MSI Journal of
Medicine and Medical Research (MSIJMMR)

Case Report

Rare Type A Esophageal Atresia: Successful Neonatal Management with Awake Intubation

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The authors declare that no funding was received for this work.

Received: 14-November-2025
Accepted: 19-December-2025
Published: 23-December-2025

This article is published in the **MSI Journal of Medicine and Medical Research (MSIJMMR)**
ISSN 3049-1401 (Online)

The journal is managed and published by MSI Publishers.

Volume: 2, Issue: 12 (December-2025)

ABSTRACT: Esophageal atresia (EA) is a rare congenital anomaly characterized by esophageal discontinuity. Type A EA, the rarest subtype, occurs in approximately 1% of cases and is not associated with a tracheoesophageal fistula, making its diagnosis and management particularly challenging. This report highlights the successful management of a neonate with Type A EA, complicated by neonatal pneumonia and congenital heart defects, using awake intubation. A 4-day-old neonate weighing 1715 grams presented with respiratory distress, hypersalivation, and failure to pass a nasogastric tube beyond 8 cm. Diagnostic imaging confirmed Type A EA, while echocardiography revealed ventricular septal defect (VSD) and patent ductus arteriosus (PDA). Awake intubation was performed with ketamine induction and an uncuffed endotracheal tube to maintain spontaneous breathing and avoid airway collapse. Thoracotomy with primary esophageal anastomosis was successfully performed, despite two episodes of intraoperative desaturation, which were resolved by repositioning the head and re-evaluating the tube. Postoperative analgesia was managed with Fentanyl, and the

https://zenodo.org/records/18030983

patient showed stable recovery in the NICU. This case highlights the importance of early diagnosis, multidisciplinary care, and precise airway management for Type A EA. Awake intubation proved to be a safe and effective approach, ensuring optimal surgical outcomes.

Keywords: *Esophageal atresia, Type A, Awake intubation, Neonate, Airway management*

Abstrak

Atresia esofagus (EA) adalah kelainan kongenital yang langka, ditandai dengan diskontinuitas esofagus. EA tipe A, subtipe paling jarang, terjadi pada sekitar 1% kasus dan tidak disertai fistula trakeoesofageal, sehingga diagnosis dan tata laksananya menjadi tantangan. Laporan ini membahas keberhasilan penanganan seorang neonatus dengan EA tipe A yang disertai pneumonia neonatal dan kelainan jantung bawaan menggunakan teknik intubasi sadar (*awake intubation*). Seorang neonatus berusia 4 hari dengan berat 1715 gram datang dengan distres pernapasan, hipersalivasi, dan ketidakmampuan melewatkan selang nasogastrik lebih dari 8 cm. Pemeriksaan pencitraan mengonfirmasi EA tipe A, dan ekokardiografi menunjukkan defek septum ventrikel (VSD) serta duktus arteriosus persisten (PDA). Intubasi sadar dilakukan dengan induksi ketamin dan pemasangan selang endotrakea tanpa *cuff* untuk mempertahankan pernapasan spontan dan mencegah kolaps jalan napas. Torakotomi dengan anastomosis esofagus primer berhasil dilakukan, meski terjadi dua episode desaturasi intraoperatif yang dapat diatasi dengan reposisi kepala dan evaluasi ulang selang. Analgesia pascaoperasi diberikan dengan Fentanyl, dan pasien menunjukkan pemulihan stabil di NICU. Kasus ini menyoroti pentingnya diagnosis dini, perawatan multidisiplin, dan manajemen jalan napas yang cermat pada EA tipe A. Intubasi sadar terbukti aman dan efektif untuk hasil bedah optimal.

Kata kunci: Atresia esofagus, Tipe A, Intubasi sadar, Neonatus, Manajemen jalan napas

Introduction

Esophageal atresia (EA) is a rare congenital anomaly characterized by the discontinuity of the esophagus, often associated with a tracheoesophageal fistula

(TEF)[1,2]. This condition arises due to embryological defects during the fourth to sixth weeks of gestation, disrupting the separation of the trachea and esophagus[3]. The global incidence of EA is estimated at 1 in 3,500 live births[1]. In Asia, including Indonesia, data on EA prevalence are scarce, but the rate is believed to be similar to global estimates. A lack of robust registries may hinder precise epidemiological reporting in developing countries[4]. However, neonatal care advancements have improved infants' survival rates with EA worldwide. In Western countries, the survival rate for patients with EA without major congenital anomalies is nearly 100%. However, in low- and middle-income countries, the mortality rate remains relatively high (30-80%)[5].

The clinical presentation of EA typically becomes apparent shortly after birth. Common symptoms include excessive drooling, inability to feed, choking, cyanosis, and respiratory distress[6]. These manifestations are often exacerbated by associated conditions, such as prematurity or congenital anomalies like cardiac defects, which are present in up to 50% of cases[6]. Early recognition and diagnosis are crucial, as delays can lead to life-threatening complications, including aspiration pneumonia and severe malnutrition[7].

EA is classified into five types based on the presence and location of any associated TEF (**Image 1**). Type A, isolated EA without a fistula, accounts for approximately 1% of cases, making it the rarest form. Type B is characterized by a proximal esophageal segment communicating with the trachea. Type C, the most common variant, involves esophageal atresia with the distal segment forming a fistula to the posterior part of the trachea. This type accounts for over 90% of esophageal malformations. Type D represents a rare form of esophageal atresia, where both the proximal and distal segments communicate with the trachea. Type E is definied by an intact esophagus with an abnormal connection to the trachea, without esophageal disruption[6,8]. Type A's rarity presents unique challenges in diagnosis and management, as it lacks the clinical clues provided by an associated fistula, such as gastric distension caused by air entering the stomach via the fistula.

Definitive diagnosis of EA can be confirmed by the coiling of nasogastric tube in the chest and the absence of a gastric bubble on a chest X-ray[1]. Chest X-ray can also

define wether the EA is accompanied by fistule or not and diagnose other anomalies related to VACTREL syndrome[7].

The complications of untreated EA, particularly Type A, are severe and potentially fatal. These include aspiration pneumonia, respiratory failure, sepsis, and failure to thrive[9]. Postoperative complications, such as anastomotic stricture and gastroesophageal reflux, are also common and require long-term follow-up[10].

Nowadays, awake intubation is gaining popularity among clinicians and patients in need due to improvements in techniques and studies to provide good safety measures to the procedures and drugs related[23]. Awake intubation is a specialized technique used to secure the airway in critically ill patients without administering general anesthesia. This approach is especially relevant in neonates with Type A EA, where maintaining spontaneous breathing during intubation is vital due to the risk of airway collapse[1]. Indications for awake intubation include congenital airway anomalies, anticipated difficult airways, and situations where rapid desaturation is a concern. In experienced hands, awake intubation has a high success rate and is considered a safe option for neonates[6]. A recent study reported success rates exceeding 90% in neonates with challenging airways, emphasizing the importance of meticulous preparation and expertise[3].

This case report highlights the management of a neonate with Type A EA using awake intubation, showcasing its efficacy and safety. As the literature on managing Type A EA remains limited, this report aims to contribute valuable insights into the optimal approach to this rare condition, particularly in resource-limited settings.

Case Description

A 4-day-old neonate weighing 1715 grams was diagnosed with Esophageal Atresia Type A complicated by neonatal pneumonia and congenital heart disease, including Ventricular Septal Defect (VSD) and moderate Patent Ductus Arteriosus (PDA). The patient presented with respiratory distress since birth, accompanied by hypersalivation and coughing during feeding attempts. At the referring hospital, a trial of nasogastric tube (NGT) insertion was performed, but the NGT could only be advanced up to 8 cm. The patient was born via cesarean section (CS) due to

premature rupture of membranes (PROM). There was no maternal history of medication use or significant illness during pregnancy.

On physical examination, the patient had a patent airway and was breathing spontaneously with Continuous Positive Airway Pressure (CPAP). There was chest wall retraction, a respiratory rate of 42 breaths per minute, and oxygen saturation (SpO_2) between 88–94% under a CPAP setting of Positive End-Expiratory Pressure (PEEP) of 7 and Fraction of Inspired Oxygen (FiO_2) of 25%. Bilateral vesicular breath sounds were heard without rales or wheezing. The extremities were warm, the capillary refill time (CRT) was less than 2 seconds, and the heart rate was 175 beats per minute, which was regular, with a systolic murmur auscultated.

The patient was being treated for neonatal pneumonia with intravenous ampicillin (90 mg every 12 hours) and gentamicin (8 mg every 36 hours), as well as for congenital heart disease with intravenous furosemide (0.9 mg every 12 hours). Laboratory evaluations were within normal limits (**Table 1**). A preoperative chest X-ray revealed bilateral pneumonia, absence of air in the intestinal tract, and no visible abnormalities of the heart or abdomen (**Image 2**). Echocardiography findings confirmed VSD with overriding aorta (50%), pulmonary atresia, and tortuous PDA measuring 2 mm.

The patient was classified as ASA IV physical status based on the clinical findings. The Esophageal Atresia was categorized as Type A (Waterston C, Spitz II), with cyanotic congenital heart disease comprising VSD, overriding aorta (50%), pulmonary atresia, and PDA, as well as neonatal pneumonia.

The patient underwent thoracotomy with primary esophageal anastomosis under general anesthesia with awake intubation. Induction medications included ketamine 1 mg. Intubation was performed using an uncuffed endotracheal tube (ETT) size 2.5 at a depth of 8 cm. Analgesia during the procedure was provided with metamizole, and anesthesia was maintained with Oxygen. Several episodes of desaturation occurred during surgery (**Image 3B**), including two significant events. The first major desaturation was resolved by pausing the surgical maneuver and repositioning the patient's head. The second was managed by pausing the surgical maneuver, returning

the patient to the supine position, repositioning the head, and re-evaluating the ETT position with laryngoscopy. Hemodynamics remained stable throughout the procedure.

The surgery lasted 3 hours, with an estimated blood loss of 5 cc. Postoperatively, analgesia was continued using Fentanyl, and the patient was transferred to the Neonatal Intensive Care Unit (NICU) for ventilatory support.

Discussion

Esophageal atresia (EA) is a congenital anomaly in which the esophagus fails to develop properly, resulting in a discontinuity between the upper and lower esophageal segments[3]. This structural defect obstructs the passage of swallowed material into the stomach. Frequently associated with a tracheoesophageal fistula (TEF), EA poses risks of aspiration, choking, and recurrent pulmonary infections[11]. EA arises during embryonic development's fourth to sixth weeks when the esophagus and trachea which originate from the foregut were saparated[4]. Although the exact etiology remains unknown[3,7,12]. Genetic predispositions, combined with environmental factors such as maternal obesity, diabetes, or teratogenic exposure, are believed to play a role in the condition's occurrence. To date, evidence suggests that mutations in approximately 54 different genes can cause EA/TEF, and the condition has been linked to over 35 genetic syndromes.[12] Advances in neonatal surgery and critical care have significantly improved outcomes for affected infants.

The global incidence of EA ranges between 1 in 3,000 to 1 in 4,500 live births[6]. Although specific data from Indonesia is limited, studies from Southeast Asia report similar prevalence rates. A Malaysian study estimated the incidence at 1 in 3,500 live births, with potential underreporting in regions with less access to healthcare. Disparities in prenatal care and environmental exposures may also influence the prevalence of EA and other congenital anomalies. EA is slightly more common in males than females, as proven by several studies[10,13].

Because of the structural abnormality, EA presents with obstructive clinical manifestation such as excessive salivation, coughing, and respiratory distress within the first hours of life. Feeding attempts typically result in choking, regurgitation, and

cyanosis. The inability to pass a nasogastric tube into the stomach is a hallmark diagnostic feature. Additionally, up to 50% of EA cases are associated with other congenital anomalies, particularly within the VACTERL spectrum (vertebral defects, anal atresia, cardiac anomalies, TEF, renal abnormalities, and limb deformities)[3,7,14]. Heart defects are the most common anomalies associated with EA and TEF[15], with ventricular septal defect (VSD) being the most frequently observed type. The prevalence of congenital heart disease (CHD) is approximately 20% in cases of non-syndromic gastrointestinal malformations in children, and more than 60% in cases of syndromic gastrointestinal malformations. A study conducted in Iran found that 36.4% of children with EA had CHD, with 71% of them having VSD[16]. In this case, the patient presented with clinical manifestations including hypersalivation, dyspnea, cough, and difficulty in inserting a nasogastric tube, which strongly indicated EA. Additionally, this case was accompanied by heart defects, including VSD and patent ductus arteriosus (PDA). In this case, the patient exhibited bilateral pneumonia as well, a common complication caused by aspiration and impaired airway clearance.

The diagnosis of esophageal atresia (EA) can be made both prenatally and postnatally. Prenatally, a combination of polyhydramnios and the absence of gastric air on ultrasound may indicate EA. A systematic review analyzing 1,760 patients with EA found that 56% had polyhydramnios, and 50% displayed a small stomach bubble on prenatal imaging[17]. Although these findings are neither highly specific nor sensitive, they are frequently used to raise suspicion of EA during prenatal care[18]. Postnatally, EA typically manifests within the first few hours or days of life. Common symptoms include hypersalivation, coughing during feeding attempts, and episodes of apnea due to milk aspiration. The diagnosis can be confirmed through the placement of a nasogastric tube (NGT). If the tube coils in a dilated proximal pouch on chest X-ray, the diagnosis of EA is established[9]. In this case, the NGT inserted into the patient bent at the level of 8 cm, consistent with the diagnosis of EA[8]. Echocardiographic evaluation plays an important role in the assessment of patients with EA/TEF. As previously mentioned, EA/TEF is often associated with cardiac anomalies, making echocardiography essential to identify these anomalies, which are crucial in determining mortality and potential complications of surgical procedures[15].

EA can be classified into five main types based on the presence and location of a tracheoesophageal fistula. Type A or pure esophageal atresia, accounts for approximately 7-8% of cases and is characterized by a complete absence of fistulae. Type B involves a fistula between the upper esophagus and trachea, with a blind-ending lower esophagus, and is extremely rare (<1% of cases). Type C, the most common subtype (87%), presents as esophageal atresia with a distal TEF[7]. Type D includes fistulae connecting both esophageal segments to the trachea, and Type E (H-type TEF) involves an isolated tracheoesophageal fistula without esophageal atresia[15].

Type A EA's rarity makes it one of the most challenging subtypes to manage. Unlike other forms of EA, patients with Type A lack communication between the esophagus and trachea, precluding air entry into the gastrointestinal tract. As a result, infants often require staged surgical interventions, such as gastrostomy for nutritional support followed by delayed primary anastomosis or esophageal replacement. Early recognition and management are critical to optimizing outcomes, particularly in resource-limited settings where delayed diagnosis increases the risk of complications.

EA is associated with numerous complications that can occur before, during, or after surgical intervention. Preoperatively, affected neonates are at risk for aspiration pneumonia, recurrent respiratory infections, and feeding difficulties. Intraoperative challenges include achieving tension-free esophageal anastomosis, particularly in Type A cases with significant esophageal gap lengths. Postoperatively, common complications include anastomotic stricture, gastroesophageal reflux disease (GERD), recurrent fistula formation, and tracheomalacia. Pulmonary complications, such as bilateral pneumonia observed in this patient, highlight the importance of meticulous airway management and nutritional support during the preoperative period[19].

Awake intubation is a specialized airway management technique used in neonates with compromised airway anatomy or respiratory stability. Awake intubation is a technique in which the anesthesiologist places a tracheal tube while the patient is awake and breathing spontaneously[20]. Unlike traditional intubation under general

anesthesia, awake intubation preserves spontaneous breathing and minimizes the risk of hypoxia or airway collapse during the procedure. This approach is particularly advantageous in neonates with EA, who are at high risk for aspiration and respiratory distress. Awake intubation requires careful preparation, including the use of topical anesthesia and minimal sedation to maintain airway reflexes while reducing procedural discomfort[15]. The decision to perform awake intubation in this case was based on several factors. The patient's anatomical challenges, including Type A EA and bilateral pneumonia, necessitated cautious airway management to avoid exacerbating respiratory distress. Additionally, the patient's clinical stability relied on maintaining spontaneous breathing throughout the procedure. Securing the airway was critical for preoperative stabilization and to facilitate subsequent surgical repair of the esophageal defect.

Awake intubation involves several critical steps. Preparation begins with assembling all necessary equipment, including endotracheal tubes (ETT), laryngoscopes, and suction devices. In this case, an uncuffed ETT was used. Disma et al[21]. noted that both cuffed and uncuffed ETTs are suitable for neonatal intubation. However, when using a cuffed ETT, practitioners must carefully select the appropriate size to minimize air leaks and reduce the risk of post-extubation stridor. Standard monitoring should include an electrocardiogram (ECG), non-invasive blood pressure measurement, and peripheral oxygen saturation (SpO_2). The placement of the pulse oximeter must account for both preductal and postductal oxygen saturations[15].

Awake intubation relies on securing the patient's airway while maintaining spontaneous ventilation. Topical anesthetics, such as lidocaine, are commonly applied to the airway to minimize discomfort and suppress reflexes. Although awake intubation can be performed using local anesthesia alone, sedation is often used to reduce the patient's discomfort and improve cooperation during the procedure. Practitioners must exercise caution to avoid oversedation, which can lead to airway obstruction, respiratory depression, cardiovascular instability, or significant morbidity and mortality.

Several sedative options are available, with remifentanil and dexmedetomidine being top recommendations. Other agents, such as midazolam and ketamine, can also be

used, although they come with certain drawbacks[21]. In this case, ketamine was chosen due to the availability of relevant sedative agents.

While effective, awake intubation carries potential risks. Ventilation during neonatal intubation presents a real challenge for anesthesiologists, as hypercapnia and desaturation are common complications[15]. According to studies, adverse events occur in 18% of neonatal intubation cases, with 48% involving severe oxygen desaturation[22]. This can be caused by acute vital changes resulting from surgical manipulation, which may disturb the position of the endotracheal tube (ETT) and gas exchange. Therefore, it is crucial for the anesthesiologist and surgeon to maintain communication throughout the procedure. In this case, the patient experienced significant desaturation twice. During the first desaturation, communication was made with the surgeon to stop the maneuver while the anesthesiologist repositioned the head. The patient's saturation returned to normal afterward. During the second desaturation, communication was again made with the surgeon to halt the maneuver, the head was repositioned, and the ETT position was checked. The saturation normalized again. A similar case was reported by Marthendro et al., where a patient with TEF type C experienced desaturation during surgery with awake intubation. Desaturation was managed by repositioning the head, checking the tube, and ensuring adequate ventilation[11]. Another case report also described desaturation in a TEF type C patient during gastrostomy and esophagostomy. Desaturation was addressed with manual ventilation, PEEP administration, and communication with the surgeon to reduce retraction if desaturation was difficult to manage[6]. This case highlights the importance of maintaining ventilation and hemodynamics during surgery in cases involving awake intubation. Despite these challenges, awake intubation remains a valuable technique in managing neonates with complex airway anomalies, such as those observed in this case.

Following the surgical procedure, analgesia in this case was maintained using Fentanyl. Postoperative pain management can be delivered through various methods, including intravenous opioid infusion, epidural catheter, subcutaneous wound catheter, or local infiltration. Currently, there is no evidence suggesting that one technique is superior to another, with the choice typically depending on local

expertise and experience. Adequate analgesia is crucial for patient recovery. Additionally, some surgeons may request the patient remain muscle-relaxed for a period postoperatively, as they believe this helps reduce shearing or distracting forces on a tensioned anastomosis[13].

Conclusion

In conclusion, this case highlights the complexity of managing Type A esophageal atresia in a preterm neonate with associated complications, including bilateral pneumonia and cardiac anomalies. The rarity of this EA subtype necessitates a multidisciplinary approach involving neonatologists, surgeons, and anesthesiologists to ensure optimal outcomes. Awake intubation was pivotal in stabilizing the patient preoperatively, allowing for definitive surgical repair of the esophageal defect. This case underscores the importance of early recognition, referral to specialized centers, and individualized management strategies for neonates with rare congenital anomalies like Type A EA.

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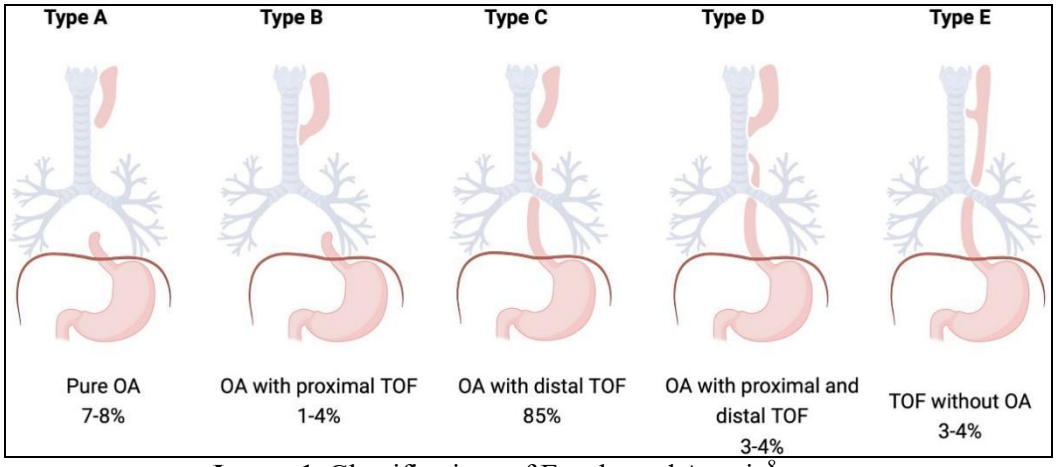

Image 1. Classifications of Esophageal Atresia[8]

Table 1. Preoperative laboratory results

Laboratory Evaluation Results	
Haemoglobin	15.4
Leukocytes	6.200
Thrombocytes	193.000
Erythrocytes	4.290
Hct	46
Na	113
K	4.4
Ca	0.67

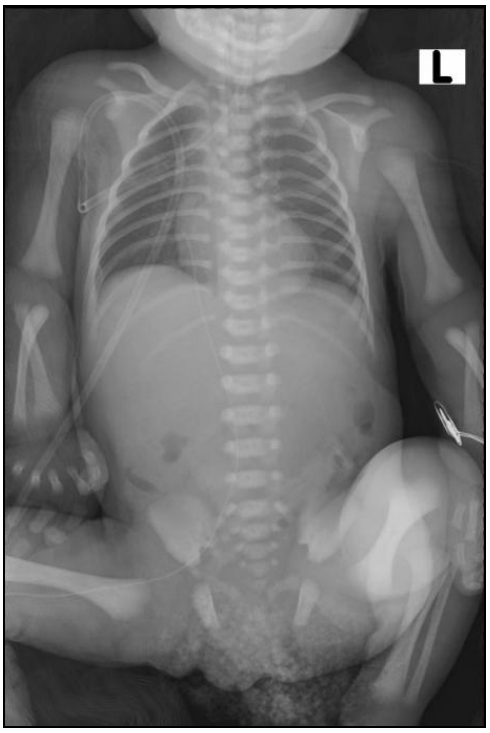

Image 2. Preoperative thoracic X-ray

Image 3. Patient's monitor evaluation; (A) Preoperative, (B) During operative, (C) Post Operative