Original Article

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Attrition rate among patients of Anorectal Malformations on colostomy: A matter of concern

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KEYWORDS

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ABSTRACT

Background: Anorectal malformations (ARM) are correctable congenital malformations with good prognosis. Traditionally it is managed by staged procedure, with diversion colostomy, followed by definitive procedure and stoma reversal in the third stage. It is recommended to complete all stages of repair by 6 months of age for better long-term continence. We had fewer patients coming for definitive surgery and stoma reversal than the patients undergoing colostomy for ARM, so we planned this study to find out the attrition rate among patients undergoing stoma for ARM and explore the reasons for the same.

Methods: An observational study was conducted in the Pediatric Surgery department of Chacha Nehru Bal Chikitsalaya, New Delhi. All the case records of patients with ARM who underwent diversion stoma at our center from January 2018 to December 2019 were retrieved. Further follow-up case records were retrieved with the same Unique Health Identification Number (UHID). When the definitive surgery/ stoma closure was not found in the hospital records till December 2022, the parents were contacted telephonically to find out if the children had undergone definitive surgery at some other center, were awaiting surgery, or had died. The attrition was calculated as the difference between the number of patients for whom a stoma was done and the number of patients who underwent definitive surgery and stoma closure till the data acquisition.

Results: A total of 105 patients were included in the study with male preponderance. Colostomy was done for 73.33% of patients in the neonatal period (77/105). The cloacal malformation was found in 7 (6.67%), rectal atresia in 6 (5.71%), and 9 had congenital pouch colon (8.57%). The age at stoma creation for ARM ranged from 1 day to 9.3 years (mean 0.67 ±1.96 years). Cardiac and renal anomalies were among the major associations. 28 of the neonates (26.67%) had stomal complications. Stomal issues were reported in 32.4%. Mortality was reported in 37 patients (35.24%) including 35 neonates. The attrition rate in our study was found to be 38.24% (26 out of 68). Factors contributing to attrition range from anemia and protracted waiting lists to the anesthesiologist's pursuit of optimal patient stabilization, alongside familial or personal matters faced by caregivers.

Conclusion: There was a high attrition rate in operated patients of ARM in whom stoma was constructed. The main reasons for the same were anemia and a long surgical waiting list.

INTRODUCTION

Anorectal malformations (ARM) are correctable congenital malformations with good prognosis. Most of the international guidelines recommend it to be managed in a staged manner, with diversion colostomy at birth, a definitive procedure around 3 months of age, and followed by stoma reversal 2-3 months later. However, in developing countries, the actual time of the procedures can seldom be complied with delayed surgery and restoration of bowel function. The delay adversely affects the long-term prognosis by preventing neuronal networks from being trained during infancy due to fecal diversion from the sphincter complex. Furthermore, many children are prevented from their fundamental right to a normal fecal passage due to various social, economic, and other factors. There is no study in English literature recording the attrition of patients undergoing stoma. The hidden mortality in these patients is either not recorded or recorded as lost to follow-up. In Indian society with its gender bias, while raising a child, delayed diagnosis due to inaccessible healthcare at delivery (and newborn screening for

gross congenital anomalies) in rural and semi-urban populations and the economic conditions forcing parents not to provide care to their children adequately, this hidden mortality and morbidity is likely to be high. In this study, we planned to find out the attrition rate in these patients and explore the reasons for the same.

METHODS

A hospital-based observational study was conducted in the Pediatric Surgery department of Chacha Nehru Bal Chikitsalaya, New Delhi.

The institutional ethics committee's approval for the study was taken with approval no. F.1/IEC/CNBC/18/01/2022/Protocol no. 102/266.

The list of patients undergoing stoma for ARM at our center from January 2018 to December 2019 was obtained from the operation theater case register. All the files included in the study were retrieved from the Hospital Information Management System and the continuation of treatment was recorded.

Patients for whom the record of a second or third surgery was not found were contacted via telephone. During these conversations, after obtaining verbal consent for participation in the study, inquiries were made to determine whether the child had undergone additional surgeries at another center, was awaiting surgery, had passed away, or was unable to attend follow-up appointments. Additionally, reasons for the inability to continue follow-up at our center were requested.

Attrition encompassed patients who underwent stoma procedures at our center but did not proceed to further surgeries (excluding those who passed away). Patients who underwent the second, third, or both stages of surgery at another hospital were excluded from attrition calculations. Those under follow-up but unable to undergo surgery due to reasons such as anesthesia unsuitability or COVID-related restrictions were deemed attrition cases. Additionally, patients unreachable via phone were considered part of attrition. Attrition was calculated as the disparity between the number of ARM patients who underwent stoma procedures and those lost to follow-up.

The data were compiled on a Microsoft Excel sheet®. The demographic profiles were calculated as numbers and percentages. The attrition rate was calculated as the ratio.

RESULTS

In the study period, a total of 105 patients diagnosed with ARM underwent stoma creation at our center and were included in the study. Among them, 62 were male, resulting in a male-to-female ratio of 1.4:1. The majority of patients were in the neonatal period (82 out of 105, 78.10%). Seven girls (6.67%) were diagnosed with cloacal malformation, with five having cloaca in conjunction with congenital pouch colon (CPC). Among males, four had CPC, while one had rectal atresia. Additionally, five female neonates were diagnosed with rectal atresia. Twenty patients had completed all three stages of surgery, including two with pouch colon. Tragically, thirty-five patients (33.33%) undergoing stoma creation passed away before definitive surgery (pull-through) could be performed.

Overall, the age at stoma creation for ARM ranged from 1 day to 9.3 years. The mean age of stoma formation was 0.67 ±1.96 years. The mean age at PSARP and stoma reversal were 1.72 ±1.94 years and 2.61 ±2.44 years, respectively. Fifteen neonates (18.29%, 15/82) had stomal complications including 10 wound dehiscence/burst abdomen, 2 stomal retractions/stenosis, and 1 prolapse. Thirteen neonates (15.85%, 13/82) suffered from stomal diarrhea while 2 (2.44%, 2/82) had stoma bleeding causing anemia. Overall stomal complications were reported in 32.4% (34/105).

The associated anomalies included cardiac and renal in 15 patients (14.29%) each, vertebral in 10 (9.52%), Down syndrome, uterovaginal anomalies, Urethral anomaly (Hypospadias/ Megalourethra) and undescended testis (UDT) in 5 each (4.76%), followed by cleft palate in 4 (3.81%) and microtia in 3 (2.86%). There were multiple stomal complications including diarrhea in 12 patients (11.43%), stoma wound dehiscence in 8 (7.62%), stomal retraction in 5 (4.76%), while stomal prolapse and adhesive intestinal obstruction in 3 each (2.86%). There was a stoma bleed causing concern to parents in 2 and stoma closure leaks in 2 patients.

The overall mortality rate was 35.24%, totaling 37 patients. Among them, 35 patients passed away after stoma creation (33.33%), one following pull-through surgery (before stoma reversal), and one after stoma closure. The causes of death were attributed to sepsis in 26 cases, cardiac issues in 3 cases, acute kidney injury (AKI) in 2 cases, a complication from stoma closure leakage in 1 case, sudden infant death syndrome (SIDS) in 1 case, and in 4 cases the cause remained unknown.

Twenty-one patients lacked recorded follow-up admissions in our Hospital Information Management System (HIMS) and could not be reached via telephone. Additionally, several patients remain on our waiting list, with surgery postponed due to specific reasons: anemia in 8 patients, a lengthy waiting list in 7 patients, an anesthesiologist's discomfort due to upper respiratory infection (URI) in 4 patients, and family issues (such as ill parents or grandparents) in 4 patients each. Two patients were unable to attend follow-up appointments due to

distance, while 1 patient's surgery was postponed due to cardiac issues. The attrition rate at our center is 38.24% (26 out of 68).

DISCUSSION

The incidence of ARM is estimated to be 1:2000 to 1:5000 live births [1]. A three-staged procedure, consisting of colostomy creation at birth, definitive pull-through around 3 months, and stoma closure before 6 months, has been recommended for high-type anorectal malformations in both sexes [2,3]. Freeman suggested that early placing the bowel in the sphincter stimulates brain-evoked potentials and is beneficial for long-term outcomes [4]. Albanese suggested that the staged procedure causes an undue delay in restoring bowel continuity, which could hamper the potential development of neuronal networks and synapses, along with the training of perineal muscles for near-normal bowel function [5]. In our institute, due to a long waiting list, completing all three procedures during infancy is rare.

High ARM has male preponderance and the same could be seen in our study [6]. The age of presentation varied from neonatal period to toddlers to pre-adolescents in various studies [6-8]. Delayed presentation is common in developing countries for reasons like social, long distance, education status, initial wrong/inadequate information by the primary caregiver, etc. [8,9].

Stoma-related complications are prevalent among children, with reported incidences ranging from 27% to 70% [7,10,11]. These complications encompass various issues such as colostomy prolapse, stenosis, retraction, excoriation, para colostomy hernia, stomal dysfunction, mislocation, bleeding, malnutrition, wound infection, and complications related to stoma closure. In our study, thirty-four patients experienced stoma-related complications, including 14 patients with stomal bleeding and diarrhea. The neonatal stoma complication rate in our study was 26.7%, consistent with findings in the literature [10,11]. Interestingly, the overall stoma complication rate was lower in our study compared to previous research [7].

In a study from India, the complication rates increased when primary PSARP was done later (mean 15 month's age) as compared to PSARP in girls under colostomy coverage [12]. We prefer to perform PSARP under stoma cover when the definitive surgery is performed after infancy.

The mortality rate associated with stomas has been reported to range from 0% to 10% [7]. In our study, although direct mortality related to stoma could not be definitively established, the high mortality rate may be partly attributed to stomal complications, poor nutrition, inadequate hygiene, and lack of education. The overall high mortality observed in our patients was primarily attributed to sepsis, which may have been indirectly related to factors such as a high volume of patients, delayed presentation, and complications arising from stomas.

In a report from North India, only 52% of patients completed the three stages [13]. They attributed the attrition either to mortality associated with associated malformations, infective or nutritional complications associated with colostomy, or social reasons and disinclination to continue treatment [13]. In our study, only 19.05% (20 out of 105) of patients completed all three stages of treatment. The reasons for attrition within our study group were multifaceted: mortality accounted for 35.24%, while loss to follow-up and non-traceability contributed to 20% of cases. Anemia was a factor in 7.62% of cases, followed by a long waiting list (6.67%), the anesthesiologist's preference for better stabilization due to frequent upper respiratory tract infections or associated cardiac anomalies (4.76%), and personal reasons of parents hindering follow-up (5.71%). Additionally, in private centers where patients are required to pay for services, 30.6% of parents cited an inability to afford treatment due to insurance denial, lack of funds, or insufficient family support [14].

CONCLUSION

There was a high attrition rate in operated patients of ARM with a stoma. The main reasons for the same were poor socio-economic background, anemia, and long surgical waiting list. This study highlights the need for early surgery or primary PSARP as soon as possible to avoid potential morbidity and mortality.

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