



Immediate primary anastomosis for isolated oesophageal atresia: A single-centre experience

Ibrahim Uygun, Hikmet Zeytun, Selcuk Otcu

ABSTRACT

Background: Isolated oesophageal atresia without tracheo-oesophageal fistula represents a major challenge for most paediatric surgeons. Here, we present our experience with six neonates with isolated oesophageal atresia who successfully underwent immediate primary anastomosis using multiple Livaditis circular myotomy. **Materials and Methods:** All six neonates were gross type A isolated oesophageal atresia (6%), from among 102 neonates with oesophageal atresia, treated between January 2009 and December 2013. Five neonates were female; one was male. The mean birth weight was 2300 (range 1700-3100) g. **Results:** All six neonates successfully underwent immediate primary anastomosis using multiple myotomies (mean 3; range 2-4) within 10 (median 3) days after birth. The gap under traction ranged from 6 to 7 cm. One neonate died of a major cardiac anomaly. Another neonate was lost to follow-up after being well for 3 months. Three anastomotic strictures were treated with balloon dilatation, and four anastomotic leaks were treated conservatively. The mean duration of follow-up was 33 months. **Conclusions:** To treat isolated oesophageal atresia, an immediate primary anastomosis can be achieved using multiple myotomies. Although, this approach is associated with high complication rates, as are other similar approaches, these complications can be overcome.

Key words: Anastomotic leak, anastomotic stricture, isolated oesophageal atresia, long-gap oesophageal atresia, myotomy

INTRODUCTION

Long-gap oesophageal atresia represents a major challenge for most paediatric surgeons, particularly

gross type A isolated oesophageal atresia without tracheo-oesophageal fistula.^[1-16] The best surgical approach for isolated oesophageal atresia repair remains undetermined. Achieving a consensus regarding the best procedure is difficult. However, the native oesophagus has been shown to be the best conduit.^[1-5] Many surgeons have reported success with several surgical oesophageal elongation approaches, performed alone or in combination, in the treatment of neonates with isolated oesophageal atresia; these approaches include external traction suture, oesophageal myotomy, delayed primary anastomosis and extrathoracic oesophageal elongation.^[6-16] If these approaches cannot be performed, oesophageal replacement surgery using a digestive organ, such as the colon, stomach, ileum, or jejunum, is necessary.^[17,18] However, this type of surgery still represents a major challenge and is associated with high complication rates than oesophageal lengthening, which aims to preserve the native oesophagus.^[1,2,17,18] Tissue engineering studies have not yet attained reliable outcomes.^[19] Conversely, experimental oesophageal transplantation studies have shown promising results for oesophageal replacement surgery; however, no study has yet been conducted in humans.^[20,21] The procedures based on preservation of the native oesophagus could be more acceptable for the treatment of isolated oesophageal atresia.^[1-16]

Herein, we present in detail our experience with six neonates with isolated oesophageal atresia who

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successfully underwent immediate primary anastomosis using multiple Livaditis circular myotomy.

MATERIALS AND METHODS

All six neonates were gross type A isolated oesophageal atresia (6%), from among 102 neonates with oesophageal atresia, treated between January 2009 and December 2013. Five neonates were female; one was male. Prenatal ultrasonography revealed polyhydramnios in all neonates and the inability to identify the stomach in one. The mean birth weight was 2300 g (range, 1700-3100 g) and the mean gestational age was 37 weeks (range, 32-40 weeks). All neonates underwent immediate primary anastomosis using Livaditis myotomy, as described below in detail. The characteristics of the cases and surgical details are summarised in Table 1 and Figures 1-3.

We performed primary oesophageal atresia repair immediately in the first operation in all neonates with all types of oesophageal atresia, including gross type A oesophageal atresia. Pre-operative evaluation consisted of routine physical examination, blood analysis, baby X-ray with oro-oesophageal feeding tube, abdominal ultrasonography, cardiology consultation and other consultations, if needed. We inserted a large radio-opaque 10-French oro-oesophageal feeding tube to confirm the diagnosis. However, we did not perform a routine barium study to avoid aspiration risk.^[12] We then operated immediately during the optimum time for the neonate after pre-operative preparation (e.g., administration of intravenous fluids and antibiotics).

Oesophageal repairs were performed by right posterolateral thoracotomy through the 5th intercostal space, to reach the both segments, using muscle-sparing technique with small skin incision and transpleural approach. After the mediastinal parietal pleura had been opened, the azygos vein was ligated and divided using a 4-0 polyglycolic acid suture (Pegesorb®; Dogsan AS, Trabzon, Turkey). The vagus nerve was identified and preserved in all cases by gentle medial displacement. The distal segment was identified using blunt dissection and then mobilised. A 4-0 polypropylene traction suture with round needle (Propilen®; Dogsan AS) was placed through the tip of the distal segment for traction.

The proximal segment was identified using blunt dissection to facilitate the insertion of a large (10-French) oro-oesophageal feeding tube, and a 4-0 polypropylene traction suture was placed through the tip of the proximal segment, and through the feeding tube, to

Table 1: Characteristics of cases (n = 6) with isolated oesophageal atresia (gross type A) underwent immediate primary anastomosis, with surgical features and results

Case	Gender	GA (weeks)	BW (g)	AA	Age (days)	Gap (cm)	OT (min)	Myotomy (n)	Anastomosis P/D/AT	AL (days)	OS/OBD (n)	VSR (days)	ICT (days)	NGT (days)	NJT (days)	Feeding (d) initial/fall/oral	HS (days)	Results	Follow-up (month)
1	Female	36	2400	None	10	6.5	175	P(2)	XL/S/T	—	—	No	No	6	no	4/6/6	8	Lost to follow-up after 3 months of wellness	—
2	Female	40	3100	None	1	6.5	210	P(2)	L/S/VT	+/42	—	6	44	3	49	3/7/43	60	Tolerated full PO well, mild GOR treated medically	54
3	Female	32	1700	Major cardiac	2	6	230	P(1), D(2)	L/S/T	—	—	4	4	4	—	—	—	Died at 4 d old from cardiac anomaly	—
4	Female	38	2330	None	3	7	300	P(2), D(2)	L/S/VT	+/42	+/2	6	45	4	52	4/8/43	79	Tolerated full PO well	34
5	Male	37	1900	Minor cardiac	5	7	240	P(1), D(2)	L/S/VT	+/41	+/1	11	41	5	40	3/7/41	47	Tolerated full PO well, mild GOR treated medically	25
6	Female	36	2500	None	1	6	210	P(1), D(1)	L/S/VT	+/40	+/2	3	42	2	38	4/8/41	46	Tolerated full PO well, severe GOR treated surgically	18

AA: Associated anomaly; AL: Anastomotic leak duration; AT: Anastomotic tension; BW: Birth weight; D: Distal segment; GA: Gestational age; GOR: Gastro-oesophageal reflux; HS: Hospital stay post-operatively; ICT: Intercostal chest tube duration; L: Large; min: Minutes; NGT: Nasogastric tube duration; NJT: Nasojejunal tube duration; OBD: Oesophageal balloon dilatation; OS: Oesophageal stricture; OT: Operation time; P: Proximal segment; PO: Peroral feeding; S: Small; T: Tense; VSR: Ventilatory support requirement duration; VT: Very tense; XL: Extra large

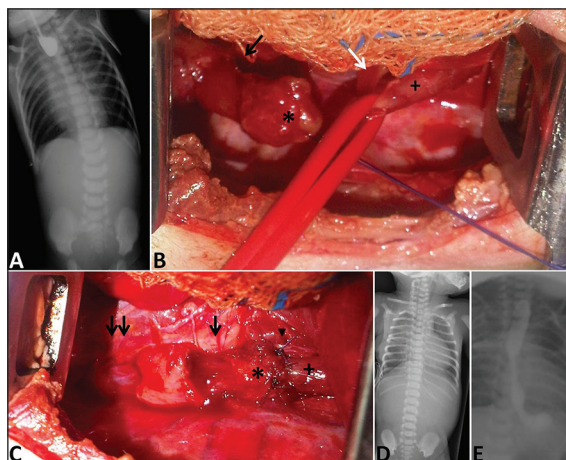


Figure 1: First case (a-e). The chest X-ray showed gasless abdomen and an extra-large proximal segment (a). In the operation (b and c), the long atretic strand (white arrow) between the trachea and distal segment (+) was preserved and retracted with a vessel loop to facilitate dissection and mobilisation of the distal segment. After two proximal (*) myotomies (black arrow, first myotomy; double black arrow, second myotomy), primary anastomosis (black arrowhead) was achieved. No chest tube was inserted (d), post-operative chest X-ray. On post-operative day 8, an oesophagogram (e) yielded good findings, and the patient was discharged on the same day

assist in the proximal segment dissection and avoid the trauma caused by repeatedly applying forceps.^[12] The gap between oesophageal segments was measured under traction before aggressive dissection. The proximal segment was dissected aggressively and mobilised by blunt dissection and by gently using a pair of scissors. The distal segment was also mobilised aggressively. After aggressive mobilisation of both segments, circular myotomy was gently performed in the proximal segment using a no. 15 blade. Multiple myotomies were performed in both segments, if required. After sufficient oesophageal elongation, anastomosis was performed using seven or eight primary separated absorbable 5-0 or 6-0 sutures (polyglycolic acid sutures [Pegesorb®] or polydioxanone [Pedesente®]; Dogsan AS) over the feeding tube (5- or 6-French) inserted into the stomach. If an extremely stretched anastomosis, an oesophageal tear, or excessive blood/fluid/air drainage occurred during surgery, a chest tube was inserted into the operating area. After surgery, the anaesthesia process was reversed and the neonates were cared for in our neonatal Intensive Care Unit. No post-operative elective ventilatory support was used to protect the anastomosis. The patients were fed as soon as possible (post-operative day 3 or 4) via a nasogastric tube, with minimal enteral feeding by measuring the gastric residual volume. We did not routinely perform barium studies to control anastomosis. If signs of anastomotic leakage were evident in the chest tube or on the chest X-ray, the nasogastric tube was converted to use as a nasojejunal tube by advancing it to the jejunum under fluoroscopic guidance, and continuous jejunal feeding was started

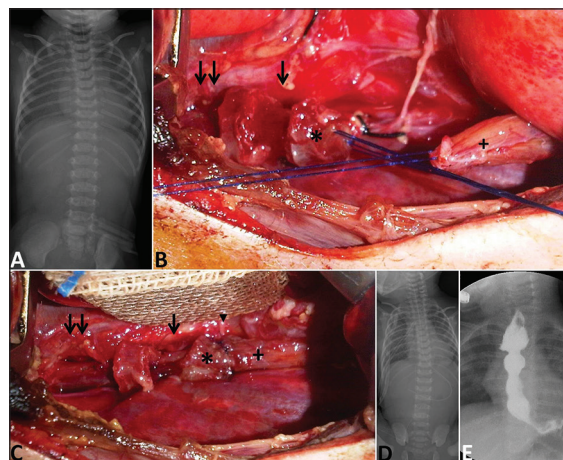


Figure 2: Second case (a-e). The chest X-ray showed gasless abdomen (a). In the operation (b and c), the distal segment (+) found to be small. After two proximal (*) myotomies (black arrow, first myotomy; double black arrow, second myotomy), primary anastomosis (black arrowhead) was achieved. The second chest tube was required to drain the oesophageal leak (d). In an oesophagogram taken 1-year post-operatively (e), no oesophageal stricture and slight oesophageal ballooning at the myotomy sites were observed

with breast milk via this tube. Within 3-5 days, a gradual increase to full feeding occurred and broad spectrum antibiotics were administered. A barium study was performed weekly to assess leak closure. If a severe anastomotic stricture was evident after 1-month post-operatively, early oesophageal balloon dilatation was performed using a 8- or 10-mm balloon (Controlled Radial Expansion [CRE™] wire-guided balloon dilator; Boston Scientific Corp., Galway, Ireland) to treat the stricture and minimise leakage. After the absence of a leak was confirmed by the barium study and chest tube drainage, the baby fed orally. If no complication occurred, the nasojejunal tube was removed first, followed by the chest tube. The baby was subsequently discharged.

RESULTS

All neonates successfully underwent immediate primary anastomosis using multiple myotomies (mean number of myotomies 3; range 2-4) within 10 days after birth (range 1-10 days; median 3 days) [Figures 1-3]. One surgeon (Dr. Uygun) performed all of the surgeries. The gaps were measured during surgery and were found to be 6-7 (mean 6.5) cm in length. The anastomoses were very tense in four cases. The aortic arch was right-sided only in our third patient. One neonate died early in the post-operative period due to a major cardiac anomaly. One neonate was lost to follow-up after 3 months of wellness. In the remaining neonates, three anastomotic strictures were treated with balloon dilatation, four anastomotic leaks were treated conservatively, two mild cases of gastro-oesophageal reflux were treated

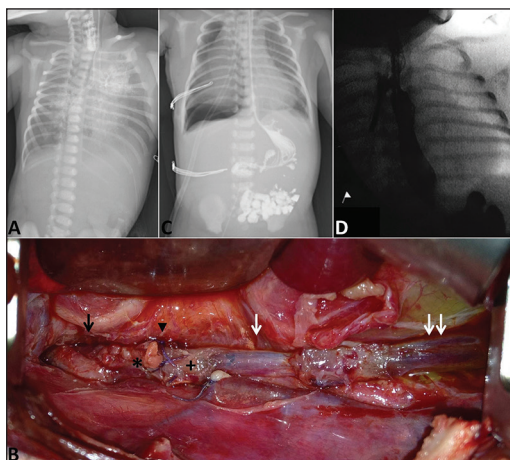


Figure 3: Fourth case (a-d). The chest X-ray with upper barium study performed at the referring clinic showed gasless abdomen, and the patient's lungs were aspirated using radio-opaque agent (bronchogram) (a). After four (two proximal and two distal) circular myotomies (black arrow, first proximal myotomy; white arrow, first distal myotomy; double white arrow, second distal myotomy; second proximal myotomy not shown), primary anastomosis (black arrowhead) was achieved (b). A second anterior chest tube was needed to drain the oesophageal leak (c and d). The anastomotic leak (d) had closed spontaneously on post-operative day 42 (*: Proximal segment; +: Distal segment)

medically, and one severe case of gastro-oesophageal reflux was treated surgically. All neonates tolerated peroral feeding well. The average duration of follow-up was 33 (range 18-54) months. The characteristics of the cases and surgical details are summarised in Table 1.

DISCUSSION

Long-gap oesophageal atresia gross type A remains a major challenge for surgeons.^[1-5] Several successful techniques and approaches to preserve the native oesophagus have been described.^[6-16] Although all of these procedures have high incidence rates of complications, such as severe gastro-oesophageal reflux, anastomotic leakage and oesophageal stricture, these complications are significantly less severe than the oesophageal replacement surgery.^[1-18] However, the best approaches to and timing of treatment remain unknown.^[2]

Oesophageal elongation using external traction sutures, described by Foker *et al.*, can be completed during primary anastomosis in the neonatal period, even if a second thoracotomy is required.^[8,10,13,22] However, this modified technique has some disadvantages while waiting for the definitive surgery, including thread breakage, pneumothorax, skin erosion, and the increased likelihood of a required gastrostomy.^[10,13,22]

Delayed primary anastomosis has been the preferred approach for the treatment of type A oesophageal

atresia since 1981 when it was reported by Puri *et al.*^[4,7] However, in our previous cases we found no reduction in gap length despite growth of the neonate. Boyle *et al.* and Giacomoni *et al.* reported similar observations.^[5,23] Boyle *et al.* recommended waiting only for the neonate to reach a good nutritional state and be vigorous (≥ 3.5 kg in weight), to enhance the likelihood of success.^[5] Disadvantages of this approach are the requirement for gastrostomy and/or additional surgery and probable long hospitalisation time for upper oesophageal pouch aspiration and its accompanying complications, such as aspiration pneumonia and sepsis.^[4,7] Another delayed primary anastomosis, the multistaged extrathoracic oesophageal elongation approach described by Kimura *et al.*, has similar disadvantages and requires multiple (1-5) surgical procedures.^[9]

Livaditis first described an oesophageal circular myotomy for elongation during oesophageal atresia repair.^[6] This technique has been widely used by most surgeons performing one or more, immediate or delayed primary anastomoses in the proximal segment and/or distal segment, alone or in combination with other techniques, for 40 years.^[3,6,12,23-25] In the Pediatric Surgery textbook, Harmon and Coran stated that he has occasionally used three proximal myotomies made through an additional cervical incision.^[12] Although complications such as anastomotic leakage and gastro-oesophageal reflux frequently occur with this technique, they are similar to those associated with other techniques.^[3,6,23-25] Additionally, although ballooning at the myotomy site may occur, this finding was reported to be unproblematic.^[24,25] Ricketts *et al.* reported that the incidence of ballooning at the myotomy site was low (17%), and it did not interfere with emptying of the oesophagus.^[25] Giacomoni *et al.* observed no ballooning at the myotomy site in any of their patients; however, Lai *et al.* reported that it was observed in all five of their patients, but peristalsis and motility of the oesophagus did not appear to be impaired by the myotomies compared with patients with oesophageal atresia who underwent primary repair without myotomy.^[23,24] Burjonrappa *et al.* resorted to an aid to myotomy in 2/15 cases in their series of patients with type A oesophageal atresia; they reported that they had moved away from this technique because of reported problems with diverticula formation.^[3] However, interestingly, they reported no evidence of diverticula formation in two cases with myotomy; one recurrent diverticula formation was observed in a patient who had undergone treatment with Fokers' technique.^[3] In our cases, we observed only one ballooning, which caused mild swallowing symptoms. Myotomy may damage the

oesophageal muscle. However, we believe that other elongation techniques based on extensive traction, such as extrathoracic multistaged and extrathoracic external traction techniques, may also cause breakage and damage to the muscle fibres of the oesophagus. The myotomy technique can be performed in all long-gap oesophageal atresia cases at any time, as a readily available aid for oesophageal elongation for paediatric surgeons.

When indicated, aggressive mobilisation of the distal segment is preferred to preserve the native oesophagus; however, the procedure should be performed gently to preserve the vagus nerve because of the high likelihood of motility dysfunction.^[8,11,12,26] Also, a circular myotomy should be performed.^[3,6,12,23-25] We also mobilise both segments and the multiple circular myotomies (two in the proximal segment and two in the distal segment) aggressively if required. In an experimental oesophageal transplantation study in rats, we observed that total oesophageal transplantation could be performed directly without vascular anastomosis and that the oesophagus and its bed have excellent compatibility and promote revascularisation and healing.^[21] Aggressive mobilisation of both segments of the oesophagus is an intervention similar to transplantation, implantation, or free grafting without vascular anastomosis, and all of these procedures can be successful. Given the anatomy and histology of the oesophagus, particularly the absence of serosa and presence of multiple thin vessels, implantation is possible. Despite the long-held opinion that the blood supply to the distal segment is tenuous and might be compromised by mobilisation, many surgeons have found that the distal segment can be, and often is, mobilised to facilitate a primary anastomosis.^[12] Finally, although the distal segment is generally very vulnerable to surgical manipulation, we encourage surgeons to perform aggressive mobilisation and additional multiple circular myotomies in both segments, handling them very carefully, if they are required to preserve the native oesophagus.

We place a traction suture through the tip of the proximal segment and the feeding tube to assist in proximal segment dissection and avoid trauma and tearing.^[12] This will not tear the proximal segment due to the strength of the oro-oesophageal feeding tube. Circular myotomy can be performed over the tube, but the tube does not allow sufficient (balloon-like) expansion. However, we could not use instruments such as Foley or Fogarty catheters in any case because they were not suitable. We believe that development of a special oesophageal traction tube with and without a balloon is needed.

Paramalingam *et al.* reported that an operative chest tube was not required during repair of oesophageal atresia with a tracheo-oesophageal fistula using extra and transpleural approaches.^[27] We prefer to use a transpleural approach because the operating time is shorter and, with current antibiotic options and intensive care, the risk of empyema after a leak is minimal.^[12] We prefer to insert a chest tube after the oesophageal atresia repair in cases in which we predict that leakage or drainage could occur. In the current series, a chest tube was not inserted in the first neonate with moderate anastomotic tension, who was treated successfully.

Intensive post-operative care is very important for neonates with long-gap oesophageal atresia.^[1,2] Some surgeons prefer post-operative elective ventilatory support to protect a tense anastomosis.^[2,5,8,11,22] By contrast, we performed no additional intervention and made no extra effort in post-operative care other than ordinary oesophageal atresia repair, such as proper fixation of the transanastomotic nasogastric tube and respiratory care. However, five neonates needed post-operative ventilatory support (mean 6 days) for respiratory failure due to pneumonia, atelectasis and anastomotic leaks. Early enteral feeding via a transanastomotic nasogastric tube is an essential part of post-operative care. Alabbad *et al.* reported that early transanastomotic nasogastric feeding reduced the total duration of parenteral nutrition and the length of hospitalisation after oesophageal atresia surgery.^[28] We have also observed that early feeding has many positive effects in the post-operative period, not only in neonates with oesophageal atresia but also in those who have undergone other surgeries. However, although we deemed early (within 24 h, or even 4 h, post-operatively) feeding to be important after routine oesophageal atresia repair without a myotomy, we hesitated to start early feeding after isolated oesophageal atresia repair; feeding was initiated 3 or 4 days post-operatively in those neonates. Finally, total parenteral nutrition was not needed in any case.

No mortality due to the surgical approach occurred in our series. One premature neonate (1700 g) died in the early post-operative period due to major cardiac anomaly (Spitz Class IV).

Anastomotic strictures are the most common complication after oesophageal atresia.^[12] Most of these respond to one or two dilatations. Risk factors for oesophageal stricture are anastomotic tension, anastomotic leakage and gastro-oesophageal

reflux.^[1,2,12,14] Chang *et al.* reported that post-operative oesophageal strictures developed more often in long-gap than short-gap oesophageal atresia, whereas a circular myotomy was not significantly associated with oesophageal stricture.^[14] In our cases, three anastomotic strictures developed in four cases with anastomotic leakage and were easily treated with early balloon dilatations.

Oesophageal leakage after oesophageal atresia repair or balloon dilatation is almost always treated conservatively.^[3,9,10] We also observed that anastomotic leakage in the patients with oesophageal atresia who had undergone surgery at our hospital, including four patients in this series, healed after the application of our oesophageal leakage treatment protocol, comprising chest tube(s) drainage, antibiotics and full nasojunal feeding instead of total parenteral nutrition. Although leakage requires long-term hospitalisation, we believe that it can resolve without severe complications.

Should type A oesophageal atresia be managed using immediate or delayed primary anastomosis? The better procedure has not been identified because of insufficient data.^[2] Although some oesophageal atresia cases had gasless abdomens on X-ray, they were reported as oesophageal atresia with a proximal tracheo-oesophageal fistula (up to 50% of cases with a gasless abdomen), oesophageal atresia with an oesophageal web without long-gap, or type A oesophageal atresia without long-gap.^[29] Therefore, the choice of delayed primary anastomosis for treatment was unnecessary and overly intricate in these cases. In our first neonate with isolated oesophageal atresia, a long atretic strand (subtype IIIb5 in Kluth's atlas classification) was observed between the trachea and distal segment [Figure 1b].^[30] Although it could not reduce the gap, it grew along the distal segment, facilitating distal segment mobilisation and anastomosis. The proximal segment is mostly hypertrophic and dilated; nevertheless, we consider the distal segment to be small, and its oesophageal lumen was also distal.

CONCLUSION

To treat isolated oesophageal atresia, paediatric surgeons can achieve immediate primary anastomosis using multiple circular myotomies. However, this approach is associated with high complication rates, as are other similar approaches, these complications can be overcome. In our limited series, six neonates with type A oesophageal atresia successfully underwent immediate primary anastomosis using multiple Livaditis circular myotomies. However, further studies are required to

investigate the long-term results of this approach in large case series.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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