Case Report

The Pattern and Outcome of Management of Acquired Anorectal fistulas in Children at a semi-urban Teaching Hospital

Samson Olori, Olabisi O Osagie

Division of Paediatric Surgery, Department of Surgery, University of Abuja Teaching Hospital, Gwagwalada, Abuja.

ABSTRACT

***Background**:* Acquired anorectal fistulas are a severe cause for parental concerns, and they pose considerable challenges in their management.

***Aim**:* The aim of this study is to document the pattern of presentation and the management outcome for the cases of acquired anorectal fistula in children seen in our centre.

***Method**:* A retrospective review of the thirteen (13) case notes of the patients with acquired anorectal fistulas managed from 2008-2011 was done. The information retrieved included; the age at presentation, sex, aetiology of the fistula, site of the fistula, the duration of the fistula to the presentation, weight loss, and other associated medical conditions, history of a developmental milestone, type of intervention, the outcome of management, HIV status of patients and parents.

***Results**:* There were two males and eleven females; eight of whom were HIV positive. Five fistulas were due to each to faecal impaction, birth trauma, trauma from rectal enema procedure, iatrogenic following posterior sagittal ano-rectoplasty (PSARP) and sexual abuse respectively.

***Conclusion**:* The fistula healing outcome for acquired anorectal fistula in HIV positive cases was weak compared to the trauma-related groups.

INTRODUCTION

Anorectal fistulas are either congenital or acquired.[1, 2] Acquired fistulas are a cause for concern and pose a significant challenge in their management because of their aetiology. Each case of acquired fistula would, therefore, be treated differently. These fistulas could be rectovestibular, rectovaginal, recto-urinary and fistula-in- ano. [3] The causes include; perirectal infections, trauma, malignancies and radiation injuries, the latter two being more common in the adults. Recently, HIV infection has been implicated in the causation of fistulas in the perineal region in children. [4, 5] Hence, perineal fistula is being proposed as one of the signs of HIV infection, the pathogenesis being microcytic abscesses in the perineal region which rupture into the anorectal canal[5]. Fistulas are a cause of concern for parents and their children and often task the surgeon's skills in their management.

This review corresponded with the time when HIV/AIDS patients were stigmatized, and many lived in denial of their status or secretly with it. Voluntary counselling services and antiretroviral drugs were not readily available. The HIV positivity of the patients with acquired fistulas and the outcome of their treatment were of interest to us. A

Correspondence:
Samson Olori
Email: samsonolori@yahoo.com,
Tel. +234(0)8065817508

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retrospective review of the pattern and issue of management of the acquired anorectal fistulas in children managed in our centre was done. In Africa, the literature on acquired anorectal fistulas in children are few; we at this moment report our experience, which we hope will add to the existing literature on the subject.

MATERIALS AND METHODS

The medical records of patients with anorectal fistulas managed in 2008-2011 were reviewed for age at presentation, sex, weight, type of fistula and location, number of fistulas, duration of the fistula to a presentation, HIV status of patients and their parents, associated medical conditions, aetiology, intervention and outcome of management. The findings are presented in tables, graph and figures.

RESULTS

There were two(2) males and eleven(11) females (M: F 1:5.5) with an age range of 1- 13yrs, the modal age at presentation was 0-1yr. Eight(61.5%) cases were due to HIV infection while five(38.5%) were due to trauma(sexual abuse, birth trauma following the breech presentation, faecal impaction in a bedridden sickle cell disease patient being managed for dislocated left hip, injury from rectal enema performed by a quack, and from a post posterior anorectoplasty for rectovestibular fistula). There were two fistula–in-ano and six rectovestibular fistulas in the HIV positive group. Three (60%) of trauma-related cases had rectovaginal fistulas (Figures1).

Figure1: Acquired rectovaginal fistula due to sexual abuse (see vagina draining feces with disrupted patulous the anus)

One of these patients presented in the first week of life following birth trauma to the vestibule, which was successfully repaired. Table 1 shows details of the clinical manifestations, aetiology type of fistula and their location with the anorectum and treatment offered and the outcome.

Table 1: Parameters extracted from the case notes, numbers 1-13 represent the patients

Parameters	1	2	3	4	5	6	7	8	9	10	11	12	13
Age	12m	11yr	14yr	7m	4m	2m	8m	4m	1d	12m	12yr	3yr	1m
Sex	F	M	F	F	F	F	F	F	F	F	F	M	F
Weightloss	+	+	+	+	+	+	+	-	-	-	+	+	-
Anaemia	+	-	+	+	+	+	+	+	-	+	-	-	+
Fistula type	RVF	FIA	RVF*	RVF	RVF	RVF	RVF	RVF	RVF	RVF*	RVF*	FIA	RVF
Fistula site	P	P	P	RL	A	P	P	P	P	P	LL	LL	P
Duration	1wk	-	4wks	6d	4wks	4d	4d	1wk	5	5d	4wks	1yr	1wk
Aetiology	HIV	HIV	FE	HIV	HIV	HIV	HIV	HIV	TR	SA	TR	HIV	TR
HIV status	+	+	-	+	+	+	+	+	-	-	-	+	-
Action	ARV	ARV	MEW	ARV/AB	ARV	ARV	-	NYS	Repair	RES	BNU	-	Col
Outcome	Died	H/A	H/A	Died	Alive	Alive	LF	Died	H/A	Died	H/A	died	H/A
Dm	D	ND	D	ND	ND	ND	D	D	D	ND	ND	ND	ND

KEY: d=day, m=month, FIA = fistula-in-ano, RVF= rectovestibular fistula, RVF*= rectovaginal fistula, FE=fecal impaction, TR=trauma, SA=sexual abuse, ARV= Antiretroviral drugs, NYS= Oral nystatin, RES =Resuscitation, AB=Antibiotics, MEW= manual evacuation and rectal wash out, Dm= Developmental milestone D= Delayed, ND =not delayed, Col=colostomy, BNU=Build up Nutrition, A= Anterior, RL=right lateral, LL=left lateral, p= posterior, + positive/present, - negative /absent, LF =loss to follow up, H/A =Healed and alive

Anaemia and weight loss were present in 9(69%) of the cases. The duration of fistula from when it was noticed to presentation ranged from 6days-1year with a mode of 1week. Of the eight (8) HIV-positive patients, two of them had a CD4 count of less 200, multiple fistulas and were among the four (50%) that have died. Clinical features of HIV/AIDS were present in about 50% of the cases (Table 3 and the graph). A summary of treatments offered and the outcome of care is shown in tables 2. The HIV status of the parents was ascertained in 30.8% (8 out of 26). Four of the parents were negative for HIV while four (two couples) who were positive died of HIV/AIDS in the course of management of their children.

Table2. Outcome of fistula management related to aetiology

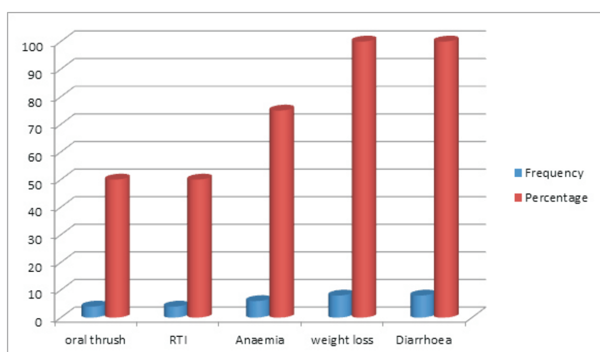
Aetiology	HIV	Trauma	Sexual abuse	Fecal impaction
Frequency (%)	8(61.5)	3(23)	1(8)	1(8)
Actions	ARV	EUA/repair/colostomy	Resuscitation	Bowel washout
Outcome	4 alive/4 died	Alive	Died	Alive

Key: EUA =examination under anaesthesia, ARV =antiretroviral drug

Table3: Symptoms associated with HIV/AIDS positive patients

Parameter	Frequency	Percentage (%)
Oral thrush	4	50
RTI	4	50
Anaemia	6	75
Weight loss	8	100
Diarrhoea	8	100

Symptoms associated with HIV/AIDS patients in graphic form



DISCUSSION

Acquired anorectal fistulas pose a significant challenge in management because of their varied aetiologies. [4] These fistulas could be rectovaginal, recto-vestibular, recto-urinary, and fistula-in-ano. The causes include; peri-rectal infections, trauma, malignancies and radiation injuries.[6] HIV infection has been implicated in perineal fistulas in children[7, 8]. According to Friesen, the manifestation of HIV depends on different opportunistic infections; this may explain why perineal fistula formation may not have been recognized as a prominent feature of this condition because of its apparent hidden location.[9] In this study, 61.5% (8)of the cases were positive for HIV infection. Perineal fistula has been proposed as one of the signs of HIV infection[8, 10].

According to ES Borgstein; 'we believe that the early appearance of acquired rectovaginal fistula in infant girls is an early manifestation of HIV infection'.[3] The number of boys in this study is similar to that of Wiersma who found the small number of HIV positive males with acquired fistulas in their report.[11] One of the fistulas in an 8month old HIV positive girl was reported to have been healed spontaneously, but the child was lost to follow up. However, an 11yr old boy who was infected through blood transfusion in the perinatal period, had his fistula healed with hypertonic saline sitz bath and ARV medication. This particular case underpins the need for safe blood transfusion. There was 38.5% overall mortality, and 50% mortality for the HIV group, which is similar to the high death recorded in previous reports.[5,8,12] All the patients received antibiotics however; the HIV infected cases in addition had antiretroviral drugs. Anaemia, diarrhoea, weight loss and respiratory infections were prominent among HIV positive patients (see Table 3) who also had delayed developmental milestones compared to the trauma-related acquired fistula group. Delayed developmental milestones are known sequelae of HIV infection in children who may not have had appropriate antiretroviral medication.[13,14]

Iatrogenic injuries following repair of congenital rectal fistulas have been well documented.[12, 15] For the patients who had a postoperative fistula and the one due to quackery from the rectal enema, diverting colostomies done and after the healing of the fistulas, the colostomies were closed, and patients passed stool usually. There were no post-repair complications. The protocol of creating a diverting colostomy to allow a distal pathology to heal or be treated has been reported in the previous study.[11,16]

Fistulas following sexual abuse have been reported.[17,18,19] An infant girl, who was abused sexually by an adult, died from sepsis and anaemia which complicated the injury following the very late presentation. Sexual assaults of children are perpetrated by close family members or associates of such families which is sometimes responsible for late presentation to hospital or reporting of such cases to the appropriate law enforcement units. Child rape cases associated with militia and military abounds.[19]

CONCLUSION

The HIV-related fistula group had more inadequate fistula healing and higher mortality in comparison with the trauma-related group.

The outcome of this study further supports the knowledge for HIV investigation in patients presenting with acquired rectal fistula with appropriate surgical management [3,11].

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