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ORIGINAL ARTICLE

# Disparity in access and outcomes for emergency neonatal surgery: intestinal atresia in Kampala, Uganda

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**Abstract**
*Background/aim* Intestinal atresia is one of the leading causes of neonatal intestinal obstruction (NIO). The purpose of this study was to analyze the presentation and outcome of IA and compare with those from both similar and high-income country settings.
*Patients and methods* A retrospective review of prospectively collected data from patient charts and pediatric surgical database for 2012–2015 was performed. Epidemiological data and patient characteristics were analyzed and outcomes were compared with those reported in other LMICs and high-income countries (HICs). Unmet need was calculated along with economic valuation or economic burden of surgical disease.
*Results* Of 98 patients, 42.9% were male. 35 patients had duodenal atresia (DA), 60 had jejunio-ileal atresia (JIA), and 3 had colonic atresia. The mean age at presentation was 7.14 days for DA and 6.7 days for JIA. Average weight for DA and JIA was 2.2 and 2.12 kg, respectively. All patients with DA and colonic atresia underwent surgery, and 88.3% of patients with JIA had surgery. Overall mortality was 43% with the majority of deaths attributable to aspiration, anastomotic leak, and sepsis. 3304 DALYs were calculated as met compared to 25,577 DALYs' unmet.
*Conclusion* Patients with IA in Uganda present late in the clinical course with high morbidity and mortality attributable to a combination of late presentation, poor nutrition status, surgical complications, and likely under-reporting of associated anomalies rather than surgical morbidity alone.
*Level of evidence* Level IV, Case series with no comparison group.

**Keywords** Neonatal surgery · Pediatric surgery · Uganda · Low- and middle-income countries · Disparities · Intestinal atresia

**Abbreviations**

| | |
|---|---|
| LMIC | Low- and Middle-Income Countries: defined by the World Bank as countries with a Gross National Income per capita, calculated using the World Bank Atlas, less than or equal to $4035 |
| HIC | High-Income Countries: defined by the World Bank as countries with a Gross National Income per capita greater than or equal to $12,476) [1] |
| NIO | Neonatal intestinal obstruction |
| IA | Intestinal atresia |
| JIA | Jejuno-ileal intestinal atresia |
| DA | Duodenal atresia |
| NICU | Neonatal intensive care unit |
| TPN | Total parenteral nutrition |
| DALYs | Disability adjusted life years |
| YLLs | Years of life lost |
| YLDs | Years of life lived with disability |

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## Introduction

Intestinal atresia is often cited as one of the most common causes of neonatal intestinal obstruction (NIO). Specifically, in low- and middle-income countries (LMICs), it ranks as the third most common cause of NIO after anorectal malformations and Hirschsprung's Disease [2–4]. The incidence of jejunal–ileal atresia and stenosis has been reported to be between 0.9 and 30 in 10,000 live births. [5].

In high-income countries (HICs), the management of intestinal atresia has greatly improved with reported mortality of approximately 4.6–11% from 30 to 50% as reported in historical controls in the literature [6]. The improvement in mortality in high-income countries (HICs) is felt to be associated with prenatal diagnosis, early presentation postpartum, improved availability of total parenteral nutrition (TPN), and access to a neonatal intensive care unit (NICU) [7].

The prevalence, management, and mortality rate for neonatal intestinal obstruction due to intestinal atresia in LMICs, however, have remained poor with mortality reported as high as 15–30% in Nigeria [4]. No studies to date have described the experience with intestinal atresia in Uganda. In this study, we set out to analyze the burden of intestinal atresia, current management strategies, and outcomes at a single center in Kampala, Uganda. Mulago Hospital is a government run 1700 bed tertiary care facility founded in 1913 as a National Referral hospital for the entire country as well as the local tertiary hospital for the Kampala metropolitan area [8].

## Methods

A retrospective review of prospectively collected data on pediatric surgery patients was reviewed. The database was reviewed for patients with a diagnosis of intestinal atresia from 1st January 2012 to 30th August 2015. The pediatric surgical database was started in 2012 to collect a variety of epidemiological data and outcomes for pediatric surgical patients. The database includes information such as date of birth, date of admission, approximate gestational age, gender, village, tribe, approximate distance traveled to reach referral hospital, mode of referral, chief complaint, surgical intervention, timing from admission to surgery, and more (see Table 1 for all data collected). These data were then compared to historical data identified through literature review. Additional chart review was performed to evaluate variables not included in the database.

Data analysis was performed using the SPSS software package (version 20.0, SPSS Inc, Chicago, IL). Data were analyzed using the $X^2$ test for categorical data with Fisher's exact test, Student's, and non-parametric $t$ tests for applicable continuous variables. Two-tailed $p$ values less than 0.05 were considered statistically significant. Data are summarized using mean and standard deviation for normally distributed variables and median for non-normally distributed continuous variables. Prior to prospective data collection, this study and the associated database were approved by the Mulago Hospital Institutional Review Board. We also attest that the human research from which this manuscript is derived is in compliance with the Helinski Declaration.

Burden of disease was calculated using disability adjusted life years (DALYs) as outlined in the Global Burden of Disease report and associated studies [9]. Disease incidence was calculated, as in our previous study, with sub-Saharan Africa references where available [10, 11]. Incidence used for GI atresias is 1 in 2860 to give an estimated annual incidence of 556 based on the current estimate population of approximately 36 million and live birth rate of 42.5 per 1000 people in Uganda [12]. DALYs for any given health condition may be described as the sum of years of life lost (YLLs) and years of life lived with disability (YLDs) [9]. For a neonatal condition such as intestinal atresia, where surgery provides a full cure and absence of surgery leads to death, DALYs are equal to YLLs which are equal to life expectancy for that country, 59 years in Uganda [13]. Averted DALYs refer to the death and disability prevented by surgical intervention, while avertable DALYs' quantify the correctable death and disability if surgical capacity were optimal, assuming that in

**Table 1** Data points collected per surgical patient, if available

| |
|---|
| Patient ID number |
| Data entry date and date of completion |
| Parent name, if available |
| Patient name, if available |
| Estimated date of birth, date of admission |
| Age in days, months, years on admission |
| Sex |
| District, village, tribe and distance to referral hospital |
| Mode of referral |
| Primary complaint on admission |
| Duration of symptoms in years, months, weeks and days if available |
| Final diagnosis, disease code |
| Surgical management including date of procedure, complications |
| Outcome, mortality |
| Duration of hospitalization post operatively and total length of hospitalization |
| Clinical notes if available |

**Table 2** Patient characteristics

| | Total, *n* (%) | Duodenal atresia, *n* (%) | Jejunal–ileal atresia, *n* (%) | Colonic atresia, *n* (%) |
|---|---|---|---|---|
| Number of patients | 98 | 35 (35.7%) | 60 (61.2%) | 3 (3%) |
| Sex | | | | |
| Male | 42 (42.9) | | | |
| Female | 56 (57.1) | | | |
| Mean age at presentation | | 7.1 days (1–9) | 6.7 days (1–11) | n/a |
| Average weight at time of presentation | | 2.2 kg (1.4–2.8) | 2.1 kg (1.25–2.8) | n/a |
| Overall mortality | 42 (43%) | 8 (10%) | 33 (55%) | 1 (33%) |

the absence of intervention, mortality would be 100% [14, 15].

Total need, therefore, may be expressed in DALYs and divided into three categories: met need, unmet need, and unmeetable need for surgical care [16]. Unmet need for many neonatal conditions involves both YLL for completely untreated patients and YLD as a consequence of incomplete or inadequate treatment. For intestinal atresia, however, this was calculated solely from YLL, since all of these patients would have died without treatment. DALY calculations were performed as Mulago Hospital was essentially the sole provider of neonatal surgeries in the country during the study period. Disease survival rates were calculated from estimates of mortality from HIC as the best possible outcome of neonatal surgical care. The DALY calculation for intestinal atresia, therefore, is as follows:

$$\begin{aligned}\text{Met needs} &= \text{YLLs}\\ &= \text{Life expectancy}\\ &\quad \times \text{number of patients discharged (survived)}\\ &= 59 \times 56\\ &= 3303.7\end{aligned}$$

$$\begin{aligned}&\text{Unmet need}\\ &= (0.5 \times (\text{expected number of patients with}\\ &\quad \text{disease} - \text{number of patients discharged})\\ &\quad \times \text{life expectancy} \times \text{disease survival})\\ &= ((566-56) \times 59 \times 0.85)\\ &= 25,576.5.\end{aligned}$$

## Results

### Patient demographics

For the research time period of 1 January 2012–30 August 2015, 98 patients were seen at Mulago Hospital with intestinal atresia. Of these patients, 42 were male and 56 were female. Duodenal atresia was reported as the primary diagnosis in 36% of patients, jejuno-ileal atresia in 61%, and colonic atresia in only 3% (Table 2). The mean age at presentation for patients with duodenal atresia was 7.14 days compared to 6.7 days for jejunal–ileal atresia ($P = 0.73$). The average weight at time of presentation was 2.2 kg for patients with duodenal atresia compared to 2.12 kg for patients with jejunal–ileal atresia. 80 patients (81.6%) had jaundice at time of presentation with indirect hyperbilirubinemia; 15 of these patients required phototherapy. 76 patients (77.6%) were found to have abnormal renal function with elevated urea.

Nearly, 43% of the patients with duodenal atresia had an associated anomaly. Though not considered a separate but associated anomaly, 57.1% of patients were found to have an annular pancreas. Of patients with an associated anomaly, 19.0% had a congenital heart defect diagnosed on echocardiogram and physical exam and 23.8% with associated Down's syndrome. In contrast, only three patients with jejunal–ileal atresia (5.0%) were found to have minor external anomalies: two with supernumerary digits and one with a periauricular skin tag.

### Prenatal work-up

Of the patients included in this study, 61.2% of mothers attended antenatal services more than once. All of the mothers who received antenatal services had an ultrasound performed, most of which were performed by an ultrasound technician as opposed to a certified radiologist. Of these 60 women, intestinal atresia was suspected in only 5 (8.3%). On chart review, history suggesting polyhydramnios was identified in 66% of mothers, but only documented by ultrasound in 35%.

### Surgical intervention

Surgical records were reviewed for all patients diagnosed with intestinal atresia. Of patients with duodenal atresia, all underwent surgical intervention with an open diamond-type duodenoduodenostomy with or without transanastomotic feeding tube placement (Figs. 1, 2). For patients with

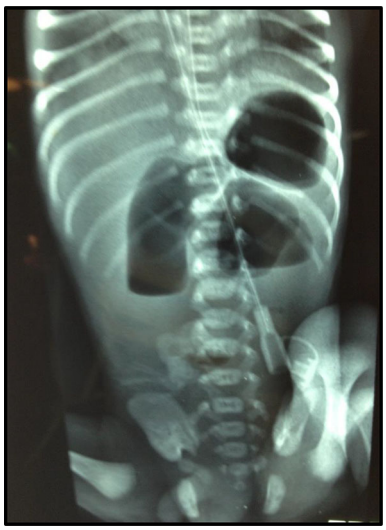

**Fig. 1** Plain film demonstrating "triple" bubble sign in child with abdominal distension, bilious emesis

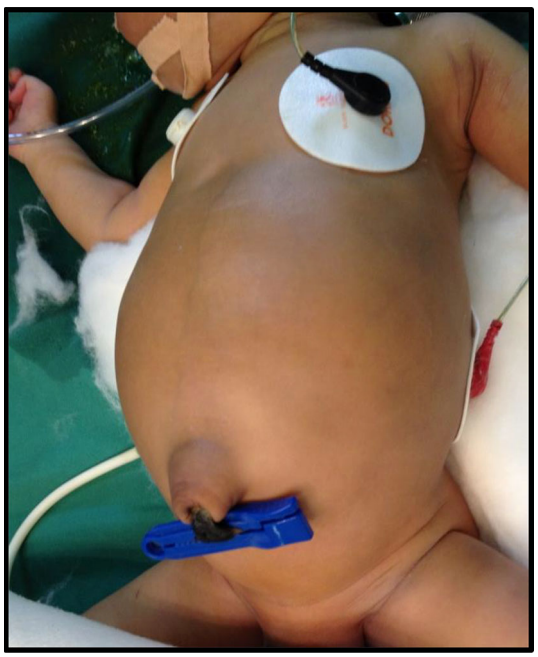

**Fig. 2** Abdominal distension in a child presenting with bilious emesis

jejunal–ileal atresia, 53 patients (88%) underwent surgery with resection of the dilated proximal bowel end, tapering enteroplasty and enteroenterostomy with interrupted, usually 5–0, absorbable sutures. During the course of the study, pediatric and adult surgical instruments were available secondary to donation by visiting surgeons. A transanastomotic feeding tube was placed when possible (Fig. 3). Procedures were all done by the licensed specialist pediatric surgeon(s) on-site and there were no procedures that were aborted intraoperatively for technical reasons. The remaining neonates with jejunal–ileal atresia were diagnosed post mortem as they were all very ill and died preoperatively. Of the patients with jejunal–ileal atresia, the majority were found to have type I (25%) followed by type II, type IIIb, type IV, and type IIIa (Table 3). All patients with colonic atresia underwent surgical intervention. All patients were taken to the general ward for routine post-operative care consisting of nasogastric tube for aspiration, intravenous fluids, and resumption of enteric feeds when possible. The mean duration from presentation to surgery was 2.2 days (range 1–8 days) for duodenal atresia and 3.4 days (range 2–6 days) for jejunal–ileal atresia).

### Outcomes

The overall mortality for this series was 42.9% (42 patients). For duodenal atresia, 8 deaths were reported (22.9% of patients with duodenal atresia) compared to 33 deaths for jejunal–ileal atresia (55.0%) which is statistically significant ($P < 0.001$). One death was reported for colonic atresia (33.3%). Reoperation was reported in 4.3% of patients, three having diagnosis of duodenal atresia, and one with jejunal–ileal atresia. All reoperations were performed for anastomotic leak. Primary reported cause of death for patients who underwent surgical procedures included aspiration, septicemia, and severe malnutrition. No patients died intraoperatively or within 3–4 days of procedure. Further details regarding cause and timing of death and complications were not available. Using mortality rates in the Western literature as the best achievable outcome in the HIC setting, the unmeetable need for intestinal atresia is 816.2 as determined in previous study [10]. Assuming that Mulago Hospital is the country's only provider of neonatal surgical services, 3304 DALYs of neonatal surgical need were calculated as met compared to 12,543 DALYs' unmet (Table 4).

## Discussion

Intestinal atresia remains a common cause of neonatal intestinal obstruction. In HICs such as The Netherlands, the US, and Canada, mortality has significantly improved from a historic rate of 35–50 to 4.6–11% [6, 17, 18]. Unfortunately, poor outcomes and mortalities as high as 28–50% in various series from Nigeria and Nepal prevail in LMICs (Table 5) [19, 20]. Advancements in surgical technique and more importantly, timely access to surgical services, availability of intensive care, and the widespread use of parenteral nutrition are largely responsible for the improvement in survival of neonates with intestinal atresia [10]. The results of this study, not surprisingly, may be

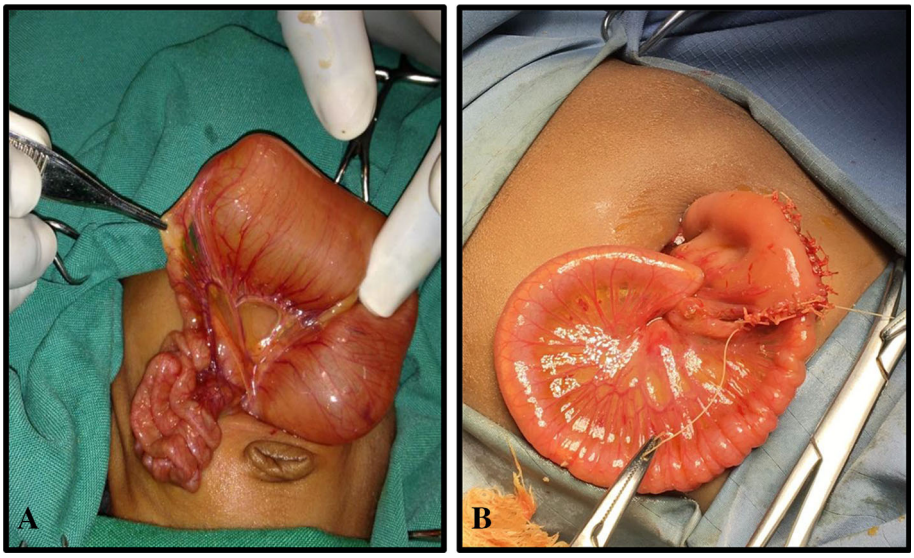


**Fig. 3** Jejunoileal atresia before (**a**) and after surgical repair with hand sewn anastomosis and placement of transanastomotic feeding tube (**b**)

**Table 3** Type of atresia

| Type of atresia | Jejunal–ileal atresia ($n = 60$) |
|---|---|
| I | 22 (36%) |
| II | 15 (25%) |
| IIIa | 5 (8%) |
| IIIb | 12 (20%) |
| IV | 6 (10%) |

attributed to any number of these factors felt to be critical to improved patient survival.

In this study population, a higher mortality rate was observed amongst patients with jejunal–ileal atresia as opposed to duodenal or colonic atresia. Based on the incidence of associated non-gastrointestinal congenital anomalies, namely, cardiac, in patients with duodenal atresia, one might suspect a higher mortality in this group. Factors associated with increased risk of death in duodenal atresia include complex cardiac anomalies, prematurity, sepsis, pneumonia, and surgical complications [21]. Cardiac anomalies are the most likely to go undiagnosed in this patient population and may contribute to late mortality, an area in need of further research. Alternatively, patients with DA and complex non-gastrointestinal congenital anomalies may be underrepresented in this cohort if they fail to present to the surgical facility.

One theory to explain the unexpectedly higher mortality amongst patients with JIA compared to DA is difference is the need to perform a tapering enteroplasty for JIA to account for the size mismatch rather than a simple, end to end or diamond anastomosis. Even when a resection of a bulbous end of a proximal atresia was possible, size mismatch still required tapering enteroplasty to achieve an end to end anastomosis. Previous studies describing this technique for tapering enteroplasty, however, reported increased rates of anastomotic leak associated with JIA compared to DA [22, 23]. It has been hypothesized that the increased mortality associated with JIA is more complex than technical considerations. Patients with JIA have been found to have a higher rate of multiple atresias as well as other associated gastrointestinal abnormalities [23, 24]. Patients with JIA, though not demonstrated in this study secondary to small sample size, may also present later as feeding intolerance may not be as immediately apparent as in DA.

## Access to specialty care

At the most basic level, access to adequate surgical services is a tremendous barrier to improvements in morbidity

**Table 4** Annual burden of disease nomenclature and calculations

| Summary | Definition | Unit | Calculation | Result |
|---|---|---|---|---|
| Surgical Burden | The overall burden of disease in the absence of surgical intervention | DALY | Cumulative (averted + avertable + unavertable) | 16,663.2 |
| Met need | The death and disability prevented by surgical intervention | DALY averted | Survival × life expectancy | 3304 |
| Unmet need | The correctable death and disability if surgical capacity was optimal | DALY avertable | Death × life expectancy | 12,542.8 |

**Table 5** Intestinal atresia patient characteristics by country

| Country | Author | Year | Study period | Methods | Number of patients, % | | | | Mortality (%) | Age at dx |
|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | Total | DA | JIA | CA | | |
| Nigeria | Chirdan et al. [2] | 2004 | 1989–2002 | Retrospective chart review | 24 | 20.8% | 70.8% | 8.3% | 41.7 | 6 |
| Nigeria | Ezomike et al. | 2013 | 2007–2012 | Retrospective chart review | 23 | 43.5% | 56.5% | 0 | 21.7 | 10.4 |
| Eastern Nepal | Shakya et al. [20] | 2010 | 2004–2008 | Retrospective chart review | 28 | n/a | 100%[a] | n/a | 28.5 | n/a |
| Netherlands | Stollman et al. | 2008 | 1971–2004 | Retrospective case series | 114 | n/a | 100%[b] | n/a | 11 | n/a |
| Netherlands | Festen et al. | 2002 | 1980–1992 | Multicenter review | 15 | n/a | 100%[c] | n/a | 20 | n/a |
| Canada | Burjonrappa et al. | 2010 | 1982–2007 | Retrospective chart review | 130 | 46% | 41% | 1% | 11 | n/a |
| New South Wales, Australia | Walker et al. [39] | 2008 | 1992–2003 | Cohort study | 286 | 53% | 47% | n/a | 8 | n/a |

[a] Only assessed jejunal–ileal atresia. 39.3% of atresia and stenosis in jejunum; 60.7% in ileum

[b] Only assessed jejunal–ileal atresia. 62% of atresia and stenosis in jejunum; 30% in ileum; 8% classified in both jejunum and ileum

[c] Apple peel atresia only. 47% single atresia, 53% multiple atresias (up to 4)

and mortality of pediatric surgical patients. By the American Pediatric Surgery Association standards of one pediatric surgeon per 100,000 children, approximately 200 pediatric surgeons would be required in Uganda [10, 25, 26]. During at least two-thirds of this study's duration, there was one pediatric surgeon practicing in Uganda and the majority of neonatal surgical care was provided at Mulago Hospital. Since the time Badrinath, et al. calculated the unmet need in 2014, two additional pediatric surgeons have joined the workforce in Uganda with a residual deficit of roughly 197 pediatric surgeons. In addition to a paucity of surgical specialists, there were no dedicated pediatric anesthesiologists in Uganda until early 2014. Though no comparative or causative data are available for this cohort of patients, other reviews have reported a substantial reduction in neonatal surgical mortality associated with the introduction of pediatric anesthesiology as a specialty [27].

Burden of disease was previously calculated for intestinal atresia as one of the six most common conditions encountered at Mulago Hospital for pediatric surgical patients using disability adjusted life years (DALYs) with methods outlined in the Global Burden of Disease report [10]. As previously described in the Global Burden of Disease study, DALYs for any given health condition are the sum of years of life lost (YLLs) plus years lived with disability (YLD) [9]. For a neonatal condition, where surgery provides a full cure and absence of surgery leads to death, as is the case with intestinal atresia, DALYs are equal to YLLs or the country's life expectancy. Unmet need or avertable burden is calculated from survival rates in HIC and incidence of disease. Even when accounting for difficulties with access to care and a high post-operative mortality, intestinal atresia care in Uganda currently has a met need of 3304 DALYs. The unmeetable need of 816 DALYs is significantly smaller than the unmet burden of neonatal atresia in Uganda of 12,543 DALYs [10]. Based on the fact that intestinal atresia has 100% mortality without surgical intervention and negligible disability following surgical repair, a profound economic benefit to repair is anticipated making congenital neonatal diseases an important area of attention.

## Nutrition supplementation

In addition to pediatric specialists, the need for nutritional therapy is a commonly cited limiting factor for pediatric conditions throughout LMICs. Tube feeds have been offered in some conditions though data on its availability and feasibility in resource-limited settings remains uncertain. The delivery of parenteral nutrition, as has become the standard of care in HICs, is limited by central venous access, availability of standard solutions and facilities for aseptic preparations, ability to perform necessary blood tests for monitoring, and cost of management of complications [28]. In this study, there were no patients receiving parenteral nutrition. During the time of data collection, more frequent use of transanastomotic feeding tubes was established in Kampala, Uganda, attempting earlier return to enteral nutrition by feeding distal to the area of repair. Moving forward, it is our recommendation that, when supplies are available, transanastomotic feeding tubes be

the standard of care. Given the short duration of this study and increased use of these tubes throughout the study, it is not possible to compare the population and associated mortality before and after the routine use of these feeding tubes. Though not specific to intestinal atresia, other studies have shown improved outcomes and intestinal function associated with early enteral feeds in both HICs and LMICs, especially in an otherwise well, full-term patient [29, 30]

### Incidence and prenatal diagnosis, referral

This study represents one of the largest of its kind with 98 patients in just under 4 years compared to 9 patients in 2 years, 22 patients in 19 years, and 23 patients in 5 years in previous Nigerian reports [3, 4, 20]. Based on limited access to prenatal care, it is unlikely that a larger volume of patients identified in this study as compared to historical controls is attributable to improved surveillance and diagnosis. It may, however, be associated with high birth rates in Uganda with the average number of births per woman ranging from 5.8 to 7.0 compared to 1.8 in the United States [31, 32].

In this series, only 35% of patients who underwent antenatal ultrasound were diagnosed with polyhydramnios and even fewer, and 8.3% of patients, who had an ultrasound, had a suspected diagnosis of intestinal atresia. In HICs, maternal death rates have shown a steady decline from 1990 to 2015 in line with the Sustainable Development Goals as part of the Millennium Development Goals. Though a decline was observed in some LMIC, the rate differs dramatically between countries [33]. In a recent study in Nigeria, a country with similar rates of intestinal atresia and comparable mortality to Uganda, 60% of women with recent live births received antenatal care from a skilled provider. The socioeconomic disparity was also evaluated as only 25% of the poorest women received antenatal care compared to 94% of their wealthiest counterparts [34]. More coordinated antenatal care may improve early referral if a bowel obstruction is suspected, but remains practically difficult given so few mothers access antenatal care.

Progress has been made in early identification and referral for children born with readily apparent congenital deformities such as cleft palate and club foot deformities. Specifically, in Tanzania, Nyamtema et al. demonstrated potential for improved maternal and neonatal outcomes with training of providers and enhanced rural referral infrastructure [35]. In Uganda, the Global Clubfoot Initiative and Christian Blind Mission have established and helped fund multifaceted programs for early identification and management of clubfoot and cleft lip/cleft palate [36, 37]. Similar results have been demonstrated with other visible congenital anomalies that are recognized by medical attendant or midwives present at time of delivery. In the case of intestinal atresia, early recognition and proper referral pose a unique challenge with delay causing detrimental effects on patient outcomes. Without a visible anomaly, the presenting symptoms of abdominal distension and food intolerance are presumed to be attributable to a number of causes, many of which would not necessitate urgent transfer to a facility with radiographic and surgical capabilities. Increased awareness of IA as a cause of neonatal intestinal obstruction and outreach programs for first line health workers are an essential first step in improving patient outcomes. As diagnostic studies and patient transfer may be financially debilitating for a family, the introduction of screening protocols for patients with recurrent vomiting or bilious emesis within the first 48 h of life may help guide more cost-effective resource utilization and patient transfer.

Efforts towards improved education and engagement of community surgical providers and pediatricians as well as nurses working on the patient wards have led to an anecdotal increase in prenatal and early diagnosis. Through ongoing assessment and characterization of patients and outcomes, it is anticipated that the rural referral infrastructure will continue to evolve for neonates born with any number of surgical conditions. With these advances, management of neonatal intestinal atresia has the potential to join the ranks of caesarean deliveries, laparotomy, and open fracture management as a Bellwether procedure, a proxy for surgical systems that have the ability to provide a broad range of procedures [38]. Previously, gastroschisis has been cited as a potential condition to represent pediatric surgery as a Bellwether procedure. While both conditions provide essential insight into the capacity of a health care system, the avertable mortality associated with intestinal atresia in the presence of neonatal surgical care may make it a more suitable metric.

Despite some perceived improvements in diagnosis and management of neonatal intestinal obstruction, the mean age at time of presentation in this study was 7.1 days for duodenal atresia and 6.7 days for jejunal–ileal atresia. This is comparable to the reported age of presentation in Nigerian series [2, 4] but statistically much later than age of presentation in HICs [6, 39]. Overall length of stay was comparable for patients with different locations of atresia similar to study by Piper et al. though this was considerably shorter in our study group compared to that in HICs as reported in the literature [18]. The patients with increased length of stay in Piper et al.'s study primarily included patients with documented associated congenital anomalies. When compared to HICs, it is likely that the shorter length of stay is associated with absence of parenteral nutrition which may, if employed routinely, delay initiation of

enteral feeds, additional work-up and intervention for associated anomalies, and other routine testing and neonatal care. In the present study, it is suspected that a proportion of associated anomalies were undiagnosed based on the already limited diagnostic capabilities and neonatal care at Mulago Hospital.

### Limitations

This study has a number of important limitations which may affect its broader application. While the sample size is large compared to similar studies, it remains small and may grossly underestimate the burden of disease in Uganda by capturing only patients who arrive at Mulago Hospital. This limitation also likely underestimates the mortality of patients who do not arrive at a referral center or who remain undiagnosed. This study is also limited by the absence of long-term follow-up for the majority of patients. As noted, many patients travel great distances to reach Mulago Referral Hospital for neonatal care. For patients who are discharged to home, follow-up with the surgical team is inconsistent at best. Additional follow-up with pediatricians and other specialists is even more challenging to ascertain. This is especially true in the case of patients with duodenal atresia and associated cardiac anomalies in a severely resource-limited setting.

A small number of private hospitals within Uganda offer neonatal surgical care, representing a likely small unaccounted for case load. For the private hospitals and regional hospitals managing patients not referred to Mulago Hospital, it is conceivable that neonates might have been treated by general surgeons. Because of this, values for met need are likely a slight underestimate, while unmet need may be a slight overestimate. This is felt to be of minimal significance, however, given the absence of other university-based hospitals with the capacity for management of intestinal atresia at the time of the study. In addition, the general surgeons operating in private or other public facilities anecdotally deny performing operations for atresias and other congenital illnesses. The calculations utilized accepted population-based estimates of incidence for anomalies, not accounting for possible variation across populations [40].

Another limitation, which is commonly encountered when performing research in under-resourced environments, is in the limited available data. While data collection improved throughout and just prior to this study, the deficits make the results' generalizability a bit limited. In particular, more detailed data around cause of death and timing of complications were not available. Since completion of this study, there has been a significant effort to develop and maintain a more detailed database of pediatric illness throughout Africa.

## Conclusion

In conclusion, this study represents one of the largest series of intestinal atresia in an LMIC and reports a disproportionately high volume of diagnosed intestinal atresia in Uganda with persistently low survival in Uganda, as in many LMICs, despite advances in surgical management and care in HICs. Unfortunately, many challenges to adequate neonatal and antenatal care are likely associated with the high mortality for children born with intestinal atresia. This study supports the urgent need for improvement in neonatal care facilities, trained personnel, and resources to improve outcomes.

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