Case Report

Vulvar Calculi: a rare complication of female genital mutilation

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Abstract

Background: Despite global eradication efforts led by the World Health Organization, female genital cutting continues to be a deeply-rooted, common practice in some parts of Asia and East Africa where Ethiopia is situated. It has cultural, social and traditional acceptance within the community for the purpose of attenuating the sexual desire of young women but often causing extreme discomfort and dangerous obstetric emergencies. The traditionally crude and unscientific nature of the practice has led to many immediate and long-term complications among the victims. Vulvar calculi deep seeded in an intermittently painful vulvar mass following type-3 female genital mutilation/cutting is one of the extremely rarelatecomplications of female genital cutting.

Case presentation: A fiveyearsold, Ethiopian, female presented with progressive, intermittently painful, vulvar swelling forthe past six months. Her history is significant for traditional female genital mutilation on the seventh day of life. An incision over the swelling was made and five discrete fragments of calculi were retrieved after which, partial correction of the vestibular narrowingwasperformed. The patient's post-operative course was uneventful.

Conclusion: In areas where female genital cutting is endemic, such as Ethiopia, clinicians need to be aware of exceedingly rare late complications of the practice, which might be misdiagnosed as other solid vulvar swelling and be prepared to treat accordingly with the required appropriate surgical management.

Key words: Vulvar calculi, female genital mutilation, infibulation.

Background

Female genital mutilation/cutting (FGM/C), also known as female genital cutting and female circumcision is defined by the World Health Organization (WHO) as "all procedures that involve partial or total removal of the external female genitalia or other injury to the female genital organs for non-medical reasons" [1]. According to the WHO, it is practiced in 28 countries in Western, Eastern, and Northeastern Africa, in parts of the Middle East, and within some immigrant communities in Europe, North America and Australia [2].

Despite global eradication efforts led by WHO, female genital cutting continues to be a deep-rooted practice in some parts of Asia and East Africa where Ethiopia is situated. In these areas the practice has cultural and socio-traditional acceptance within the community, while attenuating the sexual desire of and creating obstetric agonies foryoung women. The traditional crude and unscientific nature of the practice have led to many immediate and long term complications among the victims. Vulvar calculi deeply seeded in a painless vulvar mass, following type-3 FGM/C is one of the extremely rare scenarios only reported rarely in medical literature as a late complication of female genital mutilation/ cutting.

Case Presentation

We report a case of a five-year-old female child from the Afar region of Ethiopia, who presented with insidiously growing, and progressively painful vulvar swelling for six months after she underwent FGM/C on her seventh day of life. She and her father report she had intermittent urinary flow difficulties with dribbling, and at times painful micturition, with occasionally reddish urine discoloration for which she was referred to our center for further evaluation and management. She was pre-menarchal and the family denies any other genital trauma or foreign body introduction into her genitalia. The family did not notice any skin color changes or discharge around the swelling. She has no lower abdominal pain, bowel habit changes, or similar swelling in other parts of her body. On examination, she appears unhappy and suspicious but otherwise healthy. Pulse rate was 96 beats per minute, blood pressure was 120/78 mmHg, temperature was 37.4 ^{0}c and weight of 15.4 kg. The urogenital system examination revealed an obvious swollen vulvar mass measuring approximately 5x4 cm over the left medial labia majora, extending towards the clitoris. On palpation, the mass was firm in consistency, mildly tender to touch but the overlying skin appeared healthy and shiny. There was an arrowed introitus at the inferior aspect of the vestibule which was infibulated up to the level of clitoris, where a small dimple remained. There was no visible urethral meatus and it was difficult to inspect the hymen, even with further attempts of unfolding the small introitus through which she passes urine. There were no enlarged inguinal lymph nodes (Figure 1).

Figure1: Ill-defined swelling over the left anterior labia majora region with a small opening in the most inferior end of the vestibule outline.Note: The clitoris site is barely seen as a dimple cephalad to the swelling.

Laboratory results revealed hemoglobin of 11.6g/dl, white blood cell count of

$9.25x10^3$/ml, platelet count of $462x10^6$, Cr of 1.1 mg/dl and random serum glucose of 156mg/dl. She tested negative for HIV. Urine analysis was unremarkable. Abdominal pelvic ultrasound was unremarkable. Perineal ultrasound showed egg-shell, echogenic foci in the vulvar subcutaneous tissue measuring 1.7x2.1 cm with posterior acoustic shadow.

After well-informed written consent was obtained from the family, the patient was transferred to the operating theater and under general anesthesia with endotracheal intubation the patient was put in dorsolithotomy position and sterilization of the genitalia was done with alcohol. A vertical curvilinear incision was made over the caudal aspect of the swelling with blunt and sharp dissection using diathermy, beginning from the already existing ventral opening and taking care to avoid the underlying seated urethral meatus [Figure 2a]. **U**pon further fine dissection deeper and cephalad, multiple inclusion calculi [Figure 2b] were visualized and removed with stone forceps (Figure 3a). The redundant labial skin was partly excised and vulvar reconstruction was completed with Vicryl 3-0 using interrupted stitches after washing the incision bed with normal saline and securing hemostasis.

The urethral orifice was identified and catheterization was only possible after the stone removal, due to significant pressure on the meatal opening from the stones compressing it (Figure 3b). The wound was dressed with gauze coated with tetracycline ointment, kept NPO for eight hours with calculated maintenance intravenous fluid with the catheter in place for four days post-operatively and ceftriaxone 50 mg/kg IV BID was given for four days post-operatively.

Her post-operative stay in the hospital was uneventful and the patient was discharged home on postoperative day four with strict recommendations for sitz-bath twice daily and to return in two weeks to the referral clinic for a wound assessment (figure 4).

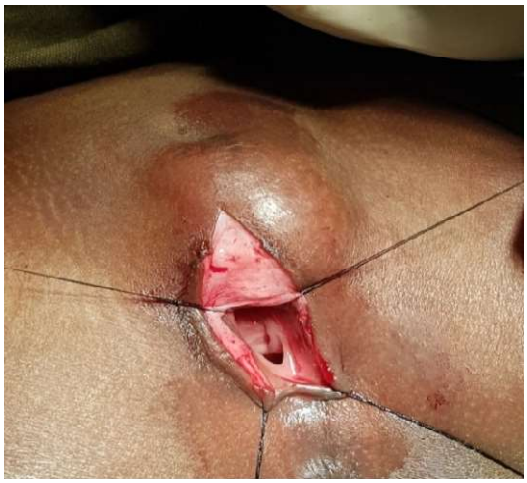

Figure 2a: Incision of the skin overlying the introitus and exposing the area of swelling of the vulva. **Note**: The hymenal ring was found to be open upon careful exam.

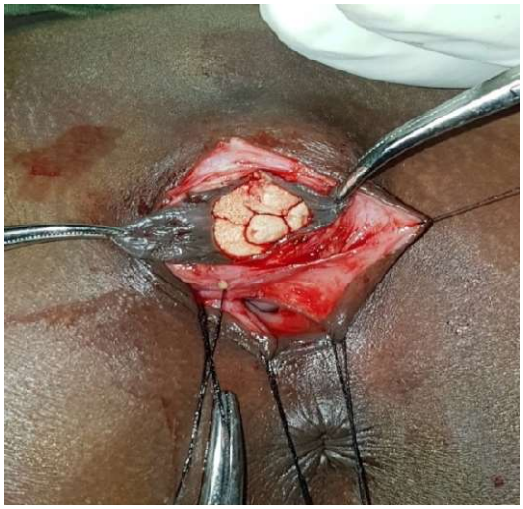

Figure 2b: Multiple inclusion stones/calculi revealed upon further dissection of tissue encapsulated in epithelized labial tissue.

Discussion

The WHO estimates about 100–140 million women and girls around the world have been subjected to the procedure, including 92 million in Africa [1]. The institution classified FGM/C into four types according to the extent of the injury: type I includes removal of the clitoral hood, type II includes removal of the clitoris and inner labia, type III is removal of all or part of the inner and outer labia and usually the clitoris and fusion of the wound leaving a small hole for the passage of urine and menstrual blood. The fused wound is opened for intercourse and childbirth [3]. Type III is the most common procedure in several countries including Sudan, Somalia and Djibouti [4]. Type IV includes several miscellaneous acts, such as piercing of the clitoris or labia and cauterization of the labia [1].

In the 28 countries in Africa and the Middle East for which data are available, national prevalence among women aged 15 years and older ranges from 0.6% (Uganda, 2006) to 97.9% (Somalia, 2006) [5]. There are some regional patterns in FGM/C prevalence. According to Demographic Health Surveys done between 1989–2002, within north-eastern Africa (Egypt, Eritrea, Ethiopia and northern Sudan), prevalence was estimated at 80–97%, while in eastern Africa (Kenya and the United Republic of Tanzania) it was estimated to be 18–38%. However, prevalence can vary strikingly between different ethnic groups within a single country [6]. FGM/C has been documented in several countries outside Africa but national prevalence data are not available [5].

With an FGM/C prevalence of 65.2% among women aged 15-49 in a population of nearly 104.5 million, Ethiopia is second only to Egypt in the total number of women and girls who have experienced FGM/C [7, 8]. Ethiopia outlawed FGM/C in 2004 and practitioners who participate in FGM/C are subject to penalization with a minimum of three months in prison or monetary fines. However, the practice is deeply rooted with a national prevalence of 74% in women of child bearing age [8]. A UNICEF policy briefing document in 2013 on FGM/C in Ethiopia revealed that 23.8 million women and girls had undergone FGM/C countrywide. The Afar National Regional State of Ethiopia is one of those which practice the most severe forms of FGM/C and one of the areas with the most prevalent practice in the nation reaching about 90.8% in Gewane Woreda [9], a district in Afar region from vicinity where the reported case under discussion comes.

A systematic review by Berg found that the most common immediate /short term complications from FGM/C were hemorrhage (5–62%), urinary retention (8–53%) and genital swelling (2–27%), although there were additional studies reporting infection and fever, and three deaths directly attributed to FGM/C [10].

Reported long-term complications of FGM/C include urinary tract infection, dyspareunia and vulvar-vaginal lacerationsduring sexual intercourse [10], epidermoid inclusion cysts [11], and vulvar stones [12] have also been reported. Lower

urinary tract symptoms are more common in women with FGM/C, particularly those with type II or type III FGM/C [13]. Poor urinary flow beneath the infibulation scar may result in symptoms of urinary obstruction, and stasis of urine, which may lead to recurrent urinary tract infection [10] and to urinary or vaginal calculi [12] a rare condition similar to our finding.

Figure 3a: Multiple calcified calculi removed.

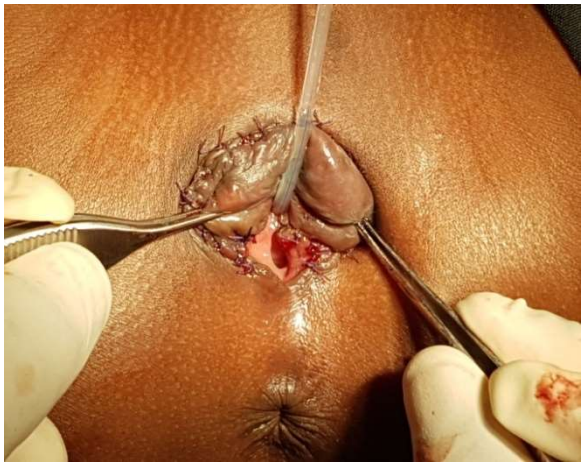

Figure 3b: Urethral meatus identified and foley catheter introduced while partially deinfibulated wound was repaired with absorbable suture in an interrupted fashion.

Figure 4: Second month post procedure revealing a healed wound and functionally open introitus.

Conclusion

Where female genital mutilation/cutting is endemic, clinicians need to be aware of exceedingly rare, late vulvar complications of the practice, which might be misdiagnosed as other solid vulvar masses. Clinicians in these areas should be prepared with the appropriate surgical management skills to treat them.

Conflict of interest

The authors declare no conflict of interests.

References

1. World Health Organization. Female genital mutilation. February2010;2:24.
2. World Health Organization. Eliminating female genital mutilation.2008; 4:22–8.
3. Momoh Female genital mutilation. Radcliffe Publishing.2005:6–7.
4. Nussbaum MG. Judging other culture: the case of genital mutilation,sex and social justice. Oxford University Press; 1999:119–20.

5. UNAIDS, UNDP, UNECA, UNESCO, UNFPA, UNHCR, UNICEF, UNIFEM, WHO. Geneva, World Health Organization, 2008.
6. Yoder P, Abderrahim N, Zhuzhuni A. Female genital cutting in the Demographic and Health Surveys: a critical and comparative analysis. Calverton, MD, Calverton, Macro International Inc. 2004;7.
7. Central Statistical Agency (CSA) [Ethiopia] and ICF (2016). Ethiopia Demographic and Health Survey: Key Indicators Report.2016:45. Available at https://dhsprogram.com/pubs/pdf/FR328/FR328.pdf .accessed 7 July 2019.
8. UNICEF. Female Genital Mutilation/Cutting: A statistical overview and exploration of the dynamics of change, 2013. p.2. Available at http://data.unicef.org/wp content/uploads/2015/12/FGMC_Lo_res_Final_26.pdf
9. United Nations Children's Fund, Female Genital Mutilation/Cutting: A global concern, UNICEF, New York, 2016.
10. Berg RC, Underland V, Odgaard-Jensen J, Fretheim A, Vist GE. Effects of female genital cutting on physical health outcomes: a systematic review and meta-analysis. BMJ Open. 2014;4:e006316.doi: 10.1136/bmjopen-2014-006316.
11. Amu, OC, Udeh, EI, Ugochukwu, AI, Madu, C, Nzegwu, MA. A case of vulval swelling secondary to female circumcision posing a diagnostic dilemma: Case Report. Int. J. Surg. 2012;3(9):431–434.
12. Yusuf L, Negash S. Vaginal calculus following severe form of female genital mutilation: a case report. Ethiop Med J. 2008;46:185–8.
13. Amin MM, Rasheed S, Salem E. Lower urinary tract symptoms following female genital mutilation. Int J Gynaecol Obstet. 2013;123:21–3.