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Pediatric Intussusception and Interventional Radiology in a Developing Country: Experience and Challenges of Ultrasound Saline Reduction Complementary to Primary Surgery

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Nonoperative reduction is considered a safe and effective treatment procedure for pediatric intussusception. However, the procedures are yet to be adopted in some developing countries. This study evaluates our experience with ultrasound saline reduction of pediatric intussusception in southeast Nigeria. Retrospective analysis of 58 children managed for intussusception from June 2009 to May 2013 at the University of Nigeria teaching hospital, Enugu. The median age at presentation was 7 months (IQR = 5 to 9 months), and the average duration of symptoms before diagnosis was 3.9 days (range, 1 to 14 days). Thirteen (22.4%) had saline hydrostatic reduction initially (11 succeeded, 2 failed), and a total of 47 cases had operative treatment (including the 2 with failed saline reduction). Operative procedures were right hemicolectomy in 24 cases, manual reduction 18, ileal resection 3, and colonic resection 2. Eighteen (38.3%) cases developed postoperative complications, and there were 2 postoperative deaths from unremitting septicemia. Comparison of the cases that had saline reduction with cases that underwent operative treatment showed a difference in the average time to diagnosis, need for preintervention transfusion, onset of oral intake, and average duration of admission. Challenges in the saline reduction procedure were delayed presentation, insufficient facilities and trained personnel. Ultrasound saline reduction procedure for pediatric intussusception is feasible in our setting. Despite the delay in presentation in most of our

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cases, this procedure may still be applied initially to patients who do not have clinical or radiologic contraindications.

Key words: Intussusception – Hydrostatic reduction – Outcome – Challenges – Developing country

Intussusception is the invagination of a portion of the intestine into the lumen of the immediately adjoining part. Approximately 90% of pediatric intussusceptions are primary or idiopathic.[1,2] Regardless of the etiology or the type, intussusception is composed of apex, internal, and returning middle layer (intussusceptum), and outer layer (intussuscipiens).[1] The preintervention diagnosis is radiologic, and currently, ultrasonographic demonstration of target sign, doughnut sign, or pseudokidney sign has diagnostic specificity rates of 88% to 100% and sensitivity rates of 98% to 100%.[3–5] Definitive treatment involves reduction of the intussusception through retrograde pressure on the apex. Prior to the advent of pressure reduction methods, definitive treatment was undertaken exclusively through operation.[6] However, presently the standard of care in most developed countries is air insufflation or saline hydrostatic enema, with surgery reserved for cases that are complicated at presentation or failed the nonoperative reduction procedures.[3,4,7–9] The enema reduction methods have been found to be safe, simple and effective.[7–10]

In many developing countries, substantial challenges have been reported to preclude the adoption of the nonoperative treatment methods.[11–13] In this setting, operative intervention for all pediatric intussusception has been routine. However, from 2009 we commenced ultrasound-guided saline reduction at the University of Nigeria Teaching Hospital (UNTH), Enugu, in southeast Nigeria.

This study evaluates our experience with saline reduction of pediatric intussusception. The focus is on comparison of patients managed by this procedure with those managed by operative intervention, and determining the challenges in ultrasound saline reduction in our setting.

Patients and Methods

At UNTH, Enugu, children with clinical suspicion of intussusception are referred from the adjoining primary and secondary health facilities in the southeast region of Nigeria. On presentation the patients are evaluated and resuscitated with intravenous fluid, electrolytes, and blood transfusion as appropriate. Diagnosis is confirmed by ultrasonographic finding of target, pseudokidney, or doughnut signs. Prior to 2009, all cases with radiologic confirmation of intussusception were managed definitively by operative intervention. From June 2009 following exposure and training of one of the consultants on nonoperative pressure methods, some of the patients with intussusception were managed by ultrasound guided saline reduction. Due to the fact that some of the surgeons prefer routine operation for intussusception, and we do not have sufficient number of ultrasound machine and available trained personnel, this method was not routinely applied. The patients who were selected for this procedure were those who presented to the unit that undertake the procedure within 3 days of onset of symptoms, and who did not have clinical or radiologic evidence of peritonitis, bowel perforation, or pathologic lead point. The rest of the patients were managed by laparotomy and, depending on the findings, either manual reduction or bowel resection were undertaken. For the cases managed by saline reduction, the procedure is undertaken in the radiology department following the confirmation of diagnosis and informed consent by the parents. The patient is placed in supine position and a size 20F Foleys catheter is passed into the rectum, and the balloon inflated with 30 to 40 mL of water. Less amount of fluid was used to inflate the balloon in younger and smaller patients. The anus is tightened around the catheter by pressure on the buttocks. Warm normal saline is run in from a drip bag positioned 1 meter above the patient and the flow and reduction is monitored with the ultrasound. In some cases, the saline bag is elevated above the 1-meter height to maintain positive pressure. Complete reduction is confirmed by the free flow of the saline into the small intestine and the disappearance of the target or the other radiological signs. Failure of reduction will warrant emergency operative treatment.

From June 2009 to May 2013, a total of 58 children were managed for intussusception at UNTH. These cases were retrospectively analyzed. From the case notes, theatre records and discharge summaries, the

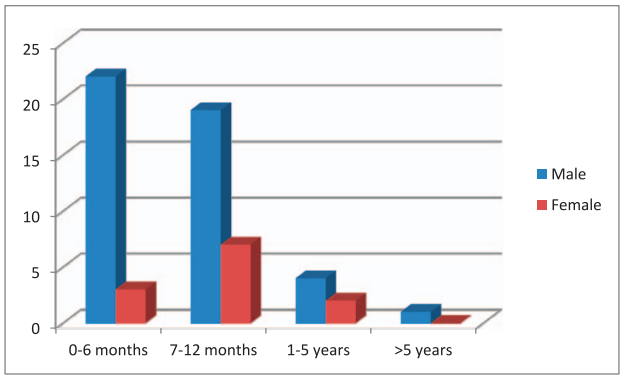

Fig. 1 Age and sex distribution of the 58 cases of pediatric intussusception.

following data were collected: age at presentation, gender, time from onset of symptoms to presentation, clinical presentation, ultrasound findings, need for pre-intervention transfusion and or electrolyte correction, definitive treatment, and reason for operative treatment. The other data collected include findings at operation, operative procedure, onset of oral intake, postoperative complications, duration of admission, follow-up duration, and documented challenges in the management of the patients.

Data Analysis

Statistical package for Social Sciences (SPSS 15.0 version, SPSS Inc, Chicago, Illinois) was used for data entry and analysis. Results were expressed as percentages, or mean. Data were analyzed by chi-square test or student *t* test as appropriate. In all, the significance level was set to $P < 0.05$.

Results

Of the 58 cases, 46 (79.3%) were male and 12 (20.7%) female. The median age at presentation was 7 months (IQR = 5 to 9 months). Figure 1 shows the age and sex distribution of the cases. The main clinical features are summarized in Table 1 and the duration of symptoms before presentation in Table 2. The average duration of symptoms before presentation to our center was 3.9 days (range, 1 to 14 days). Ultrasonography finding was target sign in 49 (84.5%) cases and pseudokidney in 9 (15.5%) cases. There was no demonstrable lead-point, or interloop fluid in any of the cases on ultrasonography.

Table 1 Main clinical features among the 58 children with intussusception

Clinical features	Frequency (%)
Abdominal pain	55 (95)
Rectal bleeding	41 (70.7)
Vomiting	39 (67.2)
Abdominal distension	21 (36.2)
Abdominal mass	44 (75.9)
Features of peritonitis	8 (13.8)

Table 2 Summary of the duration of symptoms before presentation among the 58 cases of intussusception

Duration of symptoms	n	%
1 to 2 days	14	24.1
3 to 5 days	36	62.1
>5 days	8	13.8
Total	58	100

Overall, 13 (22.4%) cases underwent saline hydrostatic reduction (11 were successful; 2 failed) and 47 (including the 2 with failed saline reduction) had operative treatment.

Patients managed by saline reduction

In this category, there were 9 (81.8%) males and 2 (18.2%) females with an average age of 9.9 ± 5.2 months at presentation. The cases had symptoms for one to three days (mean, 1.8 days) before presentation. Of these patients, 2 (18.2%) required pretreatment electrolyte correction for hypokalemia, and 2 (18.2%) required blood transfusion. Overall, 11 (84.6%) had successful reduction of the intussusception after 1 attempt and 2 (15.4%) had failed reduction. Following saline reduction, oral intake was commenced within 24 hours, and the average duration of admission was 2.4 days (range, 2 to 3 days). There was no recurrence of intussusception after a follow-up period of 2 to 5 months (mean, 3 months).

Patients managed by operative intervention

There were 47 patients managed by operative intervention. Of these, 37 (78.7%) were male and 10 (21.3%) female. Their age at presentation ranged from 3 months to 8 years (mean, 10.1 months), and they had symptoms for average duration of 4.17 days (range, 2 to 14 days) before presentation to our center. For the 29 cases that presented after 3 days of symptoms the reason documented for the delay was delayed referral in 18 cases and parents' ignorance

Table 3 Summary of the reasons for operative treatment and findings at operation

Reason for surgery	n (%)	Finding at operation		
		Reducible intussusception	Irreducible intussusception	Gangrenous intussusception
Surgeon's preference	25 (53.2)	13	12	-
Late presentation	12 (25.5)	5	7	-
Suspected peritonitis	8 (17.0)	-	1	7
Failed saline reduction	2 (4.3)	-	2	-
Total	47	18	22	7

of the problem in 11 cases. In managing these patients, 27 (57.4%) cases required preoperative electrolyte correction for hypokalemia, and 25 (53.2%) received blood transfusion. The reasons for undertaking operative treatment in these patients and the operative findings and procedure are summarized in Tables 3 and 4 respectively. Following operative treatment, 18 (38.3%) patients developed the following postoperative complication: surgical wound infection (10), prolonged ileus (3), incisional hernia (3), anastomotic leak (1), and adhesive small bowel obstruction (1). There were 2 deaths (mortality of 4.3%) from unremitting septicemia. The average duration of follow-up was 3.4 months (range, 2 to 8 months).

Comparison of patients managed by saline reduction and those managed by operative treatment is summarized in Table 5.

Discussion

The definitive treatment for pediatric intussusception has evolved from routine operative intervention to predominantly enema reduction.[1–3] The current standard of care in the more developed countries is enema reduction, with surgery reserved for cases that are complicated at presentation or failed the nonoperative reduction procedures.[7–10] Our study has shown that many of our cases were referred at an advanced stage and some cases were older than 1 year old, hence our setting may have a selection towards difficult to treat patients and therefore high incidence of primary surgery. Nevertheless, several patients who were referred at an earlier stage were clinically considered suitable for hydrostatic reduction that was frequently successful in these cases (11/13 = 85%). Therefore in a developing country, hydrostatic reduction may be a valuable complementary method in addition to standard surgical treatment.

Previous studies indicate that enema reduction techniques for pediatric intussusception have undergone significant advances in the past decades.[4,7,8] Presently barium enema reduction is no longer recommended due to significant morbidity from barium peritonitis that can accompanies perforation,[10] and the methods in use include fluoroscopic-guided pneumatic reduction or water soluble iodinated contrast enema, and sonographic-guided saline or Hartmann's solution hydrostatic reduction.[4,6–9,14–16] In the present report, the sonographic-guided saline hydrostatic reduction

Table 4 Findings at operation and the operative procedures among the 47 children who underwent operative treatment for intussusception

Operative procedure	Total	Indication/ finding (n)
Right hemicolectomy	24	Irreducible ileocolic intussusception (17) Gangrenous ileocolic intussusception (7)
Manual reduction	18	Ileocolic intussusception (17) Colocolic intussusception (1)
Ileal resection	3	Irreducible ileoileal intussusception (3)
Colonic resection	2	Irreducible colocolic intussusception (2)

Table 5 Summary of the comparison of patients managed by saline reduction and patients managed by operative treatment

Parameter	Saline reduction n = *11*	Operative treatment n = *47*	*P*
Average duration of symptoms	1.8 days	4.2 days	0.001*
Mean age at presentation	9.9 months	10.1 months	0.96
Need for preintervention transfusion	2/11	25/47	0.036*
Onset of oral intake	1 day	3.5 days	0.000*
Mean duration of admission	2.4 days	10.6 days	0.000*

* significant.

was used on our cases because of the available facilities and experience. The 22.4% of our cases managed initially by enema reduction is smaller than the figures from some other developing countries,[17,18] and significantly fell short of the 70% or more reported from developed countries.[4,7,15,19–21] The cause of this disparity may be multifaceted and some of our findings highlighted factors that might be contributory. Factors related to some surgeon's preference for operative treatment, inadequate facilities and trained personnel, and delayed presentation are found predominantly in some developing countries like ours and may negatively impact on enema reduction treatment of pediatric intussusception. The reasons why some surgeons have not accepted this current modality of treating intussusception in our setting remain uncertain. We are hopeful however, that as more of the younger surgeon and residents are trained in this procedure and with involvement of more radiologists, more of the affected children that meet the criteria for the enema reduction will be availed the opportunity. Closely related to this are the inadequate facilities and trained personnel in interventional radiology. As earlier stated, this is a major limitation in our setting. Considering that intussusception is an emergency and require prompt diagnosis and appropriate treatment, delay at the radiology unit occasioned by inadequate number of ultrasound machines and trained staff serving a large number of other hospital patients might be a cause for concern. In most cases, this may compel the surgeon on duty to opt for operative treatment. Addressing this will involve improvement of healthcare funding with provision of more ultrasound machines, training of more specialized personnel, and establishment of interventional radiology suite that will provide daily 24-hour service. This may be augmented by training and encouragement of surgeon-operated ultrasound, with the surgeon performing the sonography and the reduction in cooperation with the radiologist. Clearly, well-equipped and cooperating radiology and surgery departments would be the best solution. Delayed presentation is relatively common in many developing countries.[11–13] As previously stated, our patients had been preselected towards difficult-to-treat patients by referral from other adjoining primary and secondary health facilities, and this may have been a reason why several of our cases had required primary surgery based on clinical grounds. Previous reports indicate that referral at advanced stage of disease is not limited to intussusception but involves virtually all other childhood surgical conditions.[22,23] Delayed presentation may leads to development of complications that may preclude success with enema reduction.[2,7,9,14] It is for this reason that some authors suggested that presentation after 2 days of symptoms is a major exclusion criterion for enema reduction.[7,9,24] However there is a more recent consensus that duration of symptoms should not be used in isolation to exclude patients, but rather it should be viewed with respect to the development or presence of the complications like bowel perforation, peritonitis, presence of entrapped fluid, apex at the distal left colon, and the general state of the child to make the decision on exclusion.[10,25] Based on this current thinking, every child with intussusception may be subjected to enema reduction irrespective of the time of presentation as long as there is no demonstrable clinical or radiologic complication. While this may seem laudable in more developed settings, and the fact that some authors[4,10,25] have reported success in cases presenting after 3 days notwithstanding, caution might be applied in our setting and in some other developing economies where there is extreme delays in presentation and significant number of cases may have comorbidities like malnutrition and malaria.[11,12,26] In such setting, it is imperative to judge each patient's clinical situation based on patients condition, providers' experience, and available facilities and personnel.

The success rate of nearly 85% with enema reduction in this study may be similar to the reported success rates of 48% to 97% from centers that routinely apply this procedure.[2–4,6–9,19–21] However, our small number of cases may not compare statistically with the large series from these more developed centers. Previous reports[3,6,7,9,15,21] have indicated that a major factor in the success with enema reduction is proper patient selection with exclusion of patients that have the apex at the left colon or ultrasound evidence of free fluid, pathologic lead point, small-bowel obstruction, or trapped fluid. None of our patients, including the ones with failed enema reduction, exhibited these exclusion criteria. This may indicate that these features were not rigorously sought after in our cases. On the other hand, it may reflect that other factors might contribute to the success or otherwise of enema reduction. Delayed repeat attempts at this procedure and experienced provider have been shown to improve success.[10,20]

As documented in the present report and some earlier reports,[4,6,19,21] enema reduction of intussusception is rarely associated with procedure-related complications. The commonest complication is recurrence of intussusception which may occur in 5 to 14% of cases and mostly within 72 hours of the initial reduction.[25,27] Although recurrence was not observed in our series after follow-up of 3 months, it may probably be as a result of the small number of our cases.

Compared with operative treatment, enema reduction of pediatric intussusception is less invasive, and cost effective.[1,4,6,14,27,28] The present study has demonstrated significant advantages of the enema reduction in the areas of reduced morbidity, early oral intake, and short duration of admission. Though these advantages have been previously reported in studies from developed and some other developing countries,[4,6,14,18,27,28] a more critical evaluation show that most of the cases that had operative treatment have poorer clinical state prior to intervention. In such patients, prolonged duration of symptoms or in some cases severity of the predisposing condition might induce altered physiology that may warrant aggressive electrolyte correction and blood transfusion and other supportive perioperative therapy.

Limitation of the Study

This study was limited by the small number of cases and its retrospective nature. A larger number of cases would have availed better analysis and basis for critical comparison with published larger series. As earlier noted in a previous publication,[29] the small number of cases may not reflect the true prevalence of this disease in our setting. Most of the affected children may not have been brought to the attention of the trained medical practitioners as a result of mostly ignorance and poverty. A well-designed population-based prospective evaluation might have given better insight into some of these peculiarities of pediatric intussusception and challenges of the management in our setting.

Conclusion

Ultrasound-guided saline hydrostatic reduction of intussusception is feasible in our setting, and compared with operative treatment is cost effective and has fewer complications. Delayed presentation, and inadequate number of trained personnel are the main challenges to effective application of this modality of treatment to a large proportion of affected children. Despite the delay in presentation in most of our cases, this procedure may still be applied initially to patients who do not have clinical or radiologic contraindications. Measures geared towards improving time to diagnosis, good collaboration of surgeons and radiologists, and establishment of functional interventional radiology suite may enhance the use of this modality and ultimately minimize morbidity and mortality from pediatric intussusception.

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