CASE REPORT

Appendiceal Knotting: A Rare Complication Causing Intestinal Obstruction in a Child

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ABSTRACT

Appendicitis is a common cause of acute abdomen in children. Its common complications are; perforation, gangrene and appendix mass. The most dreaded complication is portal pyemia which could be fatal. Appendiceal knotting is a rare complication. No patient with this complication has been reported in literature. We therefore present the management of this rare complication seen as a cause of intestinal obstruction in a child at our centre.

KEY WORDS: **Appendicitis Complication, Knotting.**

INTRODUCTION

Appendicitis is the most common cause of surgical abdomen in children. Currently it accounts for approximately one third of childhood hospitalisation for abdominal pain[1]. It is also considered as a cause of acute non specific abdominal pain in children.
The life time risk of appendicitis is estimated to be 8.67% for boys and 6.7% for girls[2]. Appendicitis is most common in older children and adolescents with a peak incidence between 12 and 18 years. This condition is less common in those who take high fibre diet.

A genetic predisposition appears operational in some cases particularly in children in whom appendicitis develops before the age of 6 years[3]. Luminal obstruction causing appendicitis may result from lymphoid hyperplasia, inspissated faecal matter and sometimes parasites. The diagnosis of appendicitis in a child deviates from the classic description of the disease. This is because the presentation and manifestation in a child is completely different from an adult. It is also pertinent to note that complications ensue quite early in children when compared to adults.

Appendiceal knotting is a rare complication of this disease. No patient has been managed in our environment or reported in literature with this
condition. There is also no report of this condition as a cause of intestinal obstruction. We therefore report the management challenges of a patient who presented with appendiceal knotting causing intestinal obstruction at Usmanu Danfodiyo University Teaching Hospital, Sokoto, Nigeria.

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CASE HISTORY

HM was a 10 year old boy who presented to our hospital with a 6-day history of inability to pass stool, associated with central abdominal pain, progressive abdominal distension and bilious vomiting which became prominent 5 days after the onset of illness. He had absolute constipation. There was no history of febrile illness prior to presentation. No past history of similar abdominal pain. No history of trauma to the abdomen in the past or previous abdominal surgery.

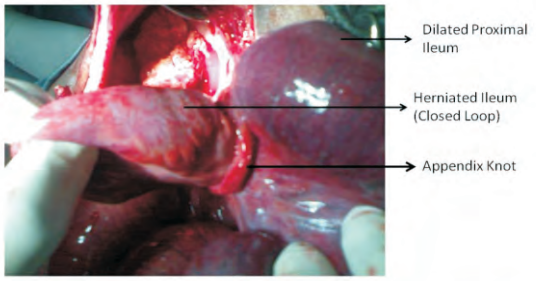
At presentation, the child was acutely ill looking, in obvious painful distress. He was however not pale, anicteric and acyanosed. He was dehydrated with a temperature of 37.0 C. He weighed 18 kg (90% of his expected body weight). Pulse rate was 100 beats per minute, regular, good volume. Blood pressure was130/90 mm Hg. He had normal heart sounds. He was dyspnoeic with a respiratory rate of 40 per minute. He had vesicular breath sounds, no added sounds.

The abdomen was grossly distended. However it moved with respiration, with visible peristalsis from left to right. The abdomen was tense but non tender. No palpable organomegaly. No masses were felt. The bowel sounds were exaggerated. Inguinal hernial orifices were intact. Rectal examination revealed an empty rectum, no mass, no tenderness, gloved finger stained with normal rectal secretions, non mucoid and non bloody.

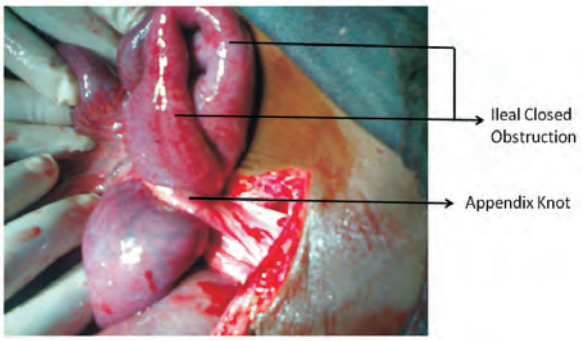
A provisional diagnosis of intestinal obstruction possibly from malrotation with midgu volvulus was made with differentials of colonic obstruction and intestinal duplication entertained.

He was resuscitated. The packed cell volume was 37% and urea and electrolytes were within normal limits after resuscitation. There was delay in obtaining plain abdominal X-ray and abdominal ultrasound before intervention. He had intervention without these investigations to avoid further deterioration.

At laparatomy, via a long lower midline incision, the intra-operative findings were; an inflamed 10 centimetre appendix with the tip knotted at the inferior surface of the terminal ileum and an inflammatory mass surrounding the tip. There was herniation of part of the terminal ileum through the knotted appendix causing a closed loop obstruction as well as simple obstruction proximal to the closed loop portion. The proximal portion of the ileum, the jejunum, duodenum and stomach were dilated. The distal ileum and the entire colon were collapsed. There were no adhesions and no intestinal duplication seen. There were no features of malrotation. There was an ischaemic portion on the terminal ileum at the level of the neck of the closed loop obstruction which recovered after relief of the obstruction, figure 1 and 2

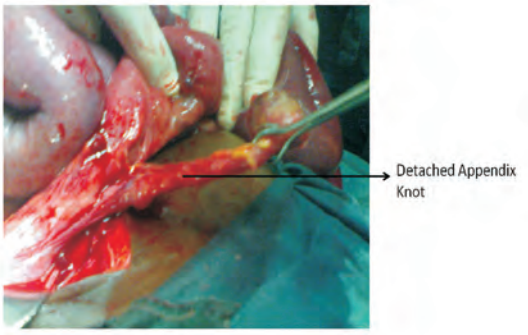


Picture 1 Appendiceal Knotting with herniated ileum in closed loop obstruction



Picture 2. Appendiceal knotting causing closed loop obstruction

The obstruction was released by detaching the tip of the knotted appendix from the terminal ileum. Appendectomy was done but the stump was not buried, figure 3.



Picture 3. Released appendix knot – relieving obstruction.

The post operative recovery was satisfactory and the patient was discharged home in good condition on the 7th day post operatevely after removal of stitches. Histology revealed schistosomal appendicitis and schistosomal appendiceal lymphadenitis.

DISCUSSION

Diagnosis of appendicitis could be quite challenging in children. The classic description of this disease in adults often deviates in children, thereby making diagnosis difficult. More so in children the various differential diagnosis may be difficult to differentiate by clinical and laboratory means. For example, neonatal appendicitis could beindistinguishable from focal necrotising

enterocolitis confined to the appendix[4].

Intestinal obstruction in children may also be difficult to diagnose. Intestinal obstruction from internal herniation is particularly difficult to diagnose pre-operatively. Most often the diagnosis is made intra-operatively as was seen in our patient. The common areas for internal herniation are the hiatus orifice, foramen of Winslow, para-duodenal recesses, the pre-ileal recess and the obturator foramen. There may also be internal herniation if there are defects on the mesentery.

Appendiceal knotting causing intestinal obstruction is a rare and strange cause of intestinal obstruction as seen in our patient. It therefore suffices to say that appendicitis when it occurs could be complicated to any dimension. The natural history of appendicitis is quite varied. There are instances where appendicitis has resolved spontaneously[5]. Perforation is a very common complication in children especially those younger than 6 years and even more in those less than 1 year[6]. The other complications are gangrene and appendix mass which could either be an appendix phlegmon or an appendix abscess[7]. Portal pyemia is a rare and dreaded complication which may result in high mortality. Due to these complications, it is advocated that appendicitis should be treated urgently in children[8]. Appendicitis when it occurs may sometimes cause intestinal obstruction in children. This obstruction occurs This obstruction occurs due to ileus caused by local inflammation around the terminal ileum and caecum. Obstruction may also occur when there is generalised peritonitis causing ileus. This obstruction is adynamic and different from the mechanical obstruction seen in our patient.

Diagnosis of appendicitis was not entertained in our patient because the child presented with features of mechanical obstruction. There were no features suggestive of appendicitis before the features of intestinal obstruction. This was however not surprising since the presentation of appendicitis in a child could be quite deviant. There was a delay in obtaining appropriate radiological and ultrasonographic investigations which might have assisted in making a diagnosis or eliminating differential diagnosis. This was a major management challenge. Computerised tomographic scan which could have been useful here was not readily available. Sometimes when available, affordability becomes a problem. Delay was a major issue. Due to these limitations, laparatomy was therefore employed in the patient as a delay might have resulted in deterioration of the clinical state of the patient or might have resulted in morbidity or mortality of the patient.

Appendiceal knotting is a very rare complication of appendicitis. This condition has not been reported in literature. The other conditions that might have simulated this condition could have been malrotation with mid gut volvulus, ileo-sigmoid knotting, strangulating intestinal adhesions or bands, intestinal duplication or intussuception.

The treatment for appendicitis is appendectomy. Our patient with appendiceal knotting had relief of intestinal obstruction by detaching the tip of the inflamed appendix from the terminal ileum with appendectomy performed without burying the stump[9,10]. This was necessary in this patient as there was associated surrounding inflammation involving the terminal ileum and caecum.

Appendicitis which is a common condition in children. It may continue to be complicated variably and dimensions may continue to be added as seen in our patient with appendiceal knotting causing intestinal obstruction from internal herniation into the recess created by the knotted appendix. It is therefore important to continue to report these new dimensions when they occur in order to inform paediatric surgeons with a view to improving morbidity and mortality.

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CASE REPORT

Open Inter Locked Nailing Without Targeting Device or X-Ray Guide in Revision Surgery for Non-Union of the Femur: A Case Report

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ABSTRACT

We present a patient with non-union of the femur and fracture of the nail after Kuntscher nailing who went on to have union after interlocked nailing with a modified Kuntscher nail (K-nail). The interlocked intramedullary nailing was done without X-ray guide or targeting device. The essence of this report is to highlight the possibilities inherent in modification of devices and established procedures. The early result calls for a closer look at this cheap, safe and effective means of handling femoral non-union in third world societies where instrumentation and implants for interlocked nailing may not be readily available.

Key Words: Interlocked-Intramedullary nail, Femur, Cortical window.

INTRODUCTION

Interlocked intramedullary nailing is a standard technique of internal fixation of femoral fractures. It has the capacity to prevent mal-rotation, shortening and angulation of comminuted femoral fractures.1 Cost of the C-arm X-ray machine, instrumentation and implants as well as the absence of a safe and readily affordable alternative technique of using interlocked nails have made this excellent fixation method inaccessible to most patients in many third world countries.

The practice of intramedullary nailing in a number of developing countries has remained to a large extent limited to the reamed intramedullary nailing technique popularised by Kuntscher.2

In addition, the occurrence of non-union after the use of K-nails is most likely in oblique fractures and may be associated with fatigue of the nails.

Further more, after the effect of nailing the on intramedullary blood flow; subsequent plating of the fracture may carry a higher risk of necrosis of the underlying bone.

Based on the above we modified a regular k-nail by creating two distal holes and one proximal hole to enable the insertion of interlocking screw.

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CASE HISTRORY

A female, 28 years old, presented with a history of pain at the upper part of her left thigh. She had fracture of the left femur following a road traffic accident three years before presentation. She had been operated upon twice by the same surgeon who .had to replace a broken Kuntscher nail (K-nail). Radiograph (Figure 1) confirmed that the second K-nail had also suffered fatigue fracture.

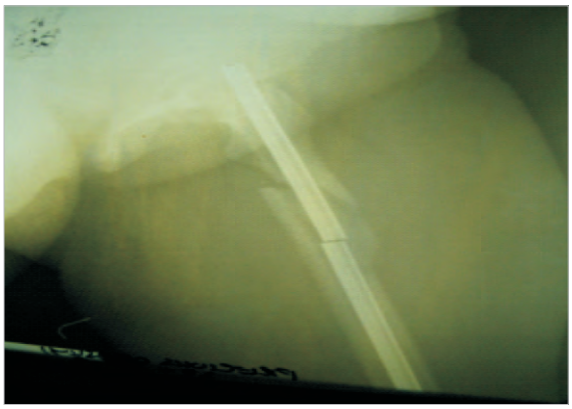

Figure 1: Pre operative X-ray (Anteroposterior view

We converted standard K-nails of sizes 12 and 13 to interlocking nails by drilling one hole at the proximal end (The hole was placed obliquely at 130° to the long axis of the nail and started at 2cm below the end of the slot for extraction) and two holes at the distal end. The

distal holes were 2cm apart and at ninety degrees to the long axis of the nail. The lower
of the distal holes was placed at 2cm from the end of the nail. All holes were placed at ninety degrees to the longitudinal slit of the nail, (Figure 2)

At surgery, which was done in the supine position under epidural anaesthesia, the limb was adducted to the extent that the ipsilateral popliteal fossa lay over the contralateral knee. The greater Trochanter was exposed through a 5cm long longitudinal skin incision. The piriform fossa was located with the gloved finger and an awl (Herzog awl) was used to perforate it to gain access to the proximal end of the intramedullary canal.

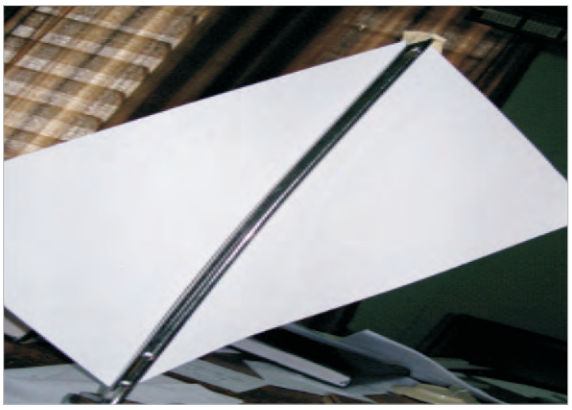

Figure 2: The modified K-nail with one proximal and two distal holes.

. An incision, approximately 10 cm (Five-finger breadth) long, was made at the lateral aspect of the thigh over the site of fracture. The incision was developed in layers to the site. The distal piece of the broken nail was removed through lateral thigh incision. The proximal piece was removed through the proximal incision. A smooth tipped 2mm guide wire was introduced into the proximal fragment through the piriform fossa. The intramedullary canal was reamed, over the guide wire with hollow reamers to size 14. The distal fragment was canalized using reamers of similar size. The appropriate length of nail was determined by inserting the guide wire into the medullary canal and measuring the depth with a sterile ruler. The guide wire was re-introduced from the piriform fossa and advanced into the distal fragment under direct vision. A size 13mm/40mm Kuntscher nail, which had been modified to enable the application of two distal 4.5mm AO cortical screws and one proximal 4.5mm malleolar screw, was threaded over the guide-wire into the distal fragment. The guide wire was removed and the nail was advanced until only a finger-breadth of the nail was palpable above the piriform fossa. The longitudinal slit in the nail was directed posteriorly. Another nail of equal length was placed parallel to the one in the intramedullary cavity and used as a rough guide to the likely site for interlockingscrews placement A 5 cm incision was made over the distal locking screws site. The intramedullary cavity of the distal end femur was exposed through a longitudinal anterolateral cortical window, measuring about 1cm by 0.5cm, which was made on the lateral condyle. With the aid of a 1 mm kirschner wire the site of each hole on the nail was identified. A 2.7mm drill bit was used to drill through the near cortex and the far cortex with the drill bit passing through the hole in the nail. A 4.5mm tap was used to tap the holes and a 4.5mm cortical Screw was placed across as an interlocking screw. The same procedure was repeated for the next hole. With the aid of the similar nail, the lateral part of the oblique proximal hole was targeted and a 2mm Kirschner wire (K-wire) was drilled through the bone and the lateral part of the hole at 130 degrees to the axis of the nail. The presence of the Kirschner wire in the lumen of the nail was confirmed by a probe placed into the lumen of the nail. The hole made by the K-wire was drilled with a 3.2mm drill bit and a self tapping malleolar screw was used as the proximal locking screw. Ipsilateral proximal tibial cancellous bonegrafts were harvested and applied at the fracture site. All wounds were irrigated with normal saline and closed in layers using nylon 2.0 for skin with suction drain tube inserted beside the fracture site.

Post-operatively, intravenous ceftriazone 1gm daily for 5days and 500mg of metronidazole 8hrly for 24 hours; Intramuscular injection of 100mg of tramadol hydrochloride 8hrly for 72 hours as well as daily intramuscular injection of 75mg of diclofenac sodium for 5 days were administered. In addition, subcutaneous enoxaparin 40mg daily for seven days from the first day after surgery and oral soluble aspirin 75mg daily for four weeks from the second day after operation were given. The operated limb was elevated on a pillow for 5 days with the knee in 15° flexion. The drain was removed on the second day post-operation. The patient commenced static quadriceps exercises on the 3rd day after the operation and was ambulated on a pair of axillary

crutches (Non-weight bearing) from the 5th day after surgery. All stitches were removed on the fourteenth day after operation.

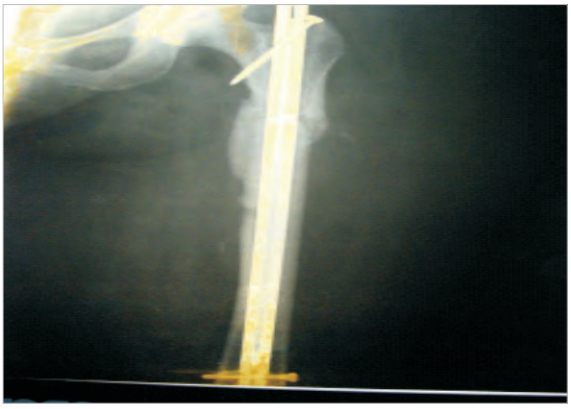

Figure 3: Post-operative X-ray, anteroposterior view

The patient was discharged home on the fourteenth day after the operation. Partial weight bearing with bilateral axillary crutches was commenced at six weeks post- operation and continued till the twenty-fourth week after surgery. Ambulation with the aid of a walking stick commenced at twenty-four weeks post-operation. At 52 weeks after surgery an anteroposterior radiograph (Figure 3) shows satisfactory union without breakage of interlocking screws. Eighteen months after surgery, the patient is ambulating without support and has no discomfort.

DISCUSSION

This case represents the best reminder that regardless of the value of the intramedullary nail, if it is not locked, the risk of non-union is real and that fatigue fracture of the nail usually ensues. It also shows the earliest results of efforts to offer stable Kuntscher intramedullary nail fixation using interlocking screws without the aid of the C-arm, targeting device and fracture table in our centre.

The presence of atrophic non-union is associated with sclerosis and narrowing of the ends of the fragments. The sclerotic ends of the fracture may need to be freshened under direct vision for the application of cancellous bone grafts at the site of atrophic non-union. Closed reduction and interlocked intramedullary nailing under image intensifier-guide is superior to the open technique we have described.

However, in a case with atrophic non-union, fractured K-nail and adjacent tissue fibrosis from previous surgery, open nailing is considerably safer in the absence of image intensifier. The technique we have described is devoid of the risk of radiation injury to the surgeon and patient.3 The quality of fixation that is derived from it is expected to be superior to the unlocked open Kuntscher nailing that is still frequently done in a number of developing countries2 because of high cost of interlocking nails and required facilities. This treatment option enhances shortened hospital stay and early ambulation without compromising the quality of fixation and will be of great value in operative treatment of fractures in such low income societies where patients' preference for traditional bone setters' treatment is rife and as a result nonunion is quite common. The anterior cortical window access for distal locking is not new and had been described by Kanellopoulos et al4 who used it as a salvage technique for distal locking When their image intensifier developed a fault. This case report ostensibly adds a window of opportunity for patients in underprivileged societies to benefit from the superiority of interlocked intramedullary nails over the unlocked Kuntscher nailing which is still in common use in such settings.

The surgical implants generation network (SIGN) interlocked nailing without image intensifier is a notable and valuable alternative that will make the distal cortical window rarely necessary but it is at this moment not widely available and accessible in Nigeria [5,6] and many other developing nations.

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CASE REPORT

Fournier's Gangrene in Children: A Report of 2 Cases

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ABSTRACT

Although Fournier's gangrene is primarily a disease of adults, It has been rarely described in children. This is a report of our experience with the management of 2 patients aged 14 and 36 days. The predisposing factors were omphalitis and in one it followed circumcision. Both patients had debridement and the wounds healed by secondary intention. They were discharged and are alive and well. The outcome of treatment of Fournier's gangrene in children is good.

Key words: Fournier's gangrene, Children, Management.

INTRODUCTION

Fournier's gangrene (FG) was initially described as a disease of young adults of unknown cause by Alfred Fournier in 1883.[1] The disease is now recognized as necrotizing fasciitis of infective origin with a definite source of infecting organisms which may be so trivial as to be undetected. There are also few reports of FG in children.[2-5] This is a report of our experience with the management of this uncommon problem in childhood.

CASE HISTORY

Case 1

A 14 day old male newborn with a wound observed on the scrotum 5 days prior to presentation. The pregnancy and labour were unsupervised and the mother delivered at home. The child was noticed to have umbilical discharge associated with progressive abdominal distension. Examination revealed a sick neonate, febrile (temperature – 39.4°C) and very pale. He was uncircumcised and had an ulcer on the anterior aspect of the scrotum (Fig. 1). The blood count revealed anaemia (pcv-15%) and wound culture yielded Escherichia coli and Staphylococcus *aureus*. He was resuscitated with intravenous fluids and blood transfusion. He was placed on parenteral broad spectrum antibiotics gentamicin,, ampclox and metronidazole. He was also given antitetanus serum. All necrotic tissue was debrided and his wound healed by secondary intention and he was discharged from hospital 20 days after admission.

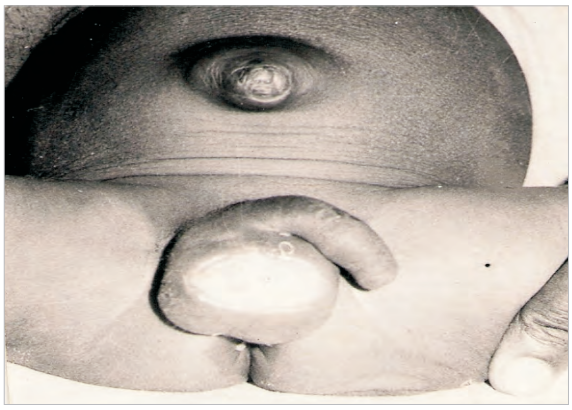

Figure 1: Fournier's gangrene with necrotic tissue sloughed off before presentation

Case 2

A 36 day old infant presented with fever and pain over the perineum. This followed circumcision 7 days earlier. Examination revealed necrotic tissue over the

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distal parts of the penis, anterior aspect of the scrotum and over the left thigh. There were also areas of hyperaemia and swelling (Fig. 2). He was resuscitated and broad spectrum antibiotics administered (gentamicin, ampclox and metronidazole). He had debridement of all devitalised tissue. He did well and his wound healed by secondary intention with wound dressing. He was discharged after 23 days on admission.

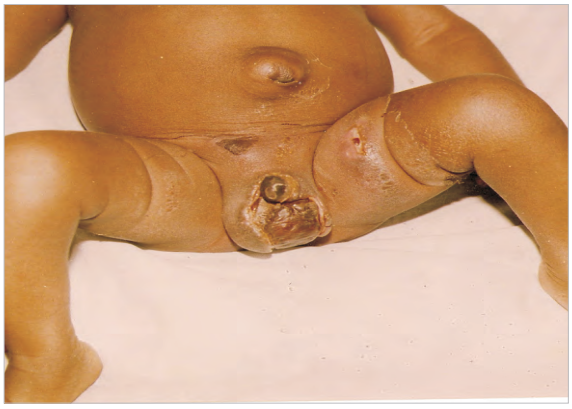

Figure 2: Fournier's gangrene with necrotic tissue

DISCUSSION

FG is primarily a disease of adults. However, it is uncommonly described in children.[2-5] Our description of the problem in a neonate and an infant will be an addition to the literature on FG in children. The original description characterized the condition as of sudden onset in a healthy young male with a rapid progression to gangrene and the absence of a definite cause.[1] However, it is now recognized as an infective necrotising fasciitis.

The source of infection may be urogenital (45%), anorectal(33%), or cutaneous (21%).[6] The predisposing factors for FG include abscesses, omphalitis, diaper rash, surgery like circumcision and herniorrhaphy, burns, insect bites , anorectal trauma, nephritic syndrome.[2-5,7] Others include systemic disorders like immunocompromised states, haematologic malignancies.[6] Omphalitis and circumcision were the predisposing factors in our patients. However, we did not screen any of the patients for human immunodeficiency virus infection (HIV). In adults systemic disorders like diabetes, alcoholism, malnutrition and HIV play important role in the development of FG.[8]

One of our patients presented with severe neonatal sepsis and anaemia. An aggressive resuscitation with intravenous fluids, parenteral broad spectrum antibiotics and including blood transfusion should be instituted. Surgical debridement in the above case was carried out by the bedside.

Infection is frequently polymicrobial gram negative organisms, gram positive organisms and even anaerobes. In one of our patients the culture grew *Escherichia coli* and *Staphylococcus* aureusWe treated both patients with debridement and allowed the wound to heal by secondary intention. This was associated with a longer hospital stay. Some authors advocate for closure once the wounds are clean as this will reduce the length of hospital stay.[5] When the source of infection is from the anorectal region or when urinary extravasation or peri-urethra inflammation is present, urinary or faecal diversion is indicated to reduce contamination and allow wound healing to take place.[9] We did not have to divert the urine or faeces in any of our patients.

The prognosis of FG is more favorable in children[2-4] than adults. Eke,[10] has also found the prognosis to be better in his Nigerian series of adults. This is probably because of the predominance of skin source of infection which is associated with a better outcome than when the source is from the anorectal region [11] and also the low incidence of premorbid systemic disorders in these group of patients.

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