics. Median duration of bowel rest was 10 days (IQR 7 – 12) and median duration of antibiotics was 7 days (IQR 7 – 10). Total parenteral nutrition was continued for a median of 20 days (IQR 7 – 39). Two neonates required operative management. The first underwent thoracotomy for washout and drainage of a mediastinal abscess, after 6 days of conservative treatment was unsuccessful. The second patient initially had a suspected

diagnosis of oesophageal atresia and therefore underwent thoracotomy to exclude this diagnosis, followed by chest drain placement.

Significant neonatal morbidity was common. Median duration of mechanical ventilation was 6 days (IQR 0.5 – 47.5) and supplemental oxygen 21 days (IQR 1.5 – 78). Three neonates had chronic lung disease (defined as oxygen requirement at 36 weeks' corrected age) and two neonates died in the neonatal period prior to hospital discharge. Both deaths were attributable to sequelae of prematurity, secondary to intraventricular haemorrhage. The two term neonates had shorter duration of TPN, and shorter duration of supplemental oxygen than the preterm neonates.

## **DISCUSSION**

This dual-centre cohort study reinforces the appropriateness of a conservative approach to management for most neonates with isolated oesophageal perforation. There has been a recent trend towards this non-operative management, with successful outcomes.<sup>2</sup> Our patient cohort was predominantly managed conservatively, with only two neonates requiring thoracotomy. This illustrates the similar, and successful, approach of two different large neonatal surgical centres across the world. This is also consistent with previous case series. In the largest published case series to date, all 25 patients were managed with bowel rest, parenteral nutrition and antibiotics.<sup>3</sup> In our case series, both thoracotomies were performed at the Australian centre, one due to suspected oesophageal atresia and the other due to mediastinitis and abscess formation. Hence, there are specific indications for operative intervention, in addition to conservative measures.

Other authors have reported variable clinical presentations of oesophageal perforation.<sup>2,4</sup> Shah *et al.*<sup>2</sup> described symptomatic presentations in all neonates with oesophageal perforation. In contrast, the majority of the neonates in the current study were asymptomatic, and the diagnosis was made due to the aberrant pathway of nasogastric tube, sometimes with associated air leak, on chest x-ray. Given the implications of enteral feeding in the presence of oesophageal

perforation, this emphasises the importance of early radiological evaluation if the diagnosis is suspected based on history.

Previous case series have demonstrated delayed attainment of full feeds, prolonged duration of supplemental oxygen and higher mortality for neonates affected by oesophageal perforation.<sup>1,2</sup> However, outcome data are influenced by the high rates of comorbidities in extremely low birth weight neonates. Consistent with this, the two neonatal deaths within our cohorts were secondary to complications of prematurity. The two term patients had less co-morbidities, shorter time on TPN and less neonatal morbidity and mortality compared to the preterm patients.

Iatrogenic neonatal oesophageal perforation is increasingly being reported in the literature. The majority of neonates in our series were extremely low birth weight, in keeping with the narrower oesophageal lumen and more frequent instrumentation required for these patients.<sup>5</sup> Oesophageal perforation is an important, and potentially preventable, complication of neonatal intensive care. It is prudent to recommend that the most skilled staff available perform oropharyngeal instrumentation, both intubation and nasogastric tube insertion. Appropriate sizing of nasogastric tubes is also important in minimising risk. Furthermore, we would recommend cessation of procedural attempts when encountering resistance or bleeding, as well as a high index of suspicion for perforation with difficult or repeated instrumentation.

Oesophageal perforation is a rare but important complication secondary to enteric instrumentation in neonates. Presentation may be asymptomatic or non-specific. Non-operative management is effective in the majority of cases, though significant neonatal morbidity is common.

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Gender – no. (%)	
Male	6 (55)
Female	5 (45)
Median gestational age (IQR) – weeks	25.3 (24.2 – 28.8)
Median birth weight (IQR) – grams	780 (676 – 1180)
Normal antenatal ultrasounds – no. (%)	11 (100)
Median time of oesophageal perforation (IQR) – hours	4 (1 – 75)
Difficult nasogastric tube insertion – no. (%)	10 (91)
Symptoms – no. (%)	
Asymptomatic	7 (64)
Respiratory distress from air leak	3 (27)
Cardiovascular instability from abscess	1 (9)
Diagnostic imaging modality – no. (%)	
Contrast study	7 (64)
X-ray	2 (18)
Bronchoscopy	1 (9)
Complications – no. (%)	
None	5 (45)
Pneumomediastinum	4 (36)
Pneumothorax (chest drain placement)	1 (9)
Pneumothorax (no chest drain placement)	1 (9)
Mediastinitis or pleural effusion	2 (18)
Management – no. (%)	
Conservative	9 (82)
Thoracotomy	2 (18)
Median antibiotic duration (IQR) – days	7 (7 – 10)
Median bowel rest duration (IQR) – days	10 (7 – 12)
Median TPN duration (IQR) – days	20 (7 – 39)
Subsequent feeding tube placement – no. (%)	

NGT (fluoroscopy)	8 (73)
NGT (at operation)	2 (18)
Nasojejunal tube (fluoroscopy)	1 (9)
Neonatal mortality – no. (%)	2 (18)

**Table 1: Patient demographics and results**

**Figure 1: Chest x-ray showing pneumothorax (superior arrows) and aberrant passage of nasogastric tube (inferior arrow)**

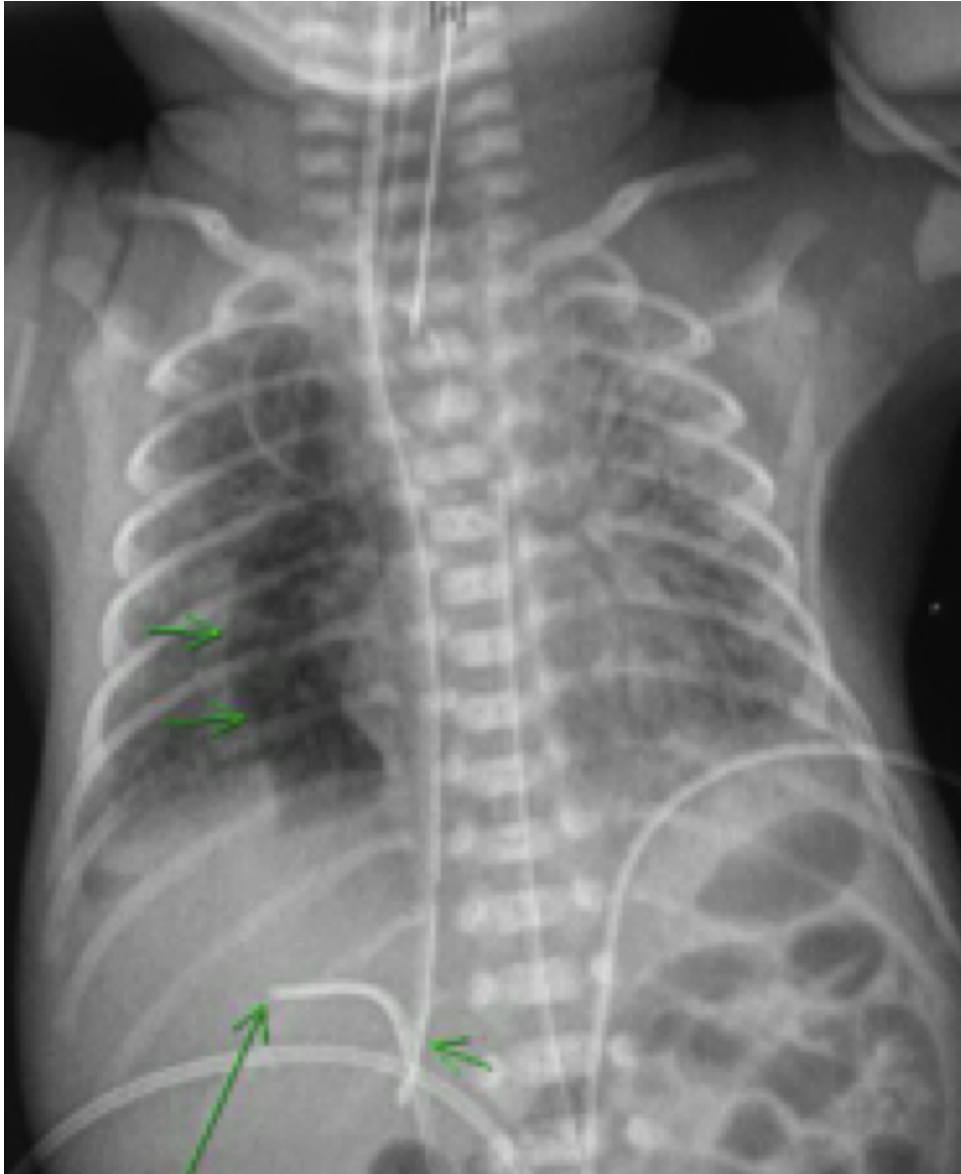


figure 1 400dpi.png

- (i) Neonatal oesophageal perforation: the role for non-operative management
- (ii) Original article
- (iii) Kate Hodgson <sup>1</sup>, Andrea Togo <sup>1,2</sup>, Aideen Moore <sup>1</sup>, Amanda Moody <sup>2</sup>, Sebastian K. King <sup>3,4,5,6</sup>, Augusto Zani <sup>7</sup>
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