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Pregnancy in a Woman with Untreated Vesico-Cloacal Extrophy: A Clinical Case

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Abstract

Introduction : Congenital malformations in a woman carrying a pregnancy are extremely rare. **Report case :** We report a case of a 16-year-old polymalformed woman with a pregnancy, referred from a rural health facility without a technical platform and qualified human resources (Batangafo District Hospital) to a facility with limited resources (Sino-Central African Friendship University Hospital Center in Bangui, the capital of the Central African Republic). The patient was successfully surgically managed: a caesarean section was performed, resulting in the cephalic extraction of a live newborn of sex female weighing 2,950 g with an Apgar score of 10/10 at the 1st, 5th and 10th minutes of life and the reconstructive surgery for various malformations. The post-operative period was simple and the patient was discharged on day 15. **Conclusion :** This case of multiple malformations during pregnancy remains an exceptional situation and

highlights the need for prenatal monitoring, ultrasound evaluation, and multidisciplinary care.

Keywords: Polymalformed ; Vesico-cloacal exstrophy ; Batangafo ; Central African Republic

Introduction

Congenital malformations are a major cause of morbidity and mortality (Sabiri N et al., 2013). Their causes are multiple; they can be due to endogenous anomalies (genetic or chromosomal) but also result from an extrinsic factor affecting embryogenesis (medications such as neuroleptics, infections, environmental exposure of one or both parents) (Thomas J et al., 2018). Sometimes, the etiology remains unknown. According to a recent study, child deaths due to congenital malformations are too high in sub-Saharan Africa compared to high-income countries (SciDevNet, 2021), and the Central African Republic (CAR) is no exception. We report a case of a minor with multiple malformations (16 years old), carrying a pregnancy in a context of very limited resources, which we managed in a multidisciplinary manner, and then we will discuss it with data from the literature.

Clinical Case

It was about a patient X, 16 years old, unemployed, living in Batangafo in the CAR. She was referred from the Batangafo District Hospital to the Sino-Central African Friendship University Hospital on 04/28/2024 for:

- Absence of closure of the left abdominal wall (laparochisis) and a congenital umbilical hernia;
- Complete absence of the labia majora (total aplasia of the labia majora);
- Absence of the vagina except for a small opening providing access to the vagina (partial congenital vaginal agenesis);
- Absence of closure of the bladder (bladder exstrophy);
- Malposition of the urethral meatus in front of the clitoris (epispadias);
- Bifid clitoris.

All based on a notion of amenorrhea of 8 months and 2 weeks according to the patient.

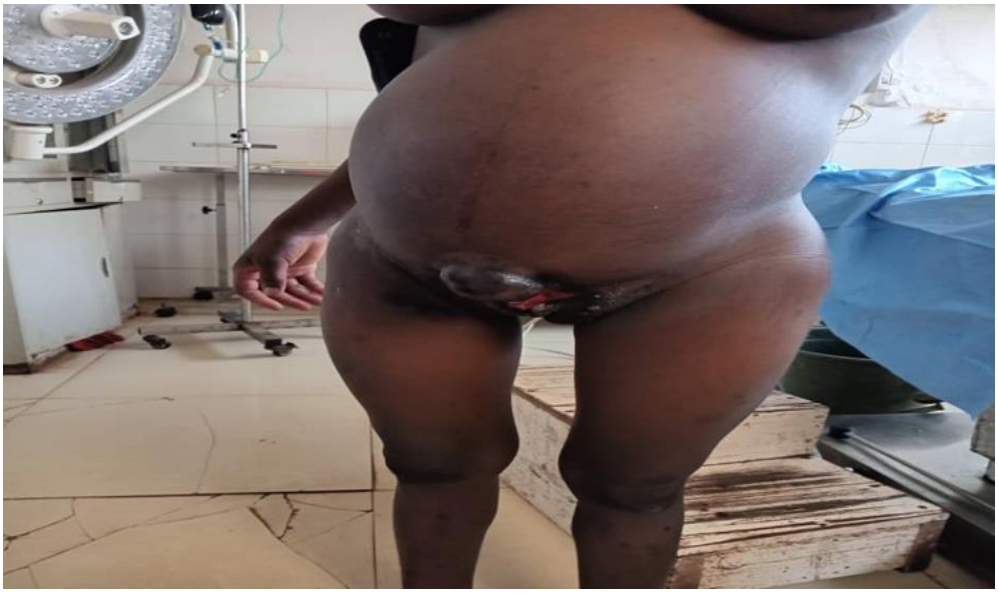

Figure 1 : Polymalformed patient carrying a pregnancy (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

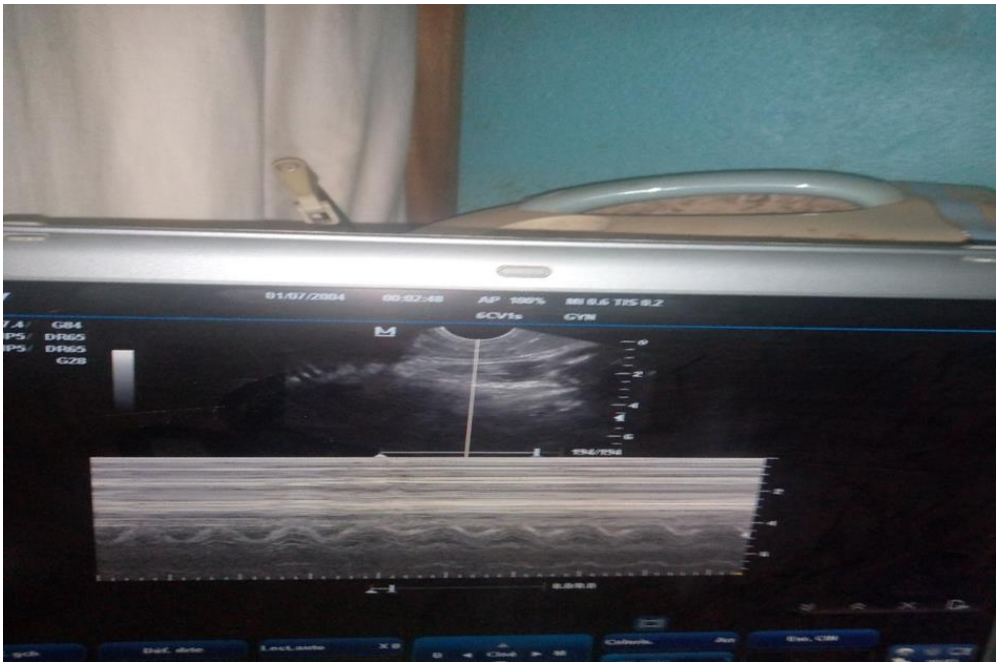

Figure 2 : Ultrasound showing the fetal heart rate (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

Family history: The patient is the 4th child in a sibling group of 7, all of whom appear to be in good health. No known hereditary defects have been reported in the family, either on the paternal or maternal side.

Personal history :

- Age at menarche: 13 years ;
- Date of last menstrual period unknown ;
- Menstrual cycle: irregular (31 days for 5 months then 28 days) ;
- Primigravida ;
- Nulliparous ;
- No history of contraceptive use ;
- Hospitalized at 7 years old for paraplegia for one week

The **CLINICAL EXAMINATION** performed on admission on 04/28/2024 finds:

- A good state of consciousness with a Glasgow score of 15/15 ;
- Good skin and mucous membrane coloration ;
- Good hemodynamic condition ;
- Weight = 55 kg, Height = 1.62 m => Body Mass Index (BMI) = 20.9 kg/m² ;
- An abdomen increased in volume, with a long longitudinal axis, breathing well ;
- Active fetal movements well perceived by the mother ;
- Uterine height at 33 cm ;
- Mobile cephalic presentation ;
- A plan of the back on the right ;
- An umbilical bulge allowing palpation of certain abdominal organs ;
- A pubic anomaly with absence of the pubic symphysis bones ;
- Absence of the labia majora ;
- The presence of a protruding suprapubic lesion with visible ureters allowing urine to flow ;
- A bifid clitoris ;
- Malposition of the urethral meatus in front of the clitoris (epispadias) and partial congenital absence of the vagina with a small vaginal opening providing access to the cervix ;
- Fetal heart sounds at 139 beats per minute in the lower right quadrant ;
- Vaginal examination impossible as the vagina is almost nonexistent except for a small opening that barely admits the little finger ;
- Rectal examination unremarkable.

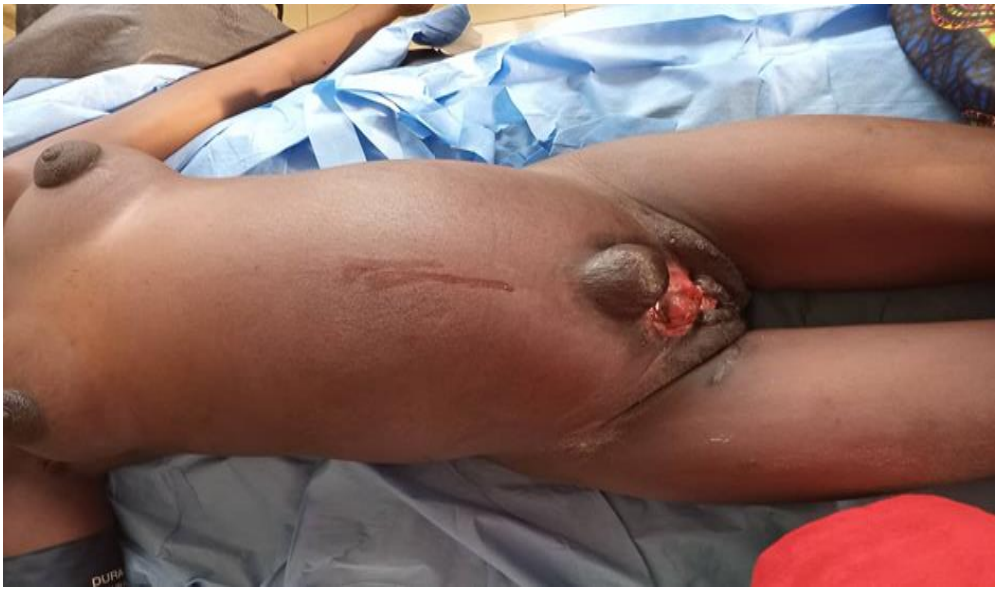

Figure 3 : Patient lying on the operating table with malformations (umbilical hernia, cloacal bladder exstrophy) (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

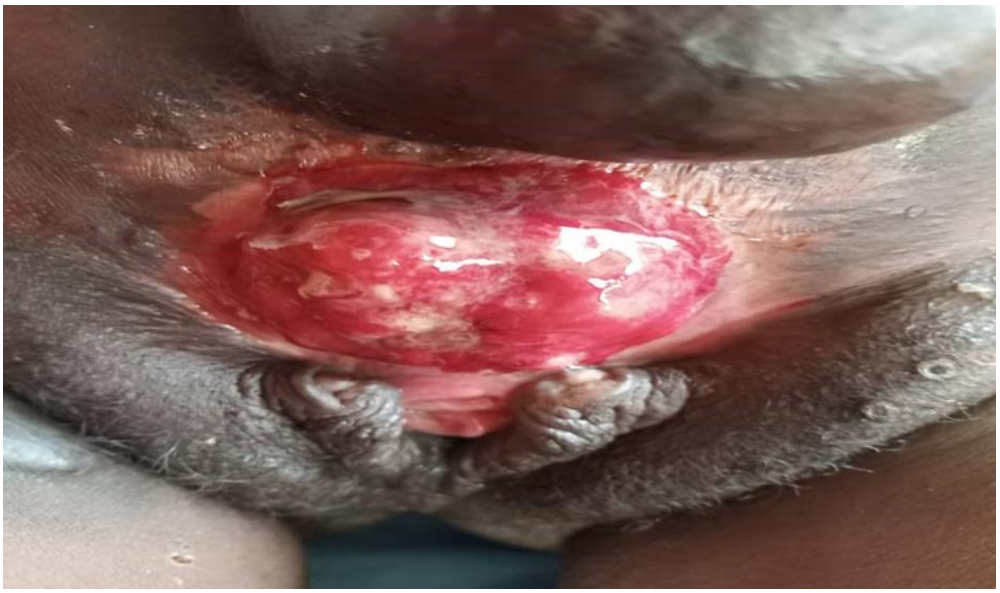

Figure 4 : Uro-digestive and genital malformations (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

The following **paraclinical examinations** were performed:

- Blood group: A / Rhesus: negative ;
- Hemoglobin level = 10.6 g/dl ;
- C-reactive protein = 4 mg/L ;

- HIV 1 and 2 serology : negative ;
- Syphilis serology : negative ;
- Urine dipstick : Nitrites = positive => Cytobacteriological examination of urine (CBEU): urinary tract infection with Staphylococcus aureus ;
- Obstetric ultrasound: Single intrauterine pregnancy at 36 weeks of amenorrhea, cephalic presentation. Anterior placenta, Grade III. Amniotic fluid normal (Amniotic Fluid Index = 4.3 => Largest Pocket). Estimated Fetal Weight = 2,700 g.
- Frontal pelvic X-ray: widening of the pubic symphysis bones less than 0.5 cm.

In total : picture of a polymalformative syndrome complicated by a urinary tract infection with Staphylococcus aureus associated with anemia in a pregnancy of 36 weeks of amenorrhea. The patient was immediately hospitalized in the department under strict maternal-fetal monitoring with antibiotic treatment and iron supplementation. After three days of treatment, the indication for a prophylactic cesarean section was made because vaginal delivery was practically impossible due to multiple malformations and pelvic diastasis. This cesarean section was performed and allowed the cephalic extraction of a live female newborn weighing 2,950 g with an Apgar score of 10/10 at the 1st, 5th, and 10th minutes of life.

Figure 5 : Painting of the patient (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

Figure 6 : Extraction of the newborn (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

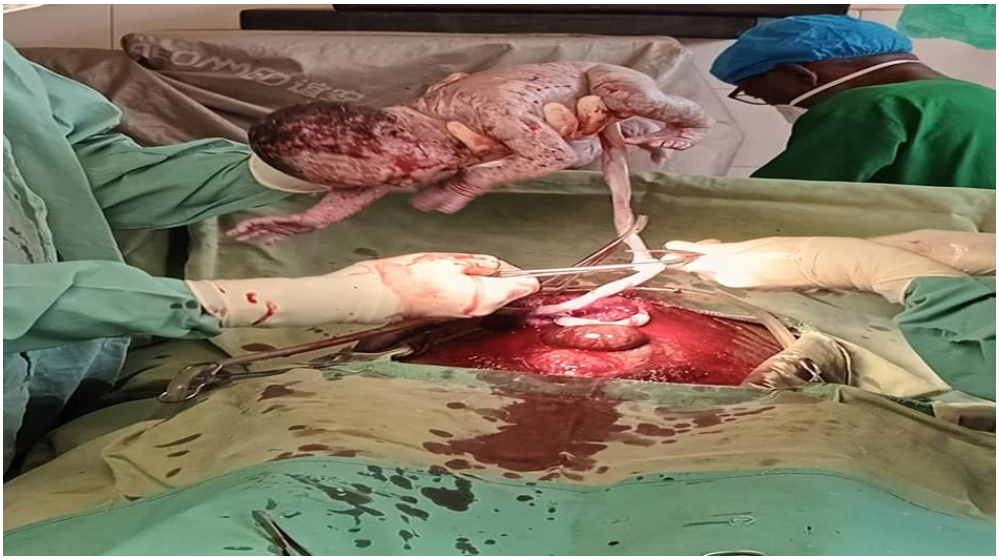

Figure 7 : Clamping then cutting of the umbilical cord (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

The procedure continued with the repair of the laparochisis and the reconstruction of the anterior surface of the bladder. For this, the nearby abdominal parietal peritoneum was harvested to close it. A capacity of about 30 ml was obtained; an indwelling urinary catheter was then placed, all of which was covered by the aponeurosis. The closure was done with separate stitches.

The procedure continued with the opening of the small opening that constitutes the vagina by enlarging the orifice to create a neo-vagina. A depth of 5 cm communicating with the urinary catheter in place was obtained for the evacuation of urine and any potential sexual intercourse.

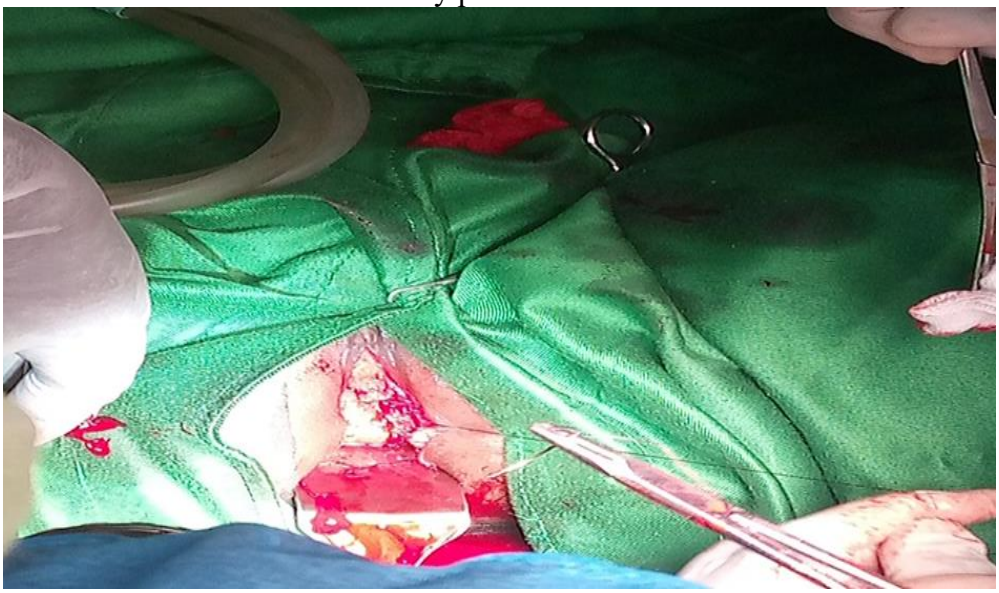

Figure 8 : Repair of laparochisis and the anterior surface of the bladder (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

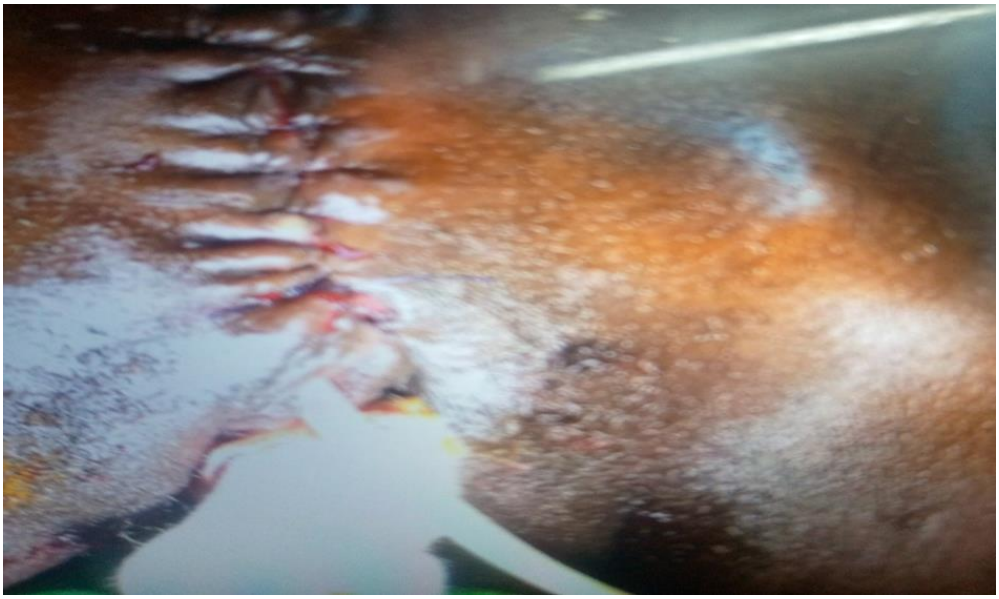

Figure 9 : Skin closure (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

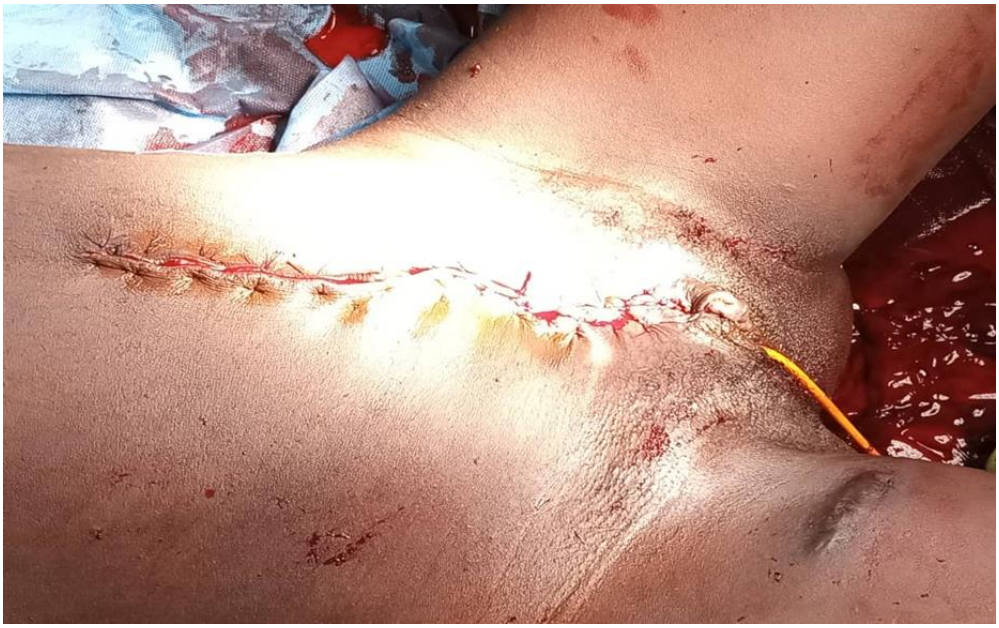

Figure 10 : Appearance of the abdominopelvic wall after repair with the presence in yellow of an indwelling catheter (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

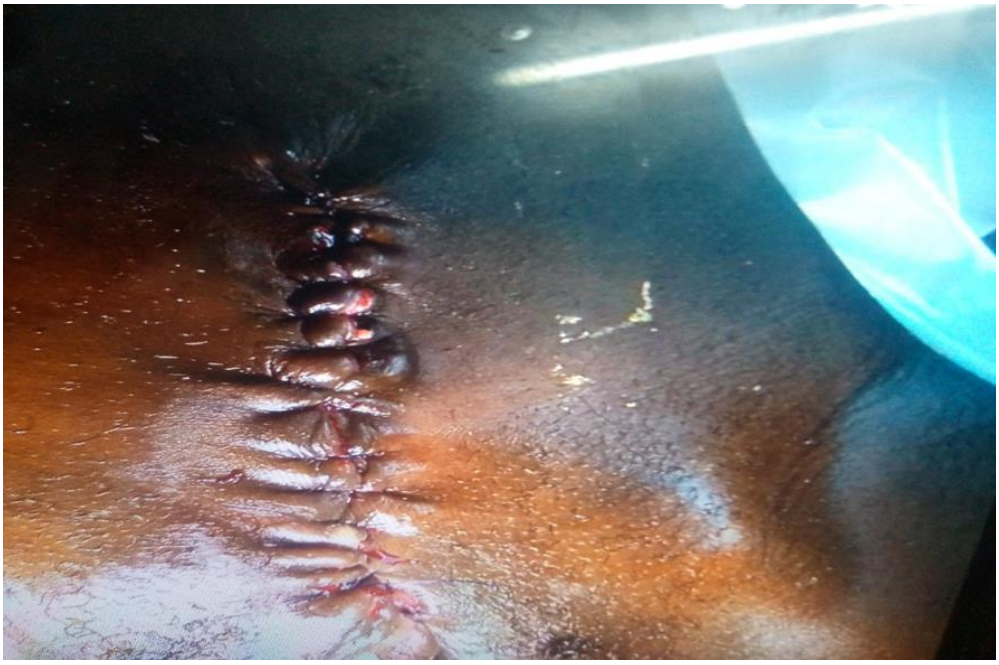

Figure 11 : Appearance of the abdominopelvic wall after repair without the indwelling catheter (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

The total duration of the procedure was 2 hours and 15 minutes. No anesthetic or surgical incidents were reported. Blood loss was estimated at 400 ml, uncompensated. The patient was transferred to intensive care and then to the ward for post-operative monitoring with the following treatment: injectable Ampicillin 2 g every 8 hours, injectable Metronidazole 500 mg every 8 hours, injectable Gentalline 160 g every 24 hours, injectable Perfalgan 1 g every 6 hours, and injectable Diclofenac 75 mg every 12 hours for 4 days, then switched to oral. An anti-D serum was systematically administered to the patient. Vital signs were normal; urine output was 1800 ml the next day. Bowel function resumed 24 hours later. Perineal rehabilitation was initiated to allow urinary evacuation. Good wound healing was achieved after 15 days. Therefore, the post-operative course was uneventful.

Figure 12 : Appearance of the abdominal wall after healing (Photo: Dr. Roch M'BETID-DEGANA and Dr. Gilles-Davy KOSSA-KO-OUAKOUA)

The patient was monitored every 15 days for 3 months, then monthly for 3 months. At the end of her six months of follow-up, the patient was discharged. Oral contraception was established after the patient's consent for at least two years, with an absolute contraindication to vaginal delivery during the next pregnancy.

Discussion

1. Age

Our patient was 16 years old. However, there are not enough scientific publications specifically discussing the age of procreation in a woman with multiple congenital malformations, due to the fact that this

situation is exceptional and very diverse. Yet, these multiple congenital malformations are often responsible for infertility and complications both obstetrical and urinary (Amaral PP and al., 2022). We found no cases in the literature of a minor with multiple malformations carrying a pregnancy. Nevertheless, age influences procreation and remains a major factor of fertility regardless of the malformations. The maximum reproductive age is between 20 and 35 years (general concept in Medically Assisted Procreation) (Gaille M, 2019). Fertility is often reduced in a woman with multiple malformations, so the use of Medically Assisted Procreation is often considered in developed countries as an alternative.

2. Mode of delivery

There is almost no scientific article specifically addressing the mode of delivery in a patient with multiple malformations. The mode of delivery is not standardized in a patient with multiple malformations. However, the obstetric literature allows us to establish solid scientific principles based on comparable situations. Thus, our patient underwent a prophylactic cesarean section at eight and a half months followed by systematic reconstructive surgery. Our approach is justified by the combination of these multiple malformations (pubic diastasis, narrow vaginal opening) preventing the passage of the fetal mass through the birth canal in an immature pelvis (Schmitz T, 2012 ; Ben-Guigui J and al., 2018). This approach is consistent with the data from the scientific literature (Merger R, 2001).

3. Types of malformations

Our patient presents with:

- The Bladder/Clocal Exstrophy Complex (BCEC): BCEC encompasses a set of congenital urological anomalies affecting the bladder, urethra, genital organs, and the pelvic musculoskeletal system (Fendereski K and al., 2024). It can impact sexual health and fertility (Bujons A and al., 2016). Its diagnosis was delayed in our patient. This situation is not exceptional in Africa (Tshimbayi M and al., 2014). Normally, BCEC is diagnosed during the first trimester of pregnancy. However, in most developing countries, it is detected at birth, as in the case of our patient. This diagnosis still poses significant challenges in rural areas, and management is complicated. This could explain why our patient was not treated for sixteen years. Its association with pregnancy is very rare but not impossible (Surer I and al., 2001).
- Malformations of the genital apparatus such as urethral meatal atresia, bifid clitoris, congenital atresia of the labia majora, and partial congenital agenesis of the vagina associated with pelvic

diastasis: Spontaneous pregnancy may be possible even without reconstructive surgery, as in the case of our patient. However, a case of congenital vaginal agenesis followed by pregnancy has been described in the literature after vaginoplasty (Aimen FJ and al., 2016).

4. Prognosis after reconstructive surgery

Our patient was placed on oral contraception after her consent. However, pregnancy after CEVC repair is not rare (Kalansuriya D and al., 2025). Pregnancy in patients operated on for cloacal exstrophy can present a high risk with obstetric complications (miscarriage, premature birth, premature placental detachment, ...) and urinary complications (urinary infection, urinary obstruction, hydronephrosis, ureteral stricture, and ureteral stones (Muecke EC, 1964; Deans R and al., 2012) throughout the pregnancy, which highlights the importance of multidisciplinary management during a subsequent pregnancy. Furthermore, the creation of a neovagina for this patient during the procedure will allow not only the restoration of fertility but also the resumption of a "normal" sexual life.

Conclusion

Very rare, the case illustrates a form of vesicocloacal exstrophy and genital malformation diagnosed late in an untreated adolescent during an advanced pregnancy discovered by chance. This type of pathology requires proper multidisciplinary management by an experienced team.

Conflict of Interest: The authors reported no conflict of interest.

Data Availability: All data are included in the content of the paper.

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