Pregnancy Complicated with a Rare Labioperineal Fusion: A Case Report

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ABSTRACT

Labial fusion is a very rare condition in the reproductive age population. It is defined as the complete or partial fusion of the labia minora and majora. Although uncommon in clinical practice, it is often seen among prepubertal girls and postmenopausal women with hypoestrogenism being its purported primary cause clinically presenting with urogenital concerns. As such, they respond to estrogen therapy, surgical separation, or combination of both.

Herein, underscores a case of a 40 year-old, near term, pregnant woman in labor with labioperineal fusion – an extra rare case. She presented with post void dribbling and dysfunctional penetrative sexual intercourse making her conception rather perplexing. Her vaginal orifice was almost totally obliterated at the midline with dense fibrotic band and an inconspicuous pinpoint <0.5cm opening. She underwent emergency cesarean section with bilateral tubal ligation and postpartum lysis of the adhesion under regional anesthesia with complete recovery within a week. Patient had resumption of sexual activity post-operatively.

Only 2 cases of pregnant patient with labial fusion are recorded in literature. This is the first documented case in this institution.

KEYWORDS: labial adhesion, pregnancy and labial fusion

INTRODUCTION

Simply stated, labial adhesion results when both labia minora adhere together causing a narrowed or obstructed vaginal introitus. It may be partial or complete and is coined in different ways: vulvar fusion, adhesions of the labia minora, labial agglutination, or conglutination of the labia minora and synechia of the vulva.

It involves mostly the prepubertal girls and postmenopausal women; however, it is rare in the reproductive age group. There are very few literatures discussing clinical cases of the labial adhesions among reproductive aged women-this being the second case pregnant at the time of diagnosis. We outline the pregnancy outcome, clinical course, and management of a pregnant indigenous woman with labioperineal fusion.

OBJECTIVE:

To discuss pregnancy outcome, clinical course, and management of a pregnant indigenous woman with labioperineal fusion

CASE PRESENTATION

A 40 year-old, G4P3 (3002), married, Filipino, Umayamnon tribe member was brought in the Emergency Department due to labor pain. She was apparently well, no known allergies, with no heridofamilial diseases and exposure to hazardous and toxic substances. She did not receive formal education and resided in a far-flung, undulating plateau in Bukidnon where resources were scarce.

She has been regularly menstruating since her menarche at 15 with relatively poor feminine hygiene. Her first 2 pregnancies were attended by a birth attendant while her youngest child succumbed to an unknown illness 2 weeks after a precipitous home delivery. No postpartum follow-up was done until her husband noted an irregular skin flap at the vaginal introitus with foul-smelling discharge which spontaneously resolved.

Three months later, there was full closure of her vaginal orifice in the midline. She further noticed difficulty in urination with fullness of her vaginal canal after micturition and dribbling of urine from a small pinhole opening with an unusual post-void dripping noticeable in an upright position. When her monthly menses resumed, mahogany-colored fluid usually admixed with her urine clearing after a week. Then on, they were not able to have a penetrative sexual intercourse rather short anal sex, fellatio, frottage culminating with her husband ejaculating into her perineal area. Attempts at penetration were unsuccessful resulting to labioperineal excoriation and ulceration.

For the next 5 months, mahogany-tinged urine was no longer appreciable and she started to have nausea, vomiting, breast tenderness, and enlarging abdomen. She consulted at a nearby health center where pregnancy was confirmed. She was then referred to a tertiary hospital for further work-up but complied at a later time due to geographical and financial constraints. She further denied any long-term history of vulvar itching or irritation and has no history of genital trauma or infections.

Seen initially at 33 weeks age of gestation, vital signs were within reference range. Abdomen was ovoid, with fundic height of 30cm, audible fetal heart tones at 145 bpm with moderate uterine contractions. Inspection of vulva revealed a complete fusion of the labia minora and majora with dense and thick midline fibrosis obliterating the urethral meatus and vaginal orifice. There were patchy areas of pinkish excoriation and a pinpoint <0.5cm opening devoid of discharges (Figure 1). These findings were consistent with labioperineal fusion but precluded speculum and bimanual pelvic examinations while rectal examination was unremarkable.

Figure 1. A. Patchy areas of pinkish excoriations; B-D. Total fusion of the labia minora in the midline with a dense fibrotic band obstructing the vagina with <0.5cm opening.

She was admitted due to preterm labor with labioperineal fusion. Tocolytics were administered which arrested the uterine contractions and dexamethasone was given to hasten fetal lung maturity. Further work-up showed complete blood count and urinalysis all within normal limits. Patient was then referred to the Dental Department where Class I caries on all molars, calcular deposits and betel nut tooth stain were noted. Fetal biometry revealed a single live intrauterine pregnancy, in cephalic presentation, 33 4/7 weeks by composite sonar aging, placenta grade II, high lying; adequate amniotic fluid index of 9.7 cm; estimated fetal weight of 2,010 grams (appropriate for gestational age); cervical length 3.2 cm; normal both ovaries. She was discharged on the third hospital day and was advised for Cesarean Section and surgical correction of labioperineal fusion at term.

Three weeks after, she was readmitted due to labor pain and subsequently underwent emergency cesarean section, with bilateral tubal ligation. She gave birth to a term male neonate, APGAR 9, 2.8 kg, 37 weeks by Ballard's with no complications. Intraoperative findings showed gravid uterus with grossly normal bilateral adnexae. 4 days post-op, surgical division of the labial fusion was done. A reconstructive pelvic surgeon was invited to perform the procedure (Figure 2)

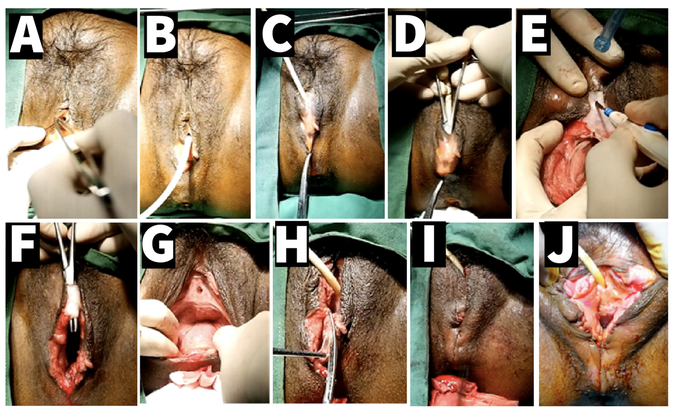

Figure 2. (A) The opening was identified; (B) A foley catheter F16 was inserted into the opening and then a balloon was subsequently inflated with 10cc sterile water; (C) A Kelly forceps was places at the perineum as a marker; (D) Kelly forceps was inserted into the opening towards the fourchette; (E and F) The anterior aspect of the fused labia was then lysed using electrocautery, using the Kelly forceps as a guide; (G) The clitoris, urethral meatus, vaginal canal and cervix were all grossly normal. No fistulous tract seen. (H) Fibrous tissues from the site of the fusion were removed form the anterior to the posterior aspects. (I) The edges of the right labia majora were apposed with simple interrupted sutures using chromic 2-0. The same procedure was done to the left labia majora. The bulbocavernous muscles were identified and reapposed using crown technique. The superficial transverse perineal muscles were identified and reapposed using overlapping simple interrupted sutures using Novosyn 2-0. (J) Hemostasis ensured.

Foley balloon catheter was removed and bladder training was done post-operatively with 9% residual urine. Patient was then discharged after 3 days.

Patient followed up at Out-Patient Department a week after with well-coaptated surgical site and no recurrence of labial fusion (Figure 3). She did not report nay urinary symptoms and was able to result normal sexual activities 3 weeks post-surgery. No recurrence of labial fusion seen on the 3rd week following surgical lysis of adhesion.

Figure 3. Post operative follow up showed a well-coaptated wound.

DISCUSSION

Labial fusion is a rare condition where two flaps of skin on wither side of the opening to the vagina are joined together. In prepubertal girls where labial fusion is more common, the incidence is estimated to be 0.6-5%. The overall prevalence of labial fusion is 1.8%, with a highest incidence seen at 13 to 23 months of age (3.3%). This benign condition is also common among postmenopausal women, rare in the reproductive age group, and extremely rare among pregnant women.

Several theories have been linked to its development but the most plausible and popular postulate was due to low serum levels of estrogen seen among prepubertal girls and postmenopausal women. Aside from hypoestrogenism, the latter cohort is predisposed due to inflammation-repair process of the vaginal epithelium inciting the unwanted fusion. However, labial adhesions may occur in normal estrogen levels. Ideally, serum FSH, LH, and estradiol are needed but in this case, a normally appearing ovaries on ultrasound and the fact that she able to conceive validates the inference that these values are within normal reference range.

In women of reproductive age where estrogen level is abundant, there are several other causes of labial adhesions. Literatures report female circumcision, diabetes, herpes simplex, lichen sclerosis, pemphigoid, caustic vaginitis, poor hygiene, surgical trauma or those related to sexual abuse or a fall, vaginal laceration following childbirth to be the risk factors for this development. It is a very rare complication following vaginal delivery.

Poor hygiene sets the stage in this case. There is a high likelihood that had vulvoperineal trauma following unattended precipitous home delivery. These factors then cause volvuvaginal infection of the labia minora. The tissue damage resulting from local trauma and irritation lead to production of fibrinous exudate facilitating union of the labial epithelium. Frequent failed attempts at penetrative sexual intercourse can also cause local irritation and local inflammation which perpetuate the inflammation-repair process and strengthen the midline adhesion. Vulvar edema following local trauma cause the denuded labial epithelium to coalesce particularly in this closely-apposed area resulting to synechial formation.

In women of the reproductive age group with posterior adhesions, difficulty in engaging in sexual activity is frequently encountered as in this case. They were not able to achieve vaginal penetration such that frottage only contributes to local irritation and further fusion. Already confounded by interplay of multifactorial etiologies, the fact that she conceived with an almost complete labial adhesion makes this case baffling. In an attempt to explain this rare conception, fistulous tract has been investigated which may be possible in anal intercourse but rectovaginal fistula was not seen in this case.

Standard treatment for prepubertal girls with labial fusion has included topical estrogen cream, betamethasone administration, manual separation, or surgery. In postmenopausal women, surgical liberation combined with postoperative estrogen administration proves complete recovery.

The most effective treatment of labial synechia is surgical division under local anesthesia in women of reproductive age as seen in this case. Surgical lysis of labial fusion under regional anesthesia, a novel procedure in this institution, was done under the guidance of a reconstructive pelvic surgeon.

The goals of management are to address hypoestrogenic state, lyse the adhesion, and prevent recurrence. Post-operatively, she was given Clindamycin 300mg TID for 7 days. No topical estrogen or antibiotics were prescribed and mechanical dilator to prevent fusion were also not used. In some patients who went on to surgery, recurrence is a possible sequela. Recurrence is common until a patient goes through puberty.

Prevention of labial adhesion should begin with proper feminine hygiene practices and prevention of and minimization of vulvar irritation in reproductive aged women where estrogen is abundant. A good postpartum perineal care is imperative. Regular inspection of the vulva should be taught in patients with lacerations. Manual separation of edematous and lacerated labia should prevent labial adhesion. Furthermore, resumption of sexual intercourse whenever comfortable can also prevent delayed adhesion.

CONCLUSION

Postpartum labial adhesions are rarely reported in literature. Further research should attempt to establish the actual incidence of this condition since it may be underreported.

The reproductive age women acquired a very rare complication of childbirth, labial fusion. She presented with the typical urinary symptoms and difficulty in sexual activities. Whilst having this benign condition, she was able to conceive and had an uncomplicated cesarean delivery. Lysis of labial adhesion was achieved surgically with complete and rapid recovery.

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