Case report

Secondary pyosalpinx after reconstructive surgery of vaginal agenesis patient with bilateral hematosalpinx: A case report

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ABSTRACT

Introduction and importance: Congenital or hypoplasia vaginal agenesis is a very rare condition caused by the failure of developmental Mullerian ducts. The prevalence is 0.001%–0.025% populations. This condition often misdiagnosed because the symptom does not appear. Acute symptoms such as abdominal pain may occur due to the obstruction of retrograde menstrual flow. In this case, we presented a case complex management of vaginal atresia with pyosalpinx, hematometra and bilateral hematosalpinx.

Presentation of case: A 12 years old teenager, non-sexually active, complained cyclic abdominal pain that worsening in seven months before admission. Patient never had menstrual blood flow during her life. Patient was diagnosed with hematometra, hematocolpos, bilateral hematosalpinx and distal vaginal agenesis. Amnion graft neovagina was performed. Five days after surgery, patient started to have fever. On the seventh days after surgery, amnion graft was removed. The next two days patient still had fever. Because of continuous fever, patient was test of COVID 19. The result was positive. On the eleventh days after the first surgery, patient complained abdominal pain VAS 3–4. Patient was diagnosed with pyosalpinx by ultrasound examination. Laparotomy was done performing adhesiolysis, bilateral salpingectomy, and omentectomy.

Discussion: In our case vaginal reconstruction surgery from vaginal approach has been done without management of the bilateral hematosalpinx because the consideration of small caliber of bilateral hematosalpinx. But then complications were developed when vaginal canal was opened, bilateral hematosalpinx were transformed into bilateral pyosalpinx and continue to developed into bilateral tubal abscess. We assume during this process, the bacteria from vagina could fastly infecting the blood and transformed it into pus and grew until tubal abscesses. We assume during this process, the bacteria from vagina could fastly infecting the blood and transformed it into pus and grew until tubal abscesses. We assume during this process, the bacteria from vagina could fastly infecting the blood and transformed it into pus and grew until tubal abscesses. We assume during this process, the bacteria from vagina could fastly infecting the blood and transformed it into pus and grew until tubal abscesses.

Conclusion: The surgical intervention in vaginal agenesis must be considered as a treatment and not only focus on the reconstruction. Laparoscopy or laparotomy may offered as options for combination treatment with vaginal approach reconstructive surgery for vaginal agenesis with obstruction complications such as hematometra and hematosalpinx to prevent the worst condition like ascending infection or misdiagnosed other severe conditions.

1. Introduction and importance

Congenital vaginal agenesis is a condition caused by unsuccessful development of Mullerian ducts. This condition is very rare and occurred in 0.001%–0.025% populations [1]. This abnormality has some forms such as transverse vaginal septum, duplicated vagina, distal vaginal atresia and vaginal agenesis. According to AFS classification by Rock and Adam, uterovaginal anomalies have subdivision categories. Hypoplasia or agenesis vagina is Class I category. If the condition is not treated, it will become further conditions like hematometra and hematosalpinx due to retrograde hematometra into salpinx. In this case report, we presented a 12-years old puberty female with cyclic abdominal pain and primary amenorrhea. The patient has distal vaginal atresia condition with pyosalpinx, hematometra, and bilateral hematosalpinx.

This case report has been reported in line with the SCARE 2020 criteria [2].

2. Patient information

A 12 years old teenager, non-sexually active with a chief complaint of cyclic abdominal pain at the beginning of the month and became
worsened in seven months before admission. The breast and pubic hair started to grow at 9 years old. The patient has never had menstrual blood flow during her life. There is no disturbance of micturition or defecation. There was no family history of the same disease. Patient is elementary school student. She has no history of drug consumption.

3. Clinical finding

From physical examination, vital signs were unremarkable. Tanner stage M4P3. Nutritional status was grade I obesity. From abdominal examination, there was a palpable mass until 3 fingers below the umbilical with tenderness. Gynecology examination, revealed no vaginal opening nor hymenal bulging (Fig. 1) In the rectal toucher bimanual examination, there was a palpable mass located starting 4 cm proximal from the anal opening sized 11 × 7 cm bulging from anterior area, suspected hematocolpos bulging into rectal canal. The mass was integrated with the previous mass palpated abdominally until 3 fingers above navel, with total mass sized 14 × 10 cm. The vaginal structure at anterior area was palpable with tenderness.

4. Diagnostic assessment

From ultrasound examination, there was enlarged uterus, uterine cavity dilated filled with fluid (hematometra) until cervical canal sized 30 × 31 mm. There was hematocolpos with size 112 × 68 mm. There were also bilateral hematosalpinx with size 56 × 10 mm (right) and 58 × 11 mm (left) (Fig. 2).

5. Therapeutic intervention

Amnion graft neovagina was performed as definitive treatment for the patient (Fig. 3). Experienced urogynecologist performed the procedure. Five days after surgery, the patient started to complain about fever. The amnion Graft with silicone was removed in the 7th days after surgery. From the gynecology examination in the 9th days after surgery, we found fluxus. In the ultrasound examination, there was bilateral hematosalpinx. In the 11th days after surgery, patient still had fever and complained about abdominal pain VAS 3–4. The Covid 19 test was positive. There was pyosalpinx in the ultrasound examination. Laparotomy approach was chosen compare to laparoscopy due to consideration for the quickest way in performing surgery for Covid-19 positive case. The patient was performed adhesiolyis, due to bilateral tubal abscess with pus found 200 cm³, bilateral salpingectomy, and partial omentectomy was performed continue with washing the intraabdominal cavity and drain insertion (Fig. 4). Empiric antibiotic was given with the goal free of fever for at least 2 × 24 h.

6. Follow up and outcome

Postoperative period was uneventful and patient was discharge in satisfactory condition. Patient was counselled on the possibility of recurrence. She is being followed up and to date (1 month post operative), there has been no sign of recurrence and no other disability. She performs molding regularly three times a day for 20 min.

7. Discussion

Congenital vaginal agenesis also called Mullerian agenesis or Mullerian aplasia is an anomaly of female reproductive tract that was first described by Action of Amanda [1]. Vaginal agenesis often has relation with congenital absence of uterus in 90–95% of cases [3]. This failure condition may have some forms such as agenesis, vertical or horizontal vaginal septum. Vaginal septum can be located everywhere, usually located at the upper and medial vaginal junction [4]. The symptoms of vaginal agenesis are underreported from the patient until the patient sexually active.

The symptoms are the inability of coitus, primary amenorrhea, and infertility. Vaginal agenesis patient may also complain about some symptoms that disturb the quality of life. The symptoms are cyclical lower abdominal pain. This pain is always felt at the relative same date every month. This pain is representation of menstrual period in patient which is obstructed and collected. Even though patients have no vagina, some patients still have normal uterus (like patient in this case) [5]. The menstrual blood causes cyclical pain and becoming worsened due to more amount blood obstructed every month.

Menstrual blood will collected inside vagina causing vaginal dilatation until the cervix, dilatation of uterine cavity and also making backflow filling both tubes and come out into abdominal cavity. This retrograde menstruation causes pelvic or membrane adhesion and gradual straightening from ostium to tuba until it makes tubal obstruction and hematosalpinx happen [5]. The capacity of menstrual blood collection may differ in every patient. A study showed the maximum quantity is 3000 ml [5]. The genital dilatation also makes mass sensation that can be palpated in the nearest umbilical [4].

Retrograde menstruation increases risk of endometriosis, pelvic adhesions, infertility and pelvic inflammatory disease (PID) [4]. Collected menstrual blood is a source for microorganism growth that will increase risk of infection [6]. PID is uncommon in non-sexually active females like in this case. The spectrum of PID is covering from upper female genital tracts disease such as endometritis, salpingitis, tubal ovarian abscess and pelvic peritonitis [7]. This goes along with this case that backflow of menstrual blood causing obstruction and increasing the risk of infection, specifically pyosalpinx and tubal ovarian abscess (TOA). TOA is an advanced form of pyosalpinx. There is no literature specifically discuss pyosalpinx pathogen in the non-sexually active patient. The study from Maraqa, a patient with pyosalpinx secondary due to agenesis ductus Mullerian, showed evidence of infection from peptostreptococcus anaerobic, prevotula biva, Streptococcus anginosus [7]. In our case, the bacterium found was Klebsiella oxytox.

Risk factors of PID that will continue to make salpingitis conditions are obesity, diabetes, poor hygiene, bacterial vaginosis, a history of pelvic surgery, sexual abuse, multiple partners, appendicitis, and the history of PID. Urogenital malformation was also reported to have a

![Fig. 1. No vaginal opening.](image-url)
Fig. 2. (A) Hematosalpinx (B) hematometra and hematocolpos in first ultrasound examination.

Fig. 3. Silicone catheter covered with amniotic membrane.

Fig. 4. Omentectomy and salpingectomy.
relation with pyosalpinx. Obesity as in the patient has a contribution to infection formation because it will make patient physically hard to clean and do personal hygiene [7].

Pyosalpinx and hematosalpinx are difficult to differentiate from imaging. Both are fluid inside fallopian tubes. In the ultrasound examination, pyosalpinx shows as tubular adnexal dilatation with complex fluid, the wall became hyperemic and thick, meanwhile hematosalpinx show tubular adnexal dilatation contained with fine low level of echoic. Ultrasound has to be done by an expert person [8].

There are some modalities to diagnose Mullerian anomaly. Laparoscopy is costly and often inconclusive because it approached from abdominal. Ultrasound and MRI are very helpful to diagnose. Three-dimensional ultrasound in expert operator has 93% sensitivity and 100% specificity to diagnose Mullerian anomaly. MRI is a gold standard that gives best estimation until 100%. MRI is not operator depended and not limited by the bowel loop disturbance even though the cost is expensive. Nevertheless, the most precise diagnosis is macroscopic appearance from intraoperative [2]. In our case, MRI is not performed because of its highly cost.

The treatment of pyosalpinx and TOA are conservative treatment with antibiotics, laparoscopy aspiration, aspiration or drainage with guided, laparoscopy salpingostomy with saline irrigation, or salpingectomy. If there is gangrene or severe tubal damage, surgery is the choice [6]. In this case, the first surgery is neovaginal amnion graft reconstruction without management for hematosalpinx because the hematosalpinx size was small, around diameter of 1.5 cm from the ultrasound imaging.

Mullerian anomaly with complications can be treated differently. This is adjusted by the condition and preference of the patient, for instance laparoscopy or laparotomy are the choices [9].

In our case vaginal reconstruction surgery from vaginal approach has been done without management of the bilateral hematosalpinx because the consideration of small caliber of bilateral hematosalpinx. But then complications were developed when vaginal canal was opened, bilateral hematosalpinx were transformed into bilateral pyosalpinx and continue to develop into bilateral tubal abscess. This was thought happened because of ascending bacteria from vagina which infecting the bilateral hematosalpinx that cannot be released once the obstructed vagina opened below. Hematocolpos and hematometra were spontaneously released during opening the vaginal obstruction when we suctioned obstructed blood from below. But obstruction of bilateral hematosalpinx was left untreated because we could not suction or release the blood inside those tubes once we open the vagina. It remained stagnant inside both tubes waiting to be absorbed spontaneously which need time several days until it totally absorbed. We assume during this process, the bacteria from vagina could fastly infecting the blood and transformed it into pus and grew until tubal abscess. Because of that, now we recommend concomitant laparoscopy or laparotomy surgery to be performed in Mullerian anomaly patients with complicated conditions such as hematometra or hematosalpinx, especially bilateral. We could drainage those from abdominal approach and would decrease risk of the obstructed menstrual blood becoming infected. And with abdominal approach we also could have better diagnostic and decrease the risk of undetected pyosalpinx, so it can be treated earlier to prevent developing focal infection that would make problems later on.

8. Conclusion

Hematosalpinx increases the risk of developing into pyosalpinx and tubal abscess. Therefore, laparoscopy or laparotomy is supposed to be done in a Mullerian duct anomaly patient with retrograde menstrual complicated conditions such as hematometra, hematosalpinx, and maybe endometriosis to prevent development of any worse condition.

The treatment goal must be looked from holistic approach and not focus only on reconstruction procedure.

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Declaration of competing interest
None declared.

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