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CASE SERIES OF UNUSUAL & VERY RARE VAGINAL PATHOLOGIES

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ABSTRACT The term vagina is from Latin meaning "Sheath" or "Scabbard" and may also be referred to as the **birth canal** in the context of pregnancy & childbirth. The vagina & vulva has evoked strong reactions in societies from time immemorial and is symbolic with female sexuality, including negative perceptions, cultural taboo's. In common speech vagina refers to the female genitals in general & not specifically to its dictionary & anatomical definitions wherein vagina refers exclusively to specific internal structure. (Wikepedia). Pelvic examination are performed by trained Gynecologists to examine for unexplained symptoms of discharge, pain, unexpected bleeding or urinary problems. The vaginal opening is assessed for position, hymen, growth, ulcers or any other pathology as a routine. We present here three very interesting & rare Case reports of vaginal pathologies that we came across during examination in Gynae OPD. We are discussing here three extremely rare cases of vaginal pathologies where in one case a stone was found in the vagina around a multiload Cu T thread in a patient with history of VVF following Ceasarian section. The second case pertains to a congenital condition called Vacteryl Syndrome where patient has grown into an adult without being aware that anal opening was absent &she was passing stool through vagina. The third case is also unique wherein post delivery the patient's vagina was packed with gauze soaked in some chemical probably caustic soda to control PPH, resulting in vaginal stenosis.

KEYWORDS : Vaginal Stone. IUCD, VVF, Absent Anus, Vaginal Stenosis

CASE 1 : RARE CASE OF VAGINAL STONE IN A PATIENT OF VVF WITH HISTORY OF PPIUCD INSERTION

INTRODUCTION

Vaginal stones (calculi or vaginoliths) are an extremely rare condition that has been registered as case reports [1] and most gynecologists may never see a vaginal stone case throughout their career . The 1st case of vaginal calculus was reported by Halban in 1900 in patient with vaginal cystocele [2] Cases reported in the medical literature are usually associated with urinary tract abnormalities, such as urethra-vaginal or vesicovaginal fistulas.[3]

Vaginal stones are classified into primary and secondary depending on the presence or absence of a foreign body [4] [5] The association of vaginal stone with vesicovaginal fistula is not uncommon these stone are commonly associated with urinary symptoms like dysuria and urinary retention and vaginal outlet obstruction.[6] [7]

Primary vaginal stones form from the deposition of urinary salts as a result of urinary leakage in association with a urethrovaginal fistula that developed after trauma, vaginal stenosis, or scarring after trauma or surgery; ectopic vaginal ureter; neurogenic bladder; hypospaidias, urethral diverticulum or vaginal outlet obstruction [8] Stasis of urine induces deposition of urinary salts.When infection by bacteria such as Proteus mirabilis, Klebsiella, or Escherichia coli occurs the metabolic activity of the bacteria can change the normally acidic pH of the vagina to more alkaline conditions and initiate creation of vaginal stones [9]

Secondary vaginal stones are the result of crystallization of urinary constituents around a foreign body in the vagina[8] Secondary vaginal stones result from the crystallization of urinary constituents around a foreign body in the vagina [10] [11]

Case Report

A 35 yr old Para 3 L 3 with history of Previous two caesarian sections reported to the OPD with the complaint of dribbling of urine from vagina for one and a half year following her last childbirth by LSCS. She also complained of continued spotting of blood per vaginum nearly every alternate day since her delivery. She further gave an unusual history that her husband used to get penile abrasion & injuries after coitus and he used to feel something hard inside the vagina, as if there was a stone inside. Her past history was not significant.
Her Last menstrual Period was six days back and her menstrual cycles were regular but she continued to have spotting for the whole month every cycle for the past one & a half year. Her obstetrical history revealed a full term normal delivery of a female child 8 years back followed by two male child both delivered by Ceasarian section 6 years & one and 1/2 year back. Post Ceasarian Copper T –Multi Load 375 had been inserted during her last childbirth.

Physical examination was did not show any significant positive finding. Gynaecological examination of vagina per speculum showed a white stone like mass inside the vagina and cervix could not be visualized. Watery discharge from vagina with uriniferous smell was observed and suggested a vesico vaginal fistula. Further history revealed that she developed this complaint soon after her delivery in the postoperative period itself, however she had not been advised any treatment for it.

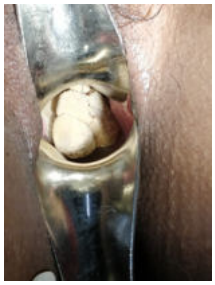

Fig 1. Vaginal Stone seen on Per Speculum Examination

Fig 2 : Stone Removed From Vagina

Fig 3 : Vaginal Stone seen hanging on the thread of Cu-T

Fig 4: IUCD Thread of ML-375 deeply embedded in stone

On Per vaginal examination the mass felt like a stone as it was stony hard and was gently manipulated and removed by the examining fingers. A foul smelling stone of approximately 4 cm by 5 cm came out with the IUCD and the thread of the Cu T –Multi Load-375, was seen embedded in it.

On further examination urine could be seen dribbling fro the anterior upper end of the vault. Cervix was pulled up high and was seen with difficulty. Uterus was normal size, anteverted with slightly restriced mobility.

An ultrasound done a month earlier had reported an IUCD in situ in the uterine cavity but had made no comment about the presence of any foreign body in the vagina and had probably been missed during the USG examination. The extreme rarity of this condition means that most radiologists may never see a case of a vaginal stone throughout their life. The diagnosis of vaginal stones can be difficult and requires that the physician is highly suspicious of this possible diagnosis.

DISCUSSION

Least common is vesicouterine fistula which is typically caused by caesarian sections, It counts 1-4 % of all urogenital fistulas.On the other hand foreign bodies such as IUDs can penetrate the uterine wall and the urinary bladder forming vesicouterine fistula [12]

The association of vaginal stone with vesicovaginal fistula is not uncommon these stone are commonly associated with urinary symptoms like dysuria and urinary retention and vaginal outlet obstruction.[6] which however was not observed in this particular patient. Here in our case poor girl has large urinary fistula since 2 yrs, due to true incontinence she developed a habit to hold urine inside the vagina which acts as a reservoir for urine stasis more over the presence of IUCD thread in the vagina, infected tissue of the bladder and the vagina may have been a possible etiological factor for vesico-vaginal stone formation hence it is difficult to ascertain in our case if the Colpolithiasis (vaginal stone) was primary or secondary. Vaginal stones are often mistaken for bladder calculi on plain radiography. Imaging studies including USG, CT, excretory urography, cystourethrography, MR urographyand Cystoscopy may be useful in diagnosing this rare condition.[3]

CASE REPORT 2 : VATER /VACTERL Syndrome Associated With Unicornuate Uterus And Non-communicating Uterine Horn

INTRODUCTION

VATER/VACTERL syndrome is an association of congenital anomalies of the vertebra/ anus/ cardiac/ trachea/ esophogus/ radius/renal/limb of unknown etiology (1–3). **3** The acronym VATER, first coined in 1973, describes an association of congenital defects: vertebral, anorectal, tracheoesophageal,and radius or renal (10**). 3** This later included cardiac and other limb abnormalities.**3** Associations between uterine and renal or anal anomalies are recognized. (4, 5). **3** Clinical and laboratory based analysis should not yield evidence of an alternative diagnosis. The diagnosis of VACTERL-H syndrome was based on the cluster of findings [1 **1** There is still no firm consensus regarding strict diagnostic criteria, though most clinicians and researchers require the presence of at least three component features for diagnosis **4** The VATER/VACTERL syndrome is used synonymously Incidence of VACTERL syndrome is very rare, 1 -9 in 100,000 live born.[1] **1** We believe this is the second or third documented case with both the VATER /VACTERL syndrome and a unicornuate uterus and non-communicating uterine horn.

This case is probably one of the rarest of rare case where the patient is 23yrs old patient is passing stools through the vagina and neither she nor her husband were aware of the absent anus and the fact that defecation was being done through the vagina. It is also surprising that even her parents did not take note of this or maybe knew about it but did not reveal it for social reasons and fear for their girl child

CASE REPORT

A 23 year old female Para 0 Abortion 1, married for one and a half year, came to OPD as a case of secondary infertility with complaints of pain abdomen and fever since past 4 days. She also had complaints of intermenstrual spotting per vaginum since 4 days. Her UPT (Urine Pregnancy test) was negative. Patient's obstetrical history revealed that she had a spontaneous abortion one year back at 3 months of period of gestation. There was no history of trauma, any excessive bleeding or any blood transfusion following her abortion & neither was it followed by dilation and curettage as the patient did not go to any doctor for check up.. Her LMP was on There was no relevant past or family history.

On general examination her vitals were stable and she had mild pallor. Cardiovascular and respiratory systems were clinically normal. Per abdomen no abnormality was detected. There was no guarding, tenderness or rigidity of the abdomen. She had a small rudimentary extra digit in her left hand.

On perineal examination it was noted that anal canal opening was not visible nor any anal dimple was seen and anus appeared to be absent.

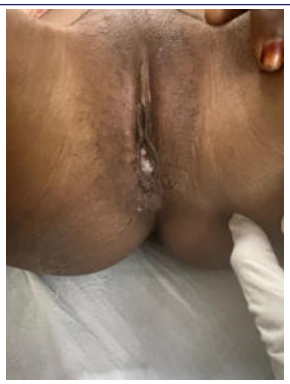

Fig 5. Vacteryl Syndrome : Appearance of Perineum

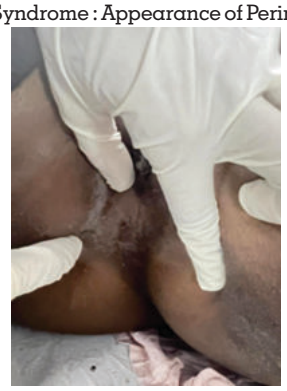

Fig 6. Vacteryl Syndrome : Absence Of Anal Opening

On Per speculum examination cervix was seen pushed up anteriorly and was visualized with difficulty. It was normal in appearance with mild circumoral erosion. There was mucoid dirty discharge in the vagina. Anteverted uterus of a size of 10-12 weeks was palpated Per vaginum. On removal of fingers gloves were found to be stained with faecal matter. Her history was asked again if she had any difficulty in passing stool but she denied any problem. It was then realized that maybe she was passing stool through vagina via a recto-vaginal opening as her anal opening was absent. Per speculum examination was repeated to look for any obvious opening but could not be visualized as patient had become very unco-operative. A repeat vaginal examination revealed a small rectal opening through vagina thus explaining her lack of knowledge about her condition.

Her USG showed an anteverted uterus with uterine size 11x4.2x5.5 cm. Uterus and cervical canal was dilated and filled with echogenic material. Left ovary showing hemorrhagic cyst of 36 x 40 mm. Right ovary was visualizes and found to be normal. The Pouch of Doughlas was empty. She has unilateral Kidney. Right kidney was not seen at its normal location. Single hypertrophied left kidney of 12 cm was seen.

HSG revealed a unicornuate uterus with rudimentary right horn containing endometrium and right cornual block. All other haematological investigations, liver & Kidney function tests were normal.

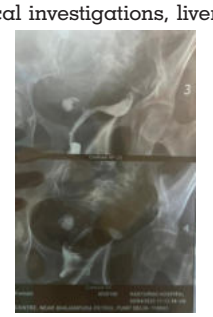

Fig 7: Vacteryl Syndrome :HSG showing Unicornuate Uterus With Rudimentary Horn

Patient was managed conservatively for her pain abdomen & improved significantly within a day and was discharged. She was referred to a GI surgeon for opinion if any further treatment was required for absent anus and to a nephrologist for her single kidney.

DISCUSSION

A central and critical question regarding VACTERL association hinges on the causes and is yet to be resolved. The etiologies of VACTERL association remain largely unknown primarily due to its rarity, it's clinical and causal heterogeneity, the typical sporadic nature of the disease, and several overlapping conditions. Coupling insights from biological models with newly available genomic technologies may begin to offer more answers about causation in the near future. 4

Embryological studies suggest that VACTERL has multiple polytopic developmental field defects [1]**1**. A disruption in differentiating mesoderm in the first 4–5 weeks may have lead to the different malformations of the VACTERL spectrum. It may also be linked to various other aetiological factors including diabetic mother, infertility treatment, exposure to hormones oestrogen or progesterone, anticonvulsants, folic acid antagonists or alcohol during the period of embryogenesis causing mutations in the genes namely ZIC3, HOXD13 and FOXF1. 1

The co-existence of anamolies of multiple organs simultaneously may be attributed to the proximity and simultaneous development of these organs..This case documents an association between VATER/ VATERL syndrome and a unicornuate uterus with a noncommunicating uterine horn.. This case emphasizes a multidisciplinary approach in the diagnosis and management of complex congenital pelvic anomalies **3** The important question relates to improving the health of affected patients and manage the psychological impact on their families. Recognizing that some congenital malformations may be subtle yet medically important, such as vertebral anomalies that can result in severe back pain later in life or renal anomalies that can predispose to infections, nephrolithiasis, and declining renal function. Managing clinicians must keep these long-term issues in mind. **4** The management of patients with VACTERL association can be intricate & complex, and the nuances of treating issues related to each component feature are not covered in this manuscript.

CASE REPORT 3 : POST PARTUM IATROGENIC VAGINAL ATRESIA

INTRODUCTION

Acquired Vaginal Atresia or Gynaetresia is seldom encountered in the present era and is often attributed to certain cultural practises still existing in African countries. In India it is usually secondary to botched vaginal deliveries conducted by untrained Birth Attendants in rural areas. We report here an interesting case of vaginal atresia following post partum insertion of a 'cloth pack' in vagina, possibly to stop the bleeding from a perineal tear. Home deliveries by untrained birth attendants should be discouraged with a concerted effort made by the Government of India to give the rural population of India access to institutional deliveries, failing which at least the attending Birth Attendant should be professionally trained so that such complications can be avoided. Secondary vaginal atresia needs extensive counselling of the couple not only prior to surgery but also post operatively for successful results of surgeries performed.

Case Report

A 21 year old Primipara diagnosed as a case of vaginal atresia was referred to our hospital by a Private Practitioner for further management. The patient gave the history of a full term normal vaginal delivery, two & a half years back at her mother's residence in the village. Her delivery had been

conducted by a 'Local Dai' (untrained Birth Attendant) and although her delivery was uneventful, her post partum period was complicated with more than average bleeding per vaginum & severe pain, which continued for the next two days. She again consulted the same Dai, who inserted a 'cloth pack' soaked in some unknown solution into her vagina and asked the patient to leave it in situ for ten days. The solution could have been caustic soda, which is easily available in most Indian households as it used as a bleaching agent for washing clothes, but cannot be definitely commented upon as the patient was unable to tell us. Her bleeding stopped thereafter and pain gradually subsided over the next few days.

After eight months she went back to her husband and started cohabiting with him, when she discovered that sexual intercourse had become very painful & difficult with full penile penetration not being possible. Taking her time, she consulted a local doctor, probably not a qualified gynaecologist, who without doing an internal vaginal check-up treated her for Candidial infection. However when the patient had no relief in her symptoms she consulted a Gynaecologist who admitted her for examination under anaesthesia. On examination under anaesthesia the doctor discovered that the cervix and vagina were badly fused to each other. She made an attempt to break the adhesions & dilate the cervix but abandoned the procedure midway as she was not successful and patient started bleeding. The patient was then referred to Kasturba Hospital for further management.

On inspection, the urethral orifice, Mons Pubis were normal but vagina appeared blind. Labial skin around the Fourchette and lower part of vulva was seen pushed high up inside the vagina for about an inch, as if pulled up inside due to scarring of the vaginal wall.

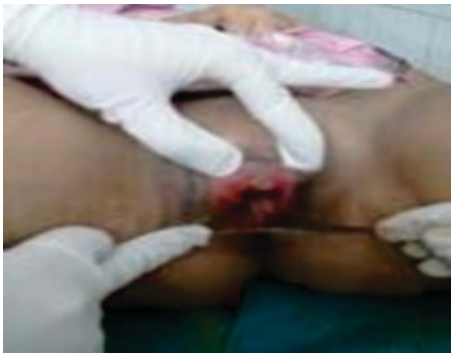

Fig 8: Vaginal Stenosis :Appearance of Vagina & Vulva on separation of Labia

Two fingers could be inserted for about half inch on per vaginal examination but cervix could neither be felt nor visualized. Nearly the whole length of vagina was obliterated with dense adhesions and fibrous tissue. Per rectal examination revealed a normal anal opening.

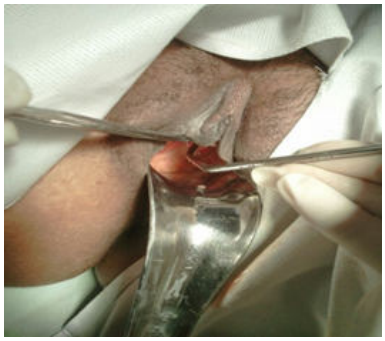

Fig 9 : Vaginal Stenosis :Attempt to identify opening in vagina with probe

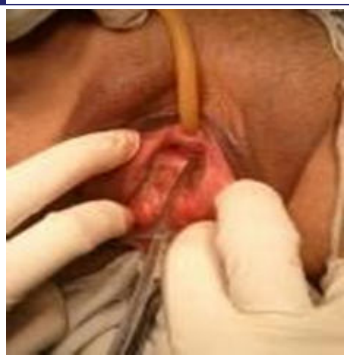

Fig 10 : Fibrosed vagina fused with cervix and normal urethra (catheterized)

Patient was planned for Mc Indoe's Vaginoplasty and all routine investigations for anaesthesia were done. Ultrasonography was unremarkable and showed normal pelvic organs. Vaginoplasty was performed under general anaesthesia. A transverse incision was given in the centre and upper & lower flaps were lifted. Space was created between the vagina & rectum by blunt & sharp dissection, bladder was dissected away meticulously and dissection continued until cervix could be visualized. Fig 4, Fig 5, Hegar's Dilator was introduced to dilate the cervix and a small amount of altered blood was seen coming out through the external os. The cervix was mobilized and cleared of all adhesions around it by blunt and sharp dissection, until vaginal space was well defined. The labial skin taken up inside the vagina around the fourcette was then released by an incision similar to that given in perineorraphy. The cicatrized tissue was removed and vaginal mucosa and skin were sutured with interrupted sutures.

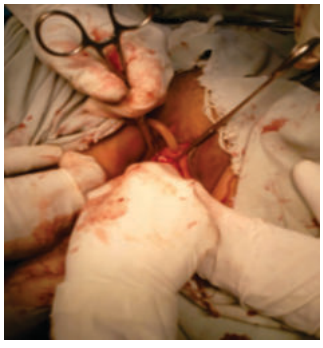

Fig 11 : Creation Of Vaginal Space By Finger Dissection

Haemostasis was achieved and a vaginal mould covered with a water-insoluble, haemostatic device Spongistone (Gelfoam) - a sterile Compressed Gelatine Sponge made of purified porcine skin gelatine was then inserted vaginally and left in situ.

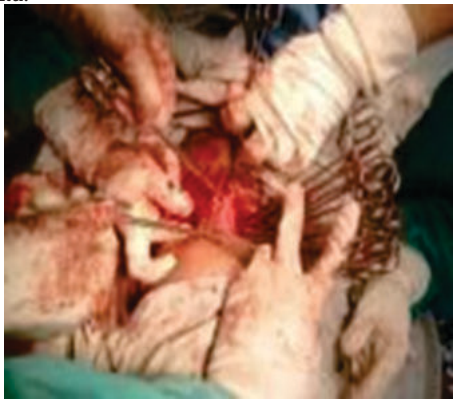

Fig 12 : Vaginal Dissection Completed And Cervix Mobilized

It was retained in position by a tight T-bandage. Patient stood the procedure well and her post operative period was uneventful. Foley' catheter was retained for 3 days and intravenous antibiotics were routinely administered. Vaginal mould was changed on Day 4 and on examination there was no pressure necrosis or ulcerations, healing was good and vaginal depth appeared near normal. Patient was trained in daily removal and self insertion of the mould with aseptic precautions and discharged. She was advised to come for regular follow up for the next four weeks and remained in follow up until she was perfectly fine and had resumed normal sexual activity.

Fig 13 : Vagina & Vulva after surgery

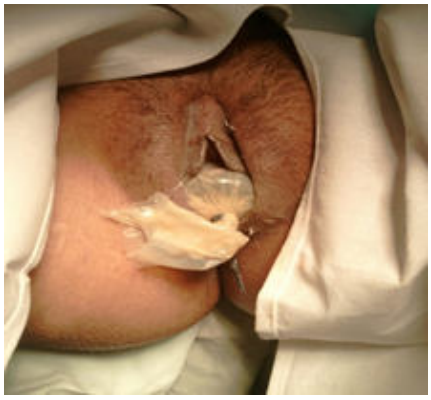

Fig 14: Patient in follow up with inserted mould

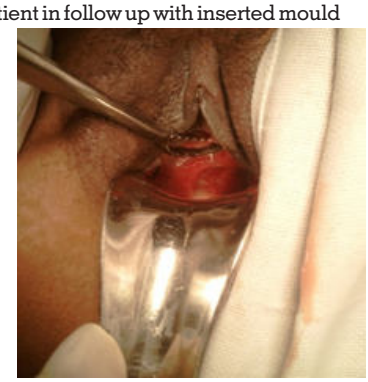

Fig 15: Patient In Follow Up Showing Normal Vaginal Space & Mobilized Cervix

DISCUSSION

Vaginal atresia can be congenital or acquired. Incidence of congenital or primary vaginal atresia is 1 in 4000 to 5000 live female births whereas secondary gynaetresia is extremely rare and exact incidence has not been reported in literature[1,2] Acquired vulvo vaginal stenosis is seldom encountered in the present era but several African countries are known entities for cultural practises resulting in gynaetresia even today. In a Nigerian study the prevalence rate was 7 per thousand with peak incidence of 20-30 years. [3, 4] Iatrogenic vaginal atresia usually occurs secondary to botched or badly managed vaginal deliveries where neglected vaginal injuries and perineal tears result in subsequent scarring. Untrained female staff i.e. 'Local Dai's, play a major role in increasing the incidence of such complications in our country. Neglected foreign bodies following induced abortion, untreated pelvic inflammatory diseases and post cancer radiotherapy constitute other important causes of vaginal adhesions. [3,5]

In rural India unattended home deliveries, deliveries by untrained birth attendants is still a very common practise. These Local Dai's as they are called are not trained professionally to conduct vaginal deliveries, nor are they familiar with the techniques of episiotimies or repair of perineal tears. Institutional deliveries of the entire pregnant population is virtually impossible in a country like India hence training of Local Birth attendants would bring down the rate of such complication to a great extent[3]

Chemical vaginitis resulting from insertion of Caustic vaginal pessaries, herbal mixtures for various reasons was another major cause of vaginal stenosis. Paula et al reported a case of labial adhesion as a result of caustic vaginitis in a post partum patient[6] much similar to our case where some kind of chemical cauterization was attempted by the Local Dai probably to arrest the bleeding per vaginum, which may have been caused by some degree of perineal tear. Kamal et al had reported a similar case of pinhole vagina following mismanaged vaginal delivery [7]

Labial adhesions in vaginal stenosis are usually dense in nature and cannot be successfully treated without surgical interventions which may vary from a simple incision to complicated vaginoplasty depending on the extent of tissue damage[3] Methods commonly used by Gynaecologist comprises of Mc Indoe's Vaginoplasty where space is created in connective tissue between bladder & rectum with use of a split thickness graft obtained from the patients buttock or anterior thigh to line the neovagina.[8] Several modifications of Mc Indoe's procedure include the use of human amnion, sigmoid colon, ileum [9] pudendal thigh flaps [10], fascio-cutaneous flap, Gracilis myocutaneous flap, Labia minora flaps, flaps raised following tissue expansion of labial pocket, peritoneum, bladder mucosa and Amnion [11(B-8)], the Interceed absorbable adhesion barrier[8] autologous buccal mucosa[5] and recently artificial dermis and recombinant basic Fibroblast Growth Factor[12] are used for Neovaginal linings.

Free skin grafts, peritoneal and local skin flaps, bladder mucosal grafts often results in scarring of the patient. Bowel segments apart from abdominal scarring also has the disadvantage of bowel obstruction, secretions, unpleasant odour and mucosal ulcerations [1] Human amniotic membrane is readily available free of cost and is already being used by surgeons for protective biological dressings of surgical wounds[13] There is no evidence of immune rejection when amnion is transplanted subcutaneously as amnion does not express HLA –A, B or DR antigens.

Amnion has the additional advantage of possessing antimicrobial properties due to Lysosome production, hence decresing the chances of post operative wound infection. The uncontaminated amniotic mesenchymal surface when applied over the raw vaginal surface results in firm adhesion of the amnion which thus facilitates re-epithelisation of the vagina and also protects the underlying granulation tissue. Amniotic membrane with meconium staining, suspected chorioamnionitis, HIV, Hepatitis B and other transmissible infections should not be used for grafting[2]

Conflict Of Interest : We have no conflict of interest to declare

Ehical Clearance : Not required

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