

A minimally interventional approach to oesophageal atresia repair with early enteral feeding is safe, optimises neonatal outcomes and reduces resource use

Nigel J Hall, Lara Kitteringham, Ori Ron, Francesca Stedman, Michael Stanton, Robert Wheeler, Ceri Jones, Rachel Smyth, Charles Keys

1. University Surgery Unit, Faculty of Medicine, University of Southampton, Southampton, UK
2. Department of Paediatric Surgery and Urology, Southampton Children's Hospital, Southampton, UK

Corresponding author:

Nigel Hall
University Surgery Unit
Faculty of Medicine
University of Southampton
Tremona Road
Southampton
SO16 6YD
UK
E-mail: n.j.hall@soton.ac.uk

Author contributions:

Study conception and design NH, CK, CJ, LK
Acquisition of data RS, CJ, NH
Analysis and interpretation of data CJ, NH, CK, LK
Drafting of manuscript NH
Critical revision of manuscript OR, FS, LK, MS RW

Level of evidence: Level 4

A minimally interventional approach to oesophageal atresia repair with early enteral feeding is safe, optimises neonatal outcomes and reduces resource use

Abstract

Purpose:

Recent series of newborn Oesophageal Atresia (OA) repair continue to report widespread use of chest drains, gastrostomy, routine contrast studies and parenteral nutrition (PN) despite evidence suggesting these are superfluous. We report outcomes using a minimally interventional approach to post-operative recovery.

Methods:

Ethically approved (15/WA/0153), single-centre, retrospective case-note review of consecutive infants with OA 2000-2022. Infants with OA and distal trache-oesophageal fistula undergoing primary oesophageal anastomosis at initial surgery were included (including those with comorbidities such as duodenal atresia, anorectal malformation and cardiac lesions). Our practice includes routine use of a trans-anastomotic tube (TAT), no routine chest drain nor gastrostomy, early enteral and oral feeding, no routine PN and no routine contrast study. Data are median (IQR).

Results:

Of total 186 cases of OA treated during the time period, 157 met the inclusion criteria of which 2 were excluded as casenotes unavailable. TAT was used in 150 infants. A chest drain was required in 13(8%) and two infants had a neonatal gastrostomy. Enteral feeds were started on postoperative day 2(2-3), full enteral feeds established by day 4(4-6) and oral feeds started on day 5(4-8). PN was required in 15%. Median postoperative length of stay was 10 days (8-17). Progress was quicker in term infants than preterm. One infant died of cardiac disease prior to neonatal discharge.

Two planned post-operative contrast studies were performed (surgeon preference) and a further 7 due to clinical suspicion of anastomotic leak. Contrast study was therefore avoided in 94%. There were 2 anastomotic leaks; both presented clinically at day 4 and day 8 after oral feeds had been started.

Conclusion:

Our minimally interventional approach is safe. It facilitates prompt recovery with lower resource use, reduced demand on nursing staff, reduced radiation burden, and early discharge home compared to published series without adversely affecting outcomes.

Keywords:

Oesophageal atresia; clinical outcomes; quality improvement

Introduction

In recent decades the focus in treating infants born with oesophageal atresia (OA) has shifted from ensuring survival to improving other important outcomes. With overall survival of term infants with OA now exceeding 95%, attention is being paid more and more to optimising other aspects of their care. In the absence of high grade of evidence to support many interventions in these infants, surgeons may rely on other methodologies to investigate relationships between treatments delivered and outcomes.¹ Where high grade evidence does therefore not exist, non-RCT epidemiological research may play an important role in identifying opportunities to improve outcomes, reduce morbidities, burden of care and resource use.

Quality improvement initiatives aim to incrementally optimise a range of aspects of care to reach these objectives. These are typically based on standardisation of care, elimination of unwarranted variation and avoidance of interventions which do not confer direct patient benefit. Variation in care delivery may exist at a number of levels; firstly within each centre where different surgeons have a different approach to the same population of infants under their care secondly within countries and third internationally. A previous epidemiological report from the United Kingdom demonstrated variation in the care provided to infants with OA.² Similarly a large multi-institution study from the USA documented variation in peri-operative management.³ Whilst less has been formally documented regarding variation in care across international boundaries, the existing literature provides adequate evidence that such variation exists. Unless there are limitations on resource use, it is difficult to imagine, and likely hard to justify, why management should differ substantially between centres within the same country and similarly why the optimum care pathway for an infant in North America should be markedly different to that in Europe.

We have been intrigued to note that recent series of infants with EA report the widespread and routine use of a number of interventions which we do not use at our centre and which we do not believe are well supported by current evidence.^{3,4} Whilst often used with the best of intentions, we suspect the routine use of some of these interventions may in fact have a number of detrimental implications both for the infant, their family and resource use. As such, avoiding such interventions may actually have benefits for all of these stakeholders but would only be acceptable if shown to be safe and effective. We also note what we believe to be unusually long length of stay post-operatively with impact on bed days and cost. The aim of our study was to report clinical outcomes for a group of patients with OA managed using an approach that both minimises interventions as well as actively promotes early enteral feeding. We hope that in reporting our experience and outcomes we may encourage other centres to challenge their existing practice.

Methods

We undertook a retrospective single centre review of all consecutive infants born with a OA between January 2000 and September 2022. For the purposes of defining a homogenous population, only infants with OA and distal trache-oesophageal fistula (TOF) who underwent primary oesophageal anastomosis at the initial operative intervention were included. This included infants with comorbidities including duodenal atresia, anorectal malformation,

cardiac and chromosomal anomalies but excluded infants having a staged repair of OA of any description.

Data were obtained from a prospectively maintained neonatal surgical database and casenotes. We describe our population, process measures and outcomes separately for the sake of clarity. Patient level data recorded included gender, gestational age, birth weight and co-existing anomalies. Process measures recorded included use of minimally invasive surgery, use of trans-anastomotic tube (TAT), use of chest drain, gastrostomy, central venous access, parenteral nutrition (PN) and oesophageal contrast study following repair. Key outcomes were recorded up to the time of neonatal discharge and included anastomotic leak, time of commencement of enteral feeds and oral feeds, time to reach full enteral feeds, length of stay, unplanned interventions of any description related to OA repair and mortality.

For the duration of the study practice in our unit remained constant. All cases were performed via a postero-lateral thoracotomy using an extrapleural approach when possible. The azygous vein was usually divided. Following fistula division the tracheal end was closed usually with interrupted sutures and the anastomosis performed with interrupted sutures taking care to take the full thickness of the oesophagus in all sutures (i.e. including mucosa). Choice of suture material was at the discretion of the operating surgeon. Our practice included routine use of TAT (standard long term 6Fr nasogastric tube), no routine use of chest drain nor gastrostomy, early post-operative extubation (with a preference for extubation at the end of the surgical procedure when clinically appropriate), no routine placement of central venous catheter nor use of PN and no routine contrast study. Enteral feeding via TAT was started early (typically at 48 hours following the surgical procedure), and oral feeding was considered (based on clinical status) from 72 hours following the surgical procedure and subsequently guided by clinical progress. Once initially tolerated, feeding was advanced as rapidly as tolerated in term infants. In preterm infants in whom a more restricted rate of advancement of feeding was appropriate, a slower rate was followed dependent on gestational age. If there was concern about excessive tension on the anastomosis infants were electively kept intubated and paralysed for 3-5 days at surgeon discretion. Infants typically remain in our centre until they are ready for discharge home and receive basic life support training prior to discharge.

Data are reported descriptively and presented as median with interquartile range. The study was approved by the UK National Research Ethics Service (reference 15/WA/0153).

Results

Patient population

During the study. A total of 186 cases of OA were treated at our centre. Of these, 157 met the inclusion criteria and all were included except 2 for whom case notes were unavailable.

Median birth weight was 2800g (IQR 2390-3165) and gestational age 39 weeks (36.5-40). Thirty-nine (25%) were defined as preterm having been born at <37 weeks completed

gestation. Ninety-eight infants (63%) were male. Twelve had a co-existing anorectal malformation and 4 had duodenal atresia.

Process measures

All procedures were performed via open posterolateral thoracotomy with no use of minimally invasive surgery. Pre-operative rigid bronchoscopy was performed based on surgeon preference. A TAT was used in 150 of 155 infants. No infant received a chest drain routinely. Thirteen infants (8.4%) received a chest drain at some point during their neonatal stay, either for concern over air leak during the operative procedure, for treatment of pneumothorax or chylothorax following surgical repair, as management of anastomotic leak or other specific indication. No infant had a routine gastrostomy placed and just two had a gastrostomy during the neonatal period. One was a growth restricted (1100g) preterm infant (31 weeks) who developed an early anastomotic stricture in whom a gastrostomy was placed to allow ongoing enteral feeds, the second was a term infant with an early recalcitrant stricture that was not amenable to resection and who underwent cervical oesophagostomy, gastrostomy and subsequent oesophageal replacement.

Two planned post-operative contrast studies were performed (surgeon preference) and a further 7 due to clinical concern for anastomotic leak. Contrast study was therefore avoided completely in 146 (94%). Overall, enteral feeds via TAT were started on postoperative day 2(2-3), and oral feeds started on postoperative day 5(4-8). Feeds were typically started earlier in term infants compared to those born preterm (Table 1). Only preterm infants were considered candidates for routine PN.

Table 1

Neonatal outcomes

Overall, full enteral feeds established by day 4(4-6) and median postoperative length of stay was 10 days (8-17). Not surprisingly these were both longer in preterm infants compared to those born at term (Table 1). PN was required overall in 15% of infants, the majority of whom required it for reasons related to prematurity. Term infants requiring PN received it for a specific indication including anastomotic leak, chylothorax, or co-existing intestinal disease. There were 2 anastomotic leaks; both presented clinically at day 4 and day 8 after oral feeds had been started. All but one infant survived to discharge home. This one death was due to non-survivable cardiac disease.

Unplanned and related further procedures during the initial neonatal stay were performed in 10 infants and listed in Table 2.

Table 2

Discussion

The aim of our study was to document outcomes achieved in a medium volume centre where we have utilised a minimally interventional approach alongside early enteral feeding in our care of infants with type-C OA over the past 20 years. Whilst we acknowledge that we do not report a cohort managed in a different way for comparison we present our series to

illustrate outcomes achievable without a number of additional interventions that we are aware are frequently used in centres worldwide but for which the evidence base to support their use does not exist. We briefly discuss the rationale for a number of aspects of care which we recognise as important.

Our approach to additional procedures at the time of OA repair has been to avoid them unless there is specific indication to use them. Therefore, we have avoided placing a chest drain routinely and have avoided the use of gastrostomy. We and others have previously documented that routine use of a chest drain is not necessary^{5,6} yet chest tubes continue to be used routinely.^{7,8} We are unclear why and propose that they confer no benefit, likely carry additional cost, themselves are associated with morbidity and likely additional radiographs performed to determine time for chest drain removal. We acknowledge that in our series a very small number of infants did receive a chest drain during the surgical procedure and that a small number (fewer than one in 10) did ultimately require one for very specific indications. Although negative outcomes for these infants, we do not believe this proportion is high enough to justify routine use of a chest drain in all cases. Furthermore, not using a chest drain likely reduces the number of chest radiographs the infant receives (although we acknowledge we have not reported data on this). Similarly, although many centres have significantly reduced the routine use of gastrostomy in OA patients in recent decades, large datasets suggest gastrostomy is being used more frequently than only in these more complicated cases – 44% of all OA cases in a recent large series of 2,509 cases.⁹ Our data suggest that routine gastrostomy for OA with distal TOF is not necessary but clearly in specific circumstances may be appropriate.

We have elected to avoid routine contrast studies following repair on the basis that we do not believe they contribute significantly to management decisions in the vast majority of cases. If infants are well and show no clinical evidence of anastomotic leak we have elected to feed them orally as they will be swallowing their own saliva from the time they are extubated. With this approach we have identified just 2 anastomotic leaks, both of which were apparent clinically after oral feeds were started. One of these was diagnosed on post-operative day 4, earlier than most centres report carrying out the routine contrast study (oesophogram). Our preference has been to reserve contrast studies for infants in whom we have specific clinical concern. In this way we have avoided contrast study in 94% of cases with resultant cost savings and reduced radiation exposure for these infants. Radiation exposure is known to be significant in this population of infants.¹¹ The real benefit to avoidance of routine contrast study though is that we have felt confident to commence oral feed and progress these based on clinical status rather than waiting for radiological confirmation of an intact anastomosis. Existing literature suggests that routine contrast studies are frequently performed prior to commencing oral feeds and that typically these have been performed at seven days following surgical repair.⁷ A recent series challenged this and suggested that the routine post operative contrast study can be safely obtained on day 5 and that this would likely result in oral feeds being initiated sooner.⁷ Indeed a post-implementation study demonstrated that this benefit could be realised.¹ Yet it is our belief that in well infants without specific concern, a routine contrast study can be avoided completely and this approach can bring with it even greater benefits.

Consistent with our aims to advance recovery following surgery as quickly as is safely possible, our approach to feeding infants with OA has been to use the gastrointestinal tract wherever possible and specifically to avoid additional feed related interventions including gastrostomy, central venous access and parental nutrition unless there is a specific indication to do so. To achieve these aims we have chosen to place a trans-anastomotic tube routinely rather than preserving them only for infants in whom we have concern about the anastomosis. This enables us to start enteral feeds soon after surgery and often on the first or second post operative day. We believe there are real benefits to avoiding central venous access and PN in all infants including those with OA. In addition to reducing cost, central venous access and PN are associated with a significant risk of infection in infants with congenital surgical anomalies and we believe there is benefit in avoiding it unless a specific indication exists. Our data support the concept of early enteral feeds and avoidance of PN. Using this approach we have been able to achieve full enteral feed on median post operative day 4 in term infants. In contrast, we note that initiation of enteral feed in reported series is often later (e.g. 8-9 days) even when a TAT is used.⁷ Such practice demands that PN be used for many days and typically up to 2 weeks to maintain adequate nutrition^{1,10} yet we believe in the majority it can be avoided completely.

With our approach of minimising interventions and progressing enteral feeds rapidly we have achieved a median length of stay of 10 days in term infants. This is markedly shorter than the majority of large recent published series^{4,8,12} and we believe represents the cumulative benefits of all steps of our treatment pathway.

We note recent concern regarding the relationship between TAT use^{7,10} and the later development of anastomotic stricture and feel it important to comment on this in light of our recommendation to use a TAT in all cases. We cannot examine this relationship in our series since almost all cases received a TAT. Whilst the incidence of anastomotic stricture is beyond the scope of this report since here we only report outcomes to neonatal discharge, we recognise that many may be concerned about using a TAT given recent concerns. We routinely monitor our anastomotic stricture rate and have found this to be consistent with both the current literature and national rates.^{13,14} Our institutional opinion is that the relationship that has been identified between TAT use and higher rate of anastomotic structure is best explained by the fact that many centres do not use a trans-anastomotic tube routinely, rather a trans-anastomotic tube is used selectively and is more likely to be used in cases where the surgeon has some concern about the anastomosis for instance because it is under tension or some other technical factor. These factors themselves are independently likely to be associated with higher chance of stricture.¹⁵ The use of a trans-anastomotic tube may therefore be associated with a higher chance of stricture formation but this relationship is associative rather than causative in nature.

Many surgeons may be surprised to find that we have not embraced minimally invasive repair of this type of OA. Put simply, we do not believe that with the volumes available to us and our current place on the learning curve there is significant benefit to infants under our care from using minimally invasive surgery. We believe that the results we present support this belief. However, we do acknowledge that in the longer-term chest wall deformity is one particular outcome that may be improved by minimally invasive surgery. Whilst we have not reported this long term outcome here, we aim to use a muscle sparing approach to postero-

lateral thoracotomy wherever possible and pay careful attention to avoid over approximation of the ribs during chest wall closure.¹⁶ Despite benefits of minimally invasive surgical repair being promoted by some, we are struck by the fact that even in large recent multicentre reports and national database studies only a minority of cases (11-15%) were performed via thoracoscopy.^{4,8} Therefore we do not believe we are an outlier in our practice. The most recent meta-analysis comparing minimally invasive and open repair reported a statistically significant reduction in a number of metrics with minimally invasive surgery including reduced time to first oral feed (of 2.8 days) and reduced hospital length of stay (of 11.9 days).¹⁷ Given that we start oral feeds on median post-operative day 5 and our median length of stay was just 10 days we are not convinced that introducing minimally invasive surgery would overall confer benefit to the infants we treat.

We recognise some limitations to our study including that we report outcomes in only a subset of cases of OA but justify this since this represents the majority of cases and allows a definable homogenous cohort to which other centres can make comparison. We also recognise that we have only reported short term outcomes to neonatal discharge and that longer term outcomes are clearly important. Here we wished to focus specifically on these early peri-operative outcomes and processes.

In conclusion, we have demonstrated that excellent short term outcomes following surgical repair of OA/TOF can be achieved without the need for a range of interventions commonly used in pediatric surgical practice and with a specific focus on early enteral feeding. We accept that reasonable surgeons may have a different approach to peri-operative management to the one we propose but whilst respecting their preferences, we wish to advance an alternative reasonable standard of care, which is less interventional, less costly, reduces a number of burdens and arguably reinstalls native physiological processes more rapidly. Our report of our experience in a large cohort of patients reveals that this is not only safe and that outcomes have not been compromised by a minimally interventional approach, but arguably that our patients have reaped the benefit of avoiding unintended consequences and complications of superfluous interventions. We urge surgeons to challenge existing surgical dogma as others have also proposed⁷ to optimize care and outcomes for this group of infants.

References

- 1 Bence, C. M. *et al.* Clinical outcomes following implementation of a management bundle for esophageal atresia with distal tracheoesophageal fistula. *J Pediatr Surg* **56**, 47-54, doi:10.1016/j.jpedsurg.2020.09.049 (2021).
- 2 Burge, D. M. *et al.* Contemporary management and outcomes for infants born with oesophageal atresia. *British Journal of Surgery* **100**, 515-521, doi:10.1002/bjs.9019 (2013).
- 3 Lal, D. R. *et al.* Perioperative management and outcomes of esophageal atresia and tracheoesophageal fistula. *J Pediatr Surg* **52**, 1245-1251, doi:10.1016/j.jpedsurg.2016.11.046 (2017).
- 4 Etchill, E. W., Giuliano, K. A., Boss, E. F., Rhee, D. S. & Kunisaki, S. M. Association of operative approach with outcomes in neonates with esophageal atresia and

- tracheoesophageal fistula. *J Pediatr Surg* **56**, 2172-2179, doi:10.1016/j.jpedsurg.2021.04.006 (2021).
- 5 Anand, S., Singh, A., Krishnan, N. & Yadav, D. K. Whether prophylactic intraoperative chest drain insertion in esophageal atresia-tracheoesophageal fistula is an evidence-based practice or just a prejudice: A systematic review and meta-analysis. *J Pediatr Surg* **57**, 1554-1560, doi:10.1016/j.jpedsurg.2021.06.015 (2022).
 - 6 Paramalingam, S., Burge, D. M. & Stanton, M. P. Operative intercostal chest drain is not required following extrapleural or transpleural esophageal atresia repair. *Eur J Pediatr Surg* **23**, 273-275, doi:10.1055/s-0032-1330845 (2013).
 - 7 Lal, D. R. *et al.* Challenging surgical dogma in the management of proximal esophageal atresia with distal tracheoesophageal fistula: Outcomes from the Midwest Pediatric Surgery Consortium. *J Pediatr Surg* **53**, 1267-1272, doi:10.1016/j.jpedsurg.2017.05.024 (2018).
 - 8 Marquart, J. P. *et al.* Thoracoscopy versus thoracotomy for esophageal atresia and tracheoesophageal fistula: Outcomes from the Midwest Pediatric Surgery Consortium. *J Pediatr Surg* **58**, 27-33, doi:10.1016/j.jpedsurg.2022.09.015 (2023).
 - 9 Patterson, K. *et al.* Quantifying Upper Aerodigestive Sequelae in Esophageal Atresia/Tracheoesophageal Fistula Neonates. *Laryngoscope* **132**, 695-700, doi:10.1002/lary.29798 (2022).
 - 10 LaRusso, K. *et al.* Effect of transanastomotic feeding tubes on anastomotic strictures in patients with esophageal atresia and tracheoesophageal fistula: The Quebec experience. *J Pediatr Surg* **57**, 41-44, doi:10.1016/j.jpedsurg.2021.09.014 (2022).
 - 11 Zamara, P. *et al.* Long-term burden of care and radiation exposure in survivors of esophageal atresia. *J Pediatr Surg* **50**, 1686-1690, doi:10.1016/j.jpedsurg.2015.05.006 (2015).
 - 12 Sfeir, R. *et al.* Risk Factors of Early Mortality and Morbidity in Esophageal Atresia with Distal Tracheoesophageal Fistula: A Population-Based Cohort Study. *J Pediatr* **234**, 99-105 e101, doi:10.1016/j.jpeds.2021.02.064 (2021).
 - 13 Vergouwe, F. W. T. *et al.* Risk factors for refractory anastomotic strictures after oesophageal atresia repair: a multicentre study. *Arch Dis Child* **104**, 152-157, doi:10.1136/archdischild-2017-314710 (2019).
 - 14 Allin, B., Knight, M., Johnson, P., Burge, D. & Baps, C. Outcomes at one-year post anastomosis from a national cohort of infants with oesophageal atresia. *PLoS One* **9**, e106149, doi:10.1371/journal.pone.0106149 (2014).
 - 15 Aumar, M. *et al.* Predictors of anastomotic strictures following oesophageal atresia repair. *Arch Dis Child Fetal Neonatal Ed* **107**, 545-550, doi:10.1136/archdischild-2021-322577 (2022).
 - 16 Wei, S., Saran, N. & Emil, S. Musculoskeletal deformities following neonatal thoracotomy: long-term follow-up of an esophageal atresia cohort. *J Pediatr Surg* **52**, 1898-1903, doi:10.1016/j.jpedsurg.2017.08.062 (2017).
 - 17 Drevin, G., Andersson, B. & Svensson, J. F. Thoracoscopy or Thoracotomy for Esophageal Atresia: A Systematic Review and Meta-analysis. *Ann Surg* **274**, 945-953, doi:10.1097/SLA.0000000000004239 (2021).

Table 1: timing of feed initiation and neonatal outcomes in term and preterm infants

		n	Enteral feed start (days)	Full enteral feeds (days)	Oral feed start (days)	PN	Chest drain	Leak
Term	>37wks	116	2 (2-3)	4 (4-5)	5 (4-6)	9	9	1
Preterm	<37wks	39	3 (2-4)	9 (5-14)	11 (5-25)	14	4	1

PN – parenteral nutrition

Data are median (IQR)

Table 2: additional unplanned and related procedures performed during neonatal stay

Procedure	Number of infants
Dilatation of oesophageal anastomosis	8
Gastrostomy formation	2
Thoracotomy and repair air leak from lung	1
Repair recurrent TOF	1
Cervical oesophagostomy*	1
Resection distal congenital oesophageal stricture	1

TOF – trache-oesophageal fistula; *- required for early recalcitrant stricture