International Journal of Contemporary Pediatrics
Basak A et al. Int J Contemp Pediatr. 2025 Feb;12(2):215-220
http://www.ijpediatrics.com
pISSN 2349-3283 | eISSN 2349-3291

DOI: https://dx.doi.org/10.18203/2349-3291.ijcp20250087

Original Research Article

Intestinal atresia: a retrospective study of 36 neonates and risk factors to mortality in Tertiary care center, Tripura

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Received: 06 June 2024
Revised: 04 July 2024
Accepted: 08 January 2025

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ABSTRACT

Background: Intestinal atresia among neonates is still a condition which has huge morbidity and mortality, particularly in the developing world. We share a case series of intestinal atresia in context of their presentation, management and outcome.
Methods: This study was conducted at Tripura Medical College and DR BRAM Teaching hospital, Tripura, over 1 year. We included all patients presenting with jejunoileal atresia, duodenal atresia and their demographic details, presentation, investigations, treatment strategies and the outcome were noted at a pre-designed proforma. All data were analysed using SPSS version 26.
Results: A total of 36 neonates with intestinal atresia were included. Most of them (77.78%) presented after 48 h of life and the mean age at presentation was 5.68±4.75 days. There were 22 male patients (61.11%) and 30 (83.33%) were full-term. The most common presenting complaint was not being able to pass meconium (88.89%). Type III atresia was the most common subtype (41.67%). Most of them underwent resection without tapering. The mean hospital stay was 12.81±6.53 and it was significantly longer among those who underwent re-exploration (P=0.034). 13 patients (36.11%) expired within 6 months of follow-up. The only significant factor for mortality was the presence of short bowel syndrome (P=0.036). All other demographic and management factors did not alter the mortality rate.
Conclusions: Management of surgical neonates is a difficult job in developing countries with limited resources. There is a high mortality rate of neonates following intestinal atresia surgeries and surgeons in these countries must fight on many fronts to improve the outcome.

Keywords: Duodenal atresia, Intestinal, Jejuno-ileal atresia, Neonates

INTRODUCTION

An atresia is a congenital defect of gastrointestinal tract that results in absolute obstruction of the lumen. Atresia of intestine is most frequent cause of bowel impediment in the newborn and can occur at any point in the gastrointestinal tract. The ileum is the most commonly affected site. Jejunoileal atresia (JIA) has a prevalence rate of 1:1000 live births, with a third of the infants being either born prematurely or small for date. JIA is a pertinent condition in neonates with<1% of babies with chromosomal or associated anomalies, however, there are much more varieties and much more surgical options in the case of JIA. It makes JIA a bit more complicated because of a variety of choice of surgical options.[1-3] Most jejunoileal atresia's or stenosis results from localized intrauterine vascular insult to the developing bowel with ischemic necrosis and subsequent reabsorption of the affected segment. Also, it is a challenging situation in developing and poor countries because of lack of resources, delayed presentation of neonates and lack of nursery intensive care unit (NICU) facilities.[4,5] We aimed to present our experience of neonates presenting with

JIA, duodenal atresia and determine the prognostic factors leading to mortality in our setup.

METHODS

Study type

This was a retrospective observational study.

Study place

This study was conducted at Tripura Medical College and DR BRAM teaching hospital (TMC), after approval of the Ethical review board. TMC is the largest centre for pediatric and neonatal surgery in the state of Tripura and the main referral centre from all over the state.

Study duration

The duration of the study was 1 year from January 2022 to December 2022.

All the neonates who underwent surgery for JIA, DA were included in the study. JIA was labelled as perioperative findings narrated by the surgeon. We did not include the other causes of small bowel obstruction like meconium ileus, volvulus without JIA or total colonic Hirschsprung's disease. Also, those who expired before any intervention were not included in the study.

Preterm was defined as gestational age< 37 weeks and low-birthweight was labelled if weight was<2500 g. Mortality was defined as demise within 30 days after the surgery. All the patients were initially resuscitated and after optimization, surgery was planned. We retrospectively went through the record of the operating room and collected all the cases of JIA and DA who had been operated during this period.

The charts of these patients were retrieved and all details were collected. The demographic details, presentation, investigations, treatment strategies and outcome were noted at a pre-designed proforma. All data were analysed using SPSS version 26. We calculated percentages for categorical variables and mean (SD) continuous variables. The odd's ratio (OR) was calculated for dependent variable (mortality) using binary logistic regression analysis.

RESULTS

A total of 36 neonates presented with JIA and DA in the study duration. 8 neonates (22.22%) presented in the first 48 h of life, while 28 neonates (77.78%) presented later. The mean age at presentation was 5.68±4.75 days and the age range of presentation was 1–25 days of life. Most of them (22, 61.11%) were males and 14 (38.89%) were female. 30 neonates (83.33%) were full-term and 6 (16.67%) were pre term. 22 neonates (61.11%) were born through the vaginal route and 14 (38.89%) were born through C section. 8 neonates (22.22%) had low birth weight (<2500 g) and 28 (77.78%) had birth weight>2500 g. In 1 patient (3.2%), JIA had been diagnosed antenatally and in 35 (97.22%) antenatal scan was normal. Regarding the presentation, the most common presenting complaint was not being able to pass meconium 32 (88.89%), followed by abdominal distention 28 (77.78%) and bilious vomiting 22 (61.11%). On per rectal examination, most of them (60, 95.2%) passed only mucous.

Figure 1: Pre-operative X-ray of a neonate suffering from proximal ileal atresia.

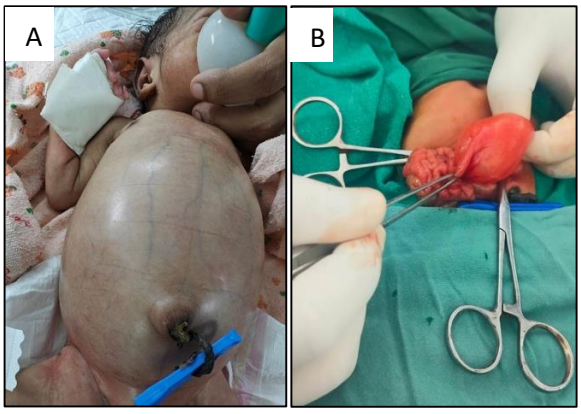

Figure 2 (A and B): Pre-operative and intra operative pictures of the neonate with proximal ileal atresia.

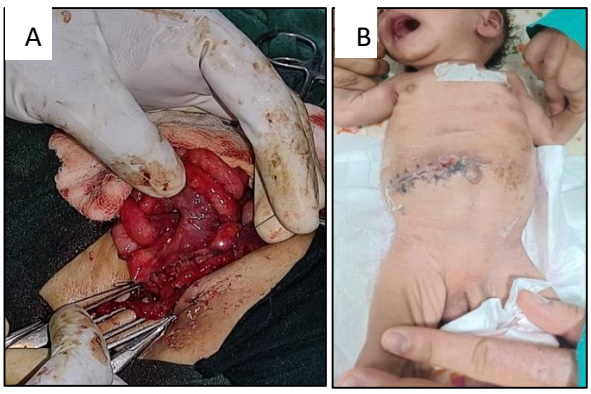

Figure 3 (A and B): Intra-operative tapering anastomosis and post operative picture of the neonate.

Further, contrast enema identified microcolon in 16 patients (44.4%) and in remaining neonates, it was not significant. All these neonates underwent exploratory laparotomy. The most commonly found type of JIA was type III atresia (15, 41.67%), followed by type 1 8 (22.22%), type II 7 (19.44%) and type IV 6 (16.67%). Among 2 patients, the volvulus of the intestine was also present along with JIA. The most commonly opted procedure was anastomosis of both portions of the gut without tapering (13, 36.11%), followed by tapering enteroplasty and anastomosis of the gut (9, 25%), stoma formation (ileostomy) 8 (22.22%) and chimney creation procedures i.e., bishop koop 4 (11.11%) and Santulli (2, 5.56%). Per-operatively, 9 patients (25%) were found to have short bowel and they were specifically advised to start total parenteral nutrition (TPN) (Table 1). The mean hospital stay was 12.81±6.53 days, with the range being 5–35 days. 4 patients (11.1%) needed a re-exploration due to any reason. Regarding the mortality within 30 days, 13 patients (36.11%) expired and 23 (63.89%) are alive (Table 1). Also, we tried to look for the factors which may lead to mortality among these patients. The only significant factor was the presence of short bowel syndrome (OR 3.643, p=0.036). Mortality was higher among patients where tapering enteroplasty or chimney procedure was opted. The age of presentation later than 48 h of life, male gender, type of atresia, associated volvulus or hospital stay had no significant effect on mortality (p=0.870). All these details are summarized in table 2.

Table 1: Demographic details, presentation, management and outcome of neonates in this study.

Age at presentation	
Within 48 h	8 (22.22%)
After 48 h	28 (77.78%)
Gender	
Male	22 (61.11%)
Female	14 (38.89%)
Gestational age	
Preterm	6 (16.67%)
Full-term	30 (83.33%)
Mode of delivery	
Vaginal route	22 (61.11%)
Cesarean section	14 (38.89%)
Weight	
<2500 g	8 (22.22%)
>2500 g	28 (77.78%)
Antenatal scan	
Yes	1 (3.2%)
No	35 (97.22%)
Presentation	
Bilious vomiting	22 (61.11%)
Abdominal distension	28 (77.78%)
Inability to pass meconium	32 (88.89%)
Surgical procedure opted	
ETEA without resection	13 (36.11%)
Tapering enteroplasty with ETEA	9 (25%)
Ileostomy	8 (22.22%)
Bishop-Koop procedure	4 (11.11%)
Santulli procedure	2 (5.56%)
Type of atresia	
Type I	8 (22.22%)
Type II	7 (19.44%)
Type III	15 (41.67%)
Type IV	6 (16.67%)
Short bowel syndrome	
Yes	9 (25%)
No	27 (75%)
Outcome	
Expired	13 (36.11%)
Alive	23 (63.89%)

Table 2: Logistic regression for factors leading to mortality.

Variables	Number of patients with mortality (%)	OR: (95% CI): P value
Age at presentation		
Within 48 h	3/8	1.08 (0.212-5.494)
After 48 h	10/28	0.926
Gender		
Male	8/22	1.029 (0.255-4.156)
Female	5/14	0.969
Gestational age		
Preterm	2/6	0.864 (0.135-5.508)
Full-term	11/30	0.876
Mode of delivery		
Vaginal route	8/22	1.028 (0.254-4.156)
Caesarean section	5/14	0.967
Surgical procedure opted		Reference
ETEA without resection	5/13	
Tapering enteroplasty with ETEA	4/9	0.971:(0.222–4.243):0.968
Ileostomy	2/8	1.833:(0.392–8.566):0.441
Bishop-koop procedure	1/4	3.667:(0.557–24.132):0.177
Santulli procedure	1/2	
Type of atresia		Reference
Type I	3/8	
Type II	2/7	0.227: (0.040–1.299):0.096
Type III	6/15	0.595: (0.114–3.102):0.538
Type IV	2/6	0.441: (0.105–1.854):0.264
Short bowel syndrome		
Yes	6/9	5.714 (1.118-29.207)
No	7/27	0.036
Volvulus		
Yes	1/3	0.458 (0.036-5.789)
No	12/33	0.546
Re-exploration		
Yes	3/6	2.000 (0.34-11.756)
No	10/30	0.4431

DISCUSSION

In developing countries, delayed presentation and mortality are quite common in surgical neonates. In this series, 77.78% of the neonates presented after 48 h of life. Authors have reported similar conditions from Nigeria, who reported delayed presentation in 63.2% of cases.[6] Another study from Ethiopia reported 72% of cases having delayed presentation with GI atresia.[7] It is a different situation in developed countries, where authors report an early presentation.[8,9] There are many reasons for this delayed presentation. First and foremost are the healthcare facilities for mother and child in developing countries. Most of the children are born in the peripheral centres without optimal neonatal care and once they are referred to us, it is already more than 48 h passed.[6,7] Gender distribution was almost equal in both genders in this study. No particular gender propensity has been noted in JIA in previously reported reports from Nigeria and Spain.[6,8] An antenatal scan was done in only 1 patient (3.2%) in this series and only his parents knew about the condition prenatally. Similar proportions have been narrated from other developing countries. However, in developed countries, it has been reported in up to 86.6% of cases.[8] Virgon C et al, conducted a meta-analysis over the data and reported the accuracy of prenatal ultrasound for the diagnosis of JIA varying 10–100%, with an overall prediction being 50.6%. Also, they narrated that the accuracy for diagnosing jejunal atresia is higher than ileal atresia (66.3% vs. 25.9%).[10]

Although ultrasound machines are available everywhere in Pakistan, the proportion of these conditions being diagnosed is very low. A recent huge population-based study concluded that almost 50% of the women visit healthcare workers during pregnancy two or fewer times.[4] There may be various reasons for this problem. Most probably, there is a lack of trained radiologists and at most of the centres, gynecologists or technicians are performing antenatal scans. These people are not trained

to diagnose JIA and other GI anomalies.[11] Also, most centres have poor resolution probes and old machines, which probably cannot pick up these conditions.[6] We found that the most common type of JIA was type III, followed by type I, type II and type IV. Most of the previous reports have narrated similar findings.[6,12]

However, one centre had reported type II being most common type.[13] Another study from Pakistan had also reported type III as the most common subtype.[12] Tis variability in the frequency among types of JIA highlights that globally, it varies from one area to another. It may be another important area of future studies to investigate this aspect and determine the reason for this variance. An important aspect in the management of surgical neonates in developing countries is the high mortality rate. In this series, we found mortality in 36.11% of cases.

When we analysed the data of JIA cases and their mortality, we found it 34.2%, 25%, 33%, 37% from developing countries.[6,7,14,15] However, in developed countries, it is clearly less than 10%.[8] The obvious reason for high mortality is increased complication rate, higher wound infection rate, sepsis, delayed presentation of the neonates, lack of NICU services and TPN and hypothermia. Another report from Pakistan narrated the mortality in 42.5% of cases with GI atresia.[12] Chaudhry et al, reported complications in 40.8% of all children presenting with intestinal obstruction. These higher numbers show the conditions of the facilities and the level of care being provided.[5]

The outcome can be improved with the provision of peri-operative anesthesia services, good post-operative care, trained NICU staff, good nutritional service and a team-based approach to these cases.[16] The only variable which had significant effect on mortality was the presence of short bowel syndrome. The mean hospital stay in patients with short bowel syndrome was 16.6+7.67 days and 11.63+5.71 days (P 0.009) in those without SBS. All of the patients with SBS were started with TPN within the hospital. As we do not have the facility for home TPN, so usually, patients are discharged from the hospital, once they are tolerating oral feed and are stable.

As mentioned, that in our setup, it makes a lot of sense because of poor provision of resources and no facility to provide home-based TPN. Because of these scarce resources, almost all such patients succumb to death and have no good outcome. Another interesting finding was that those who underwent some chimney procedure (like Bishop-koop or Santulli), had higher mortality. Most of the literature shows a better outcome with chimney formation procedures.[17,18] We went through the charts of these patients to find out the reason, but could not find any pertinent reason for this phenomenon in our patients. However, we noticed that most of these procedures were opted when level of atresia was too proximal in the jejunum. We do not understand the reason for this observation; nonetheless, we speculate that it may be because of complications of a stoma, which may be too proximal in the jejunum. Another similar finding has been shared recently where authors found higher short-term and long-term complications among those who underwent stoma than those who underwent anastomosis.[19] Although a major limitation of this case series was being a single-centre report, it highlights the situation and difficulties we observe in managing JIA in our setup. It also highlights a high mortality rate. We need to focus on the peri-operative care and provision of nutritional services to these neonates to improve the outcome.

CONCLUSION

The overall survival rate amongst newborn babies suffering from intestinal atresia has improved from a dismal 10% in 1952 to 90% at present. This is because of the change in surgical procedure from primary resection anastomosis without resection to liberal resection of blind proximal and distal ends followed by end to end anastomosis. Atresia still carries a mortality of 19% predominantly due to gangrene of proximal end of the distal segment (7%), anastomotic leak (15%) and stricture formation (15%). Post-surgical complications like anastomotic leak, stricture formation, temporary intestinal dysfunction, blind loop syndrome and short bowel syndrome can be minimised by careful attention to the anatomical detail, meticulous surgical procedure and maximal bowel preservation procedures. Survival rate of 46%-70% can be expected in most infants with less than 25 cm jejunoileum ideally. Full bowel adaptation takes 6-18 months to become accomplished.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: The study was approved by the Institutional Ethics Committee

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Cite this article as: Basak A, Sil A, Sarkar M. Intestinal atresia: a retrospective study of 36 neonates and risk factors to mortality in Tertiary care center, Tripura. Int J Contemp Pediatr 2025;12:215-20.