Original Article

Post-Circumcision Urethrocutaneous Fistulae: Presentations, Repairs and Outcomes in a Tertiary Centre

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Abstract

Background: Circumcision is regarded as the most common surgical procedure world over. It is also perceived to be a simple and safe procedure; however, it could be fraught with major urological complications such as urethrocutaneous fistula (UCF). Repair of these fistulae poses a great reconstructive challenge to the paediatric surgeon/urologist with varied outcomes. **Aim:** We seek to review the presentations, repair and outcome of post-circumcision UCFs managed in a tertiary centre. **Patients and Methods:** This is a retrospective review of all the consecutive cases of repaired post-circumcision UCFs in Nnamdi Azikiwe University Teaching Hospital, Nnewi, South-East Nigeria, over a period of 9 years (January 2012–December 2020). Data on demography, size of fistula, type of repair and outcome were retrieved from the Records Department and patients' case notes and were analysed using SPSS (version 22, Chicago, Illinois). **Results:** A total of 22 boys had post-circumcision UCF repair within the period, and they were aged between 2 weeks and 108 months with a median age of 4 months at presentation and aged between 8 months and 144 months with a median age of 24 months at the time of surgery. Circumcisions were by freehand technique in 21 (95.5%). Nurses performed most of the circumcisions in 19 (86.4%), and most of the circumcisions were performed in private hospitals 7 (31.8%), maternity homes 3 (13.6%) and general hospitals in 10 (45.5%) each, respectively. Most 17 (77.3%) fistulae were coronal, and the size of defect ranged from 1 mm to 10 mm. The most commonly employed technique of repair was simple closure in 16 (72.2%). Meatal stenosis and re-fistulation occurred in 2 (9.1%) and 4 (18.2%), respectively. Only two (12.5%) required reoperation. There was no relationship between the size of defect and re-fistulation, $P = 0.377$. **Conclusion:** Majority of the Post-circumcision urethrocutaneous fistulae were from free hand technique of circumcisions . These were performed mostly by nurses in general, private hospitals and maternity homes. Hence, there is a need to ramp up training of providers of neonatal circumcision in our environment. Most UCF in children could be repaired with simple closure technique reinforced with dartos flap.

Keywords: Circumcision, urethrocutaneous fistula, urethroplasty

Introduction

Circumcision is perhaps the oldest and most common surgical procedure performed world over with about 37%–39% of males above 15 years circumcised globally.[1] The circumcision rate in Nigeria has been reported to be 87% by Okeke *et al*.[2] in Ibadan, western Nigeria. The rate could vary according to regions and their cultural and religious practices hence may be higher in South-East Nigeria where virtually every male is circumcised at the neonatal period for religious and cultural reasons.[3,4] There are various personnel involved in circumcisions worldwide including doctors, nurses, traditional circumcisionists and Mohels in the Jewish religion. By extension, techniques, locations and facilities in which these procedures are carried out vary also.[5]

Circumcision has been perceived as a short and simple procedure; however, it could be fraught with complications ranging from simple and self-limiting events such as oedema to more complex and daunting complications such as urethrocutaneous fistula (UCF), severe haemorrhage and penile amputations.[4,6]

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Post-circumcision UCF) though rare is commonly found following procedures by untrained personnel and the converse is true when performed by trained personnel and in developed countries.[4,7] UCF is a largely avoidable but severe nonlethal complication which has a huge psychological impact on the child and family alongside untold economic hardships.[7] Aside these aforementioned burdens, repairs are quite challenging to the surgeon with the chances of recurrence and multiple procedures.[4,6,8,9]

Repair techniques for UCF are many and are largely determined by the size of the fistula, location, availability of tissues and surgeons' expertise. Commonly deployed techniques include simple closure, modified Mathieu's repair and modified Snodgrass technique.[6,9]

Our centre is a referral tertiary centre for circumcisions and repair of circumcision-related complications in South-East Nigeria. In this report, we aim to review all consecutive cases of post-circumcision UCF to highlight the presentations, repair techniques and outcomes. This we hope will guide modifications in future efforts at repair and to add to the body of literature in this area.

Patients and Methods

This is a retrospective review of all consecutive cases of UCFs arising from circumcisions from January 2012 to December 2020 in a tertiary health centre, South-East Nigeria. Fistulae secondary to hypospadias repair were excluded from the study.

All the patients were referred to our centre from alternative healthcare facilities. They were first seen at the paediatric surgery outpatient clinics. After evaluation at the clinics, the patients were scheduled for repairs as elective cases. Our choice of repair technique was determined by the location of the defect and size. Small fistulae >5 mm were repaired by simple closure performed in two layers and reinforced with dartos fascia before skin resurfacing. All repairs were performed with appropriate-sized 100% silicone catheters in 9 (40.9%) or feeding tubes 13 (59.1%) and the tubes lasted for 6–12 days with a mean duration of 9.86 ± 1.61 days before decatheterisation.

Data were extracted from the medical records, theatre and ward registers on; the type of circumcision, category of circumcisionist, type of institution where circumcision was carried out, associated complications, type of repair, and outcome. The data were fed into a spreadsheet and analysed using Statistical Package for Social Sciences (IBM SPSS version 22 Chicago Illinois). Results are presented in counts, frequencies and means using tables and charts. The relationship between categorical variables was tested using Chi-square tests, and $P \leq 0.05$ was considered statistically significant.

Results

There were 22 boys who were managed for post-circumcision UCFs within the study period. They were aged between 2 weeks and 108 months with a median age of 4 months at presentation and 24 months at the time of surgery. All the boys had their circumcisions during the neonatal age without anaesthesia.

The category of circumcisionists is represented in Table 1. Nurses performed most of the circumcisions and majority of the procedures were carried out in and General hospitals, private hospitals and maternity homes as seen in Table 1. Freehand technique of circumcision was used in 21 (95.5%) of the cases while Plastibell technique was employed in 1 patient (4.5%). There was associated primary and reactionary haemorrhage in 10 (45.5%) boys while 12 (55.5%) had no other associated complications during and immediately after circumcision. The fistula locations are represented in Table 2.

The fistulae size ranged from 1 mm to 10 mm with a mean of 4.0 mm ± 2.4. Repairs were carried out using the following techniques, as represented in Table 3. The most commonly used technique was simple closure followed by modified tabularised incised plate urethroplasty and Thiersch-Duplay techniques.

There were complications in 6 (27.3%) of the patients as follows: meatal stenosis in 2 (9.1%) and re-fistulation in 4 (18.2%). They were followed up for 8–36 months with a mean follow-up duration of 9.36 ± 10.40 months.

Table 1: Category of circumcisionists and facility of circumcision

	Frequency (%)
Circumcisionist	
Nurses	19 (86.4)
Doctors	2 (9.1)
Traditional	1 (4.5)
Total	22 (100)
Facility of circumcision	
Home	1 (4.5)
Tertiary centre	1 (4.5)
Private hospital	7 (31.8)
Maternity homes	3 (13.6)
General hospital	10 (45.5)
Total	22 (100)

Table 2: Location of fistulae

Location	Frequency (%)
Glans penis	2 (9.1)
Coronal	17 (77.3)
Distal penile	3 (13.6)
Total	22 (100)

Table 3: Techniques of repair

Techniques	Frequency (%)
Thiersch-Duplay	1 (4.5)
TIPU	5 (22.7)
Simple closure	16 (72.7)
Total	22 (100.0)

TIPU: Tubularised incised plate urethroplasty

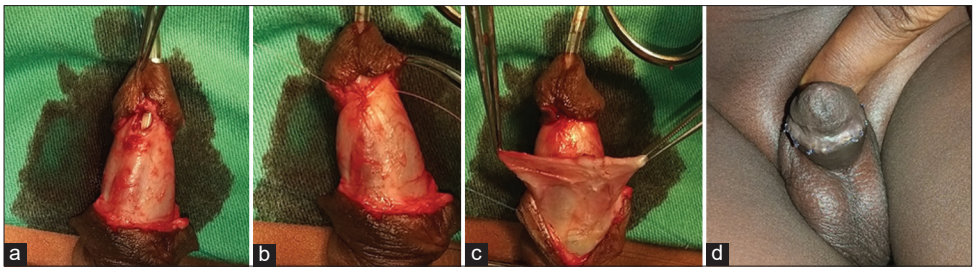


Figure 1: (a-d): Simple closure of a coronal fistula. (a) Degloved penis for a fistula at the subcoronal region. (b) Simple closure of fistula done in two layers. (c) Vascularised dartos fascia for reinforcement. (d) 3 weeks post-operative

Discussion

Urethrocutaneous fistulae following circumcision is a major complication of this seemingly simple procedure. It portends a huge financial and psychological burden on the parents and the patients with risk of multiple reconstructive surgeries.[7,10] Neonatal circumcision rate in South-East Nigeria remains quite high and indications are largely for cultural and religious reasons.[3]

A total of 22 children had repairs of post-circumcision UCF in our facility giving an average of 2.5 cases/year. This rate is less than the figure reported by Ikuerowo *et al*.[6] who had 31 cases in Lagos, Nigeria, over a 3-year period but similar to the findings by Takure *et al*.[9] in Ibadan who had 25 cases in 10 years and higher than the number reported by Ekenze and Ezomike[7] over a 6-year period in Enugu, South-East Nigeria. These discrepancies could be a result of regional differences in circumcision practices and population densities within the country.

All the patients had their circumcisions during the neonatal period. This preference for neonatal circumcision could be informed by culture and religion which are the most common indications except where otherwise indicated by neonatal illnesses and conditions which may preclude neonatal procedures.[3-6,11] Both median age of presentation and repair were found to be late. This is similar to previous reports in Nigeria.[6,9,10] Perhaps, this late presentation could be explained by delayed recognition of the complication as most of the children go on diapers until late infancy when parents would detect the anomaly. Second, there were usually multiple referrals and occasional assurances of 'spontaneous closure' before referrals to the major centres with expertise particularly if the initial procedure was performed by non-doctors.[4]

Majority of the cases were from circumcisions performed by nurses [Table 1], followed by doctors and traditional circumcisionists. Again, freehand technique of circumcision was the most predominant technique. Plastibell clamp was only used in a patient. This trend of nurses performing most of the circumcisions causing UCF and using the freehand technique to do so has been reported in previous studies.[5,6,9,10,12] Nurses in private hospitals, maternity homes and general hospitals performed most of the circumcisions. Osuigwe *et al*.[10] had previously found out that there were fewer complications in circumcisions performed in teaching hospitals by doctors than those performed by midwives in other categories of health facilities. Similarly, Okeke *et al*.[2] reported more complications amongst nurses than doctors. Osifo and Oriaifo[4] also reported more complications in procedures performed by paramedical practitioners with no training followed by nurses before doctors. The assumption that circumcision is a minor procedure for all practitioners in healthcare (both traditional and orthodox) underpins this generalist approach to the procedure. A coordinated training programme both at basic and postgraduate levels for healthcare providers who may wish to perform circumcision in their practice is imperative to mitigate the rate of severe complications such as UCF. The trend in developed countries is different as circumcisions performed by nurses have less complications and this is a result of adequate training on the procedure.[11,13] Training in circumcision amongst doctors in residency training in South-East Nigeria, has been found to be suboptimal; in spite of this, majority of these doctors plan to perform circumcision in future.[12] The scenario in the training of nurses and paramedics may be poorer in our environment even when they provide the highest number of this service.

No form of anaesthesia was used in all the patients and close to 50% of the patients had associated excessive primary and reactionary haemorrhage after the circumcisions. It could easily be adduced that freehand circumcisions predispose more to haemorrhage likely from the frenular artery and attempt to control the bleeding with haemostat and sutures led to urethral injury and fistula formation.,[6,10] The use of clamps such as Plastibell, Gomco and Mogen have been found to be protective and haemostatic, thereby preventing UCF. Plastibell is easy and simple to use with very low risk of UCF in trained hands.[3] The most common location of the UCF is the coronal and subcoronal regions of the penis followed by the distal penile region [Table 2]. This finding is consistent with previous studies.[6,7,10] At these regions, the urethra is more superficial and prone to injury, hence attempt to control haemorrhage at these sites would readily lead to UCF.

Repairs of post-circumcision UCFs remain a huge challenge to the surgeons who undertake them.[6,10] The most commonly employed technique in our series was simple closure [Table 3 and Figure 1]. Our choice of repair was determined

by the location of the defect and size. Small fistulae >5 mm were repaired by simple closure performed in two layers and reinforced with dartos fascia before skin resurfacing. Those that were wider or close to the natural meatus were connected to the meatus and either a tabularised incised plate urethroplasty or Thiersch-Duplay technique performed. Modified Mathieu's and double pedicled skin flap techniques have also been used by some surgeons for wide defects.[5,14] The re-fistulation rate in this study was 18.2% and was similar to 16% reported by Takure *et al*.,[9] though their series included post-hypospadias fistulae. Two of those that had recurrence closed spontaneously and this might have been because of the multilayered repair and dartos fascia reinforcement. Dartos overlapping cover and tunnelled tunica vaginalis reinforcements have been found to improve outcomes in repairs.[9,15]

Conclusion

There is a need to scale up training in neonatal circumcision amongst providers in our environments. This invariably reduces complication rates following this procedure including UCF. Trainings would include the use of anaesthesia and protective clamps for neonatal circumcisions as these would reduce the rate of post-circumcision UCF. In the event of UCF, meticulous surgical techniques with dartos interface have been found to improve the outcome of repaired post-circumcision UCF.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

References

1. Morris BJ, Wamai RG, Henebeng EB, Tobian AA, Klausner JD, Banerjee J, *et al.* Estimation of country-specific and global prevalence of male circumcision. Popul Health Metr 2016;14:4.
2. Okeke LI, Asinobi AA, Ikuerowo OS. Epidemiology of complications of male circumcision in Ibadan, Nigeria. BMC Urol 2006;6:21.
3. Ekwunife OH, Ugwu JO, Okoli CC, Modekwe VI, Osuigwe AN. Parental circumcision preferences and early outcome of plastibell circumcision in a Nigerian tertiary hospital. Afr J Paediatr Surg 2015;12:251-6.
4. Osifo OD, Oriaifo IA. Circumcision mishaps in Nigerian children. Ann Afr Med 2009;8:266-70.
5. World Health Organization. Male Circumcision: Global Trends and Determinants of Prevalence, Safety and Acceptability. Geneva: World Health Organization; 2008.
6. Ikuerowo SO, Bioku MJ, Omisanjo OA, Esho JO. Urethrocutaneous fistula complicating circumcision in children. Niger J Clin Pract 2014;17:145-8.
7. Ekenze SO, Ezomike UO. Complications of neonatal circumcision requiring surgical intervention in a developing country. J Trop Pediatr 2013;59:292-7.
8. Okoro PE, Ekeke NO. Severity of malcircumcisions and circumcision-related complications in three tertiary health facilities in Southern Nigeria. Ann Pediatric Surg 2018;14:130-3.
9. Takure AO, Adebayo SA, Sotunbi PT, Olapade-Olaopa EO, Okeke LI, Shittu OB. Experience with managing childhood urethrocutaneous fistula at Ibadan. J West Afr Coll Surg 2017;7:44-58.
10. Osuigwe AN, Ikechebelu JI, Okafor PI. Circumcision-related complications in the male: Experience amongst the Igbo's of Southeast Nigeria. Afr J Urol 2004;10:246-51.
11. Brisson PA, Patel HI, Feins NR. Revision of circumcision in children: Report of 56 cases. J Pediatr Surg 2002;37:1343-6.
12. Ekenze SO, Ugwu JO, Onumaegbu OO. Evaluation of neonatal circumcision training for resident doctors in a developing country. J Pediatr Urol 2015;11:6.e1-6.
13. Weiss HA, Larke N, Halperin D, Schenker I. Complications of circumcision in male neonates, infants and children: A systematic review. BMC Urol 2010;10:2.
14. Ademuyiwa AO, Ojewola RW, Elebute OA, Jeje EA, Bode CO. Surgically correctable morbidity from male circumcision: Indications for specialist surgical care in Lagos. Niger J Surg 2012;18:71-4.
15. Obi AO. Giant "double barreled" urethrocutaneous fistula complicating neonatal circumcision – A case report. Arch Urol 2018;1:1-4.