CASE REPORT

Acquired gynatresia

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SUMMARY

Vaginal stenosis (gynatresia) is commonly congenital, occurring as a part of Mayer-Kustner-Hauser syndrome. Those occurring postabortal or postpartum are rarely seen nowadays. Here, we report two cases, one in which there was complete stenosis postpartum following a retained gauze and another case, with chemical vaginal burns following attempted abortion by a local untrained midwife.

BACKGROUND

Acquired vaginal stenosis or gynatresia is a rare problem in the developed world but common and grossly under-reported in the developing world where it may be the result of intervention by untrained health workers, quacks or as a result of infection. In the Arab world, rock salt was inserted everyday in a postpartum woman to restore the vagina to a nulliparous state resulting in complications like sepsis, vaginal stenosis, haematocolpos, haematometra, etc.¹ The other part of the world where acquired gynatresia is common is Africa and especially Nigeria, where incidence is reported to be around 7/1000² women, with the commonest cause being insertion of caustic vaginal pessaries, birth injury and also as a complication of vesicovaginal fistula repair.³ In the developed world, it has been reported to be associated with chronic graft-versus-host reaction⁴ or after radiotherapy. Here we report two cases, one with postpartum total vaginal stenosis secondary to negligence by a healthcare worker and another case secondary to chemical vaginal burns.

CASE PRESENTATION

Case 1

A 30-year-old woman para 1 with no live issues reported to the outpatient department (OPD) of our hospital with secondary infertility, dyspareunia and dysmenorrhoea for past 10 years. Except for the dysmenorrhoea her menses were regular. According to the patient, she had delivered vaginally, a live full-term female baby 10 years back after a prolonged labour at a primary health centre (PHC). The baby expired soon after birth. According to the patient, she received an episiotomy which was sutured, soon after which she was discharged. Post delivery, according to the patient, she developed vaginal discomfort and discharge and on follow-up at the same PHC 2 weeks after delivery, a neglected gauze swab inserted at the time of delivery was removed from the vagina. The patient was discharged with the advice of abstinence for 2 months. After 2 months, when the patient attempted intercourse she had severe

Figure 1 Complete vaginal stenosis.

dyspareunia and penetration was impossible. On examination, there was severe vaginal stenosis and even a small paediatric Sim's speculum could not be inserted. A per vaginam examination was not possible due to intense pain. The patient was posted for examination under anaesthesia (EUA) with vaginal adhesiolysis. EUA revealed a cicatrised vagina half an inch in length and a small hole posteriorly. There was dense fibrosis between the anterior and posterior vaginal walls (figure 1).

Case 2

The patient was a 40-year-old lady, para 5, came to the emergency room reporting cessation of menses for 2 months, bleeding and pain for 6 days. According to the patient, she was approximately 2 months pregnant when she consulted a local Dai (untrained midwife in rural India) for termination of pregnancy. The Dai inserted some chemical into the vagina. Soon after the insertion, she developed intense pain in the lower abdomen and buttocks and bleeding per vaginam. On examination, the general condition of the patient was poor, she was dehydrated, febrile and had a pulse of 120/min, her blood pressure was 100/70 mm Hg. The patient had marked pallor but there were no other significant findings in systemic examination.

Per abdominal examination revealed a bluish-black discolouration of the lower abdominal skin and inner aspect of the high extending to buttocks (figure 2). The lower abdomen was tender; however, there was no organomegaly or any sign of collection in the abdomen.

Local examination of the genitalia revealed bluish-black, hypopigmented areas around the vulva suggestive of chemical burns (potassium

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Figure 2 Chemical burns on the mons and labia.

permanganate) with bullae formation in some areas. Per speculum examination showed same findings in post wall of vagina and around the cervix. There was profuse foul smelling discharge seen coming from the cervical os. The cervix was torn and macerated due to burns and infection. On per vaginam examination the uterus was bulky, soft and tender. The adnexa was clear but the cervix was ballooned up with a tear on the left side. The cervical os was open and products of conception (POC) could be felt through os. There was tenderness present on pelvic examination.

INVESTIGATIONS

Case 1

Haemoglobin:11.8 g%, total leucocyte count (TLC): 9500/mm³, differential leucocyte count: neutrophils 76%, lymphocytes 20%, eosinophils 4%, liver function test: within normal limits, blood urea: 27.3 mg/dL, serum creatinine:1.0 mg/dL; ultrasonography showed a normal sized uterus with normal ovaries.

Case 2

The investigations were as follows: haemoglobin 6 g, TLC 8500/mm³ with 80% neutrophils, blood urea 100 mg and serum creatinine 1.8 mg and rest of the investigations were normal. USG showed a bulky uterus with minimal fluid in the pouch of Douglas and some retained POC in the uterine cavity.

TREATMENT

Case 1

Adhesiolysis was carried out with careful sharp dissection till the cervix became visible followed by regular vaginal dilation by placement of a mould made of alginate a material used for dental impression. She was advised to wear it 24 h initially later decreasing it to night insertions only.

Case 2

The patient was put on broad spectrum antibiotics with oxygen, blood and intravenous fluids, a barrier cream was given for local application on burns. However, the patient developed cellulitis in the right thigh after 48 h in hospital and went into septic shock. She needed debridement under general anaesthesia. She responded to third-generation cephalosporin and resuscitation. After debridement daily cleaning of the vulva and vagina was carried out.

OUTCOME AND FOLLOW-UP

Case 1

Patient reported to the OPD after 6 weeks with a well-formed, epithelialised vagina and cervix (figure 3). Patient also reported satisfactory intercourse. We are happy to report that the patient conceived after 2 months of the adhesiolysis operation.

Case 2

On a follow-up visit, it was seen that the vagina had considerably shortened and so the patient was advised daily vaginal self-dilation with a mould and to start sexual intercourse as soon as she was comfortable. Patient recovered fully with a well-developed vagina.

DISCUSSION

Congenital vaginal aplasia or agenesis occurs as a part of MRKH (Mayer-Rokitansky-Kustner-Hauser) syndrome which has an incidence of about 4000–10 000 female live births.⁵ However, acquired vaginal cicatrisation or stenosis is of infrequent occurrence. Vaginal stenosis is typically defined as an inability to insert two fingers on examination⁶ Cases of acquired vaginal cicatrisation or stenosis reported from developed world occur associated with chronic graft-versus-host reaction either after receiving a bone marrow transplantation,⁷ or as a part of Steven-Johnson syndrome⁴ or after toxic epidermal necrolysis⁸ but more commonly after radiation therapy. Vaginal stenosis has also been reported after retention of a foreign body in the vagina.⁹ Only one case similar to our first case has been reported in the literature. In this case, postpartum vaginal agglutination occurred because of hypoestrogenic state during lactational amenorrhoea.¹⁰ However, in our case, vaginal stenosis occurred probably because of sepsis following retention of a swab in the vagina.

Vaginal stenosis as a result of herbal vaginal pessary insertions is commonly reported from African countries especially Nigeria, where it is the commonest cause of vaginal stenosis followed by

Figure 3 Same patient (case 1) after adhesiolysis and vaginal dilation.

obstetric injury and female genital mutilation.^{11 12} These caustic vaginal pessaries are inserted for treatment of fibroids, infertility and prolapse.¹² Vaginal atresia can also occur as a complication of fistula repair.³

Vaginal stenosis may have long lasting psychological effects and loss of sexual function as seen in our first case.

Many procedures have been used for the treatment of acquired gynaetresia. A procedure involving pudendal thigh flap has been described in the literature and has the advantage of being simple and reliable with robust tissue that can be used to create a neovagina with a natural angle and with sensation.¹³ It can be easily applied in single-stage reconstruction of genital deformities. This, however, has its own set of problems like hair growth and numbness in the vagina. Sigmoid vaginoplasty also gives good results but has the disadvantage of excessive mucus production and periumbilical pain.¹⁴ These procedures though associated with good results may be difficult to perform and require surgical expertise. Acquired vaginal stenosis may be treated by a simple division of adhesions followed by regular vaginal dilation as restenosis is a common complication. Dilation causes stretching of mucosa, mitosis and growth of new mucosa. Adhesiolysis is easy to perform and combined with vaginal dilation has a good success rate.

Learning points

- ▶ Most commonly vaginal stenosis is congenital; however, it can occur associated with an autoimmune reaction or after radiotherapy.
- ▶ It may rarely also occur because of vaginal mutilation caused by mismanaged puerperium or septic abortions.
- ▶ Treatment can be accomplished by simple division of adhesions followed by regular vaginal dilators made of dental alginate impression material which is inexpensive and freely available.
- ▶ Prevention of adhesion should be considered especially for those with excessive vaginal denudation.
- ▶ The general population should be educated about the dangers of procuring illegal abortions and healthcare workers should be as careful about postoperative instrument and swab count after a vaginal procedure as after a laparotomy.

Prevention of vaginal stenosis, though not routinely practiced, is recommended as a preventive measure in patients who have excessive vaginal mucosal denudation. Any type of vaginal dilator or vibrator can be used, none having any benefit over another. In these cases, frequent vaginal intercourse may also be helpful.¹⁵

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