

Complete uterine septum, cervical septum and longitudinal vaginal septum: a challenging differential diagnosis with double cervix

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ABSTRACT

Background: The presence of complete uterine septum, cervical septum and longitudinal vaginal septum (class U2bC1V1 according European Society of Human Reproduction and Embryology/European Society for Gynaecological Endoscopy classification) is a rare congenital anomaly of the female genital tract. The diagnosis of this anomaly is very challenging, significantly influencing the type of treatment to be performed.

Objectives: We propose a one-stop diagnosis through the combined use of 2D-3D ultrasound (US) and hysteroscopy and the minimally invasive endoscopic treatment of this anomaly, emphasising the diagnostic and therapeutic differences compared to U2bC2V1 anomaly.

Participant: Stepwise demonstration with video footage of an integrated approach in the management of a patient with a class U2bC1V1 anomaly. The patient was 23 years old and presented with dyspareunia and a previous miscarriage. We performed a one-stop diagnosis through the combined use of diagnostic hysteroscopy and 2D-3D pelvic US and a minimally invasive endoscopic treatment with a 15Fr bipolar miniresectoscope.

Intervention: Hysteroscopic control performed 40 days after the procedure showed a regular vagina, a normal single cervix and a normal uterine cavity. No intra- or postoperative complications occurred. The patient was discharged 3 hours after the procedure. The total operation time was 24 minutes.

Conclusions: Making an accurate diagnosis of a single cervix with cervical septum and a double cervix is crucial in the management of patients with complex genital anomalies. An accurate diagnosis is possible when combining hysteroscopy and US. Minimally invasive endoscopic treatment of U2bC1V1 anomaly with a 15 Fr bipolar miniresectoscope is an effective and safe procedure, easier when compared to the treatment of U2bC2V1 anomaly.

What is New?: This videoarticle describes the hysteroscopic criteria for the differential diagnosis between single cervix with cervical septum and double cervix.

Keywords: Cervical septum, uterine malformation, ultrasound, hysteroscopy, U2bC1V1

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Introduction

The simultaneous finding of complete uterine septum, cervical septum, and non-obstructive longitudinal vaginal septum is a rare anomaly of the female genital tract classified as U2bC1V1 according to the European Society of Human Reproduction and Embryology/European Society for Gynaecological Endoscopy (ESHRE/ESGE) classification.¹ The real incidence of this complex anomaly cannot be estimated because of its rarity and lack of data in literature.^{2,3} The most frequent symptoms are dysmenorrhea and dyspareunia, which are often associated with adverse obstetric outcomes such as infertility, recurrent miscarriages, preterm deliveries, and intrauterine growth restriction. In a yet undetermined proportion of patients, this condition remains entirely asymptomatic.⁴⁻⁶

The non-specificity of symptoms, its rarity, and the absence of standardised diagnostic techniques often result in misdiagnosis or diagnostic delay.

Diagnosis can be very challenging and may require the use of different techniques and multiple steps. In the past, diagnosis was obtained through a combination of hysteroscopy and laparoscopy. Due to improved diagnostic techniques, currently the most widely used methods are magnetic resonance, 2D-3D pelvic ultrasound (US), and hysteroscopy.⁷

Crucial aspects in diagnosis involves the study of the external uterine profile and of the cervix or cervixes.

The correct assessment of these two parameters allows a differential diagnosis in the first case between septate uterus and bicorporal uterus (U2b vs. U3b); in the second case between the presence of single cervix with cervical septum and double cervix (C1 vs. C2).

While the enormous progress made in 3D US allows for a remarkably accurate investigation of the external uterine profile, the cervix evaluation appears, to date, still controversial and is the main diagnostic challenge in the evaluation of these complex anomalies.⁸

Evaluation of the cervix is particularly complex in patients with intact hymen and/or vaginal congenital anomalies. In these cases, the speculum examination may not be feasible or may be hindered by the vaginal anomaly, not allowing complete and accurate visualisation of the cervix or cervixes.

Moreover, even when well visualised, it is not always easy to distinguish between a double cervix and a single

cervix with a cervical septum, due to the lack of clear guidelines providing precise parameters for a differential diagnosis. The techniques that have been found to be most effective in the study of the lower genital tract are vaginoscopy as well as the 3D saline-contrast sonovaginocervicography.^{9,10}

The differential diagnosis is fundamental as it results in substantially divergent surgical treatments. In fact, considering the external uterine profile, many studies have shown that removal of the uterine septum improves obstetric outcomes, while there are no surgical indications in cases of bicorporal uterus.¹¹

More controversial is the treatment of the cervical anomaly. In literature, data are scarce and quite contradictory. While it seems, there is no indication to treat double cervix,^{12,13} some evidence, although limited, showed that the removal of the cervical septum associated with metroplasty makes the procedure safer, easier, and less complicated.¹³⁻¹⁵ Women treated with cervical septum incision have no significant differences in reproductive outcomes compared to patients with preservation of the cervical septum. Moreover, the caesarean section rate is lower after the removal of cervical septum.^{14,16}

The aim of our study is to describe the characteristics to make the differential diagnosis between double cervix and single cervix with cervical septum easier. We describe the key differential aspects between single cervix with cervical septum and double cervix, and we share our one-stop¹⁷ minimally invasive approach for the diagnosis and treatment of this anomaly.

Methods

A 23-year-old woman was referred to our hospital - Fondazione Policlinico Agostino Gemelli IRCCS of Rome, Italy - for a suspicious of complex uterine anomaly. The patient presented with dyspareunia and in her obstetric history she reported a previous spontaneous miscarriage.

The diagnosis was obtained in our Digital Hysteroscopic Clinic - CLASS Hysteroscopy - through a one-stop office procedure with the integration of 2D-3D pelvic US and hysteroscopy both performed at the same time by an experienced operator (U.C.). Through 2D US, in the transverse scan, the presence of a complete uterine septum was observed. At 3D reconstruction, the external uterine outline showed a convex profile and the presence of a complete uterine septum that reached the internal uterine orifice with the evidence of two distinct, non-

communicating uterine hemicavities. The uterine septum appeared to continue into the cervix resulting in the presence of two distinct cervical canals. Ultrasonographic evaluation was performed according to the diagnostic criteria of the 2016 ESHRE/ESGE consensus.⁷

Vaginoscopy showed the presence of two hemivaginas and of a non-obstructive complete longitudinal vaginal septum. From the left hemivagina, we entered the left cervical canal, reaching the left uterine hemicavity visualising the ipsilateral tubal ostium. At this point, exiting from the left cervical canal, we noticed that the cervical septum was not in junction with the vaginal septum. So, from the left hemivagina, we directly entered the right cervical canal, overpassing the vaginal septum. Through the right cervical canal, we reached the right uterine hemicavity visualising the ipsilateral tubal ostium.

The two uterine hemicavities appeared completely separated by the presence of a complete uterine septum that continued with the cervical septum without interruption. It was the first time that we found this scenario and we carefully evaluated the cervix. It was single with a single external uterine orifice. The complete cervical septum originated from the external uterine orifice, it was not in continuity with the vaginal septum and the apex of the cervical septum was covered by endocervical glandular epithelium. The ectocervix was covered by squamous epithelium. No intercervical cleft, covered by squamous epithelium, was observed.

The patient was diagnosed as a class U2bC1V1 anomaly according to the ESHRE/ESGE classification and the endoscopic treatment was scheduled after 30 days of progesterin hormone therapy.

Table 1 shows the diagnostic criteria for single cervix with cervical septum.

Surgical treatment was performed according to an ambulatory model of care,¹⁸ under general anaesthesia with laryngeal mask. The minimally invasive endoscopic

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|---|
| Table 1. Diagnostic criteria for single cervix with cervical septum. |
| Hysteroscopic diagnostic criteria |
| - Single cervix covered by squamous epithelium |
| - No intercervical cleft |
| - Single external uterine orifice |
| - Cervical septum apex covered by endocervical glandular epithelium |
| - Non-continuity between the vaginal septum and the cervical anomaly |

technique was performed with a 15 Fr bipolar miniresectoscope, as follows:

1. Vaginoscopic complete incision of the vaginal septum with a Collins loop.
2. Anterograde incision of the cervical septum and of the complete uterine septum up to the interostial line, using a Collins loop.
3. 2D transabdominal coronal US scan, evaluating the fundal myometrial thickness.
4. Resection of the redundant tissue on the anterior and posterior uterine walls, with a 90° angled bipolar cutting loop.

Results

At the end of the procedure, the total fundal myometrial thickness at 2D-3D US, was 10 mm.

No intra- or post-operative complications occurred. The total surgery time was 24 minutes. The patient was discharged in good clinical condition 3 hours after the procedure.

The hysteroscopic office control performed 40 days after the procedure, showed a regular vagina, a normal single cervix and a normal uterine cavity (class U0C0V0 according ESHRE/ESGE classification). Mild fundal cuts were performed with 5 Fr scissors to optimize the surgical result obtained on the fundus. No intrauterine adhesions were observed. At 3D US, the fundal total myometrial thickness was 9 mm.

Discussion

In this video article we presented our integrated approach in the diagnosis and minimally invasive treatment of patients with vaginal septum, single cervix with cervical septum and complete uterine septum, demonstrating the key aspects in the differential diagnosis with double cervix and analysing the differences in the treatment.

Through the integrated use of 2D-3D transvaginal US and hysteroscopy, we obtained a precise and accurate diagnosis at the same time, avoiding multiple diagnostic steps and unnecessary delays. Furthermore, our combined approach makes magnetic resonance not required for diagnosis. This approach, in the hands of an experienced operator, is effective in diagnosing other complex anomalies of the genital tract, as complete uterine septum, double cervix and vaginal septum.¹⁹

Although the cervix can be clinically assessed by the speculum examination, in many cases the presence of concomitant vaginal malformations can make the precise evaluation impossible. Moreover, it cannot be performed in patients with an intact hymen. It is important to emphasise that, even if well visualized, it is not always easy to distinguish between a single cervix with cervical septum and a double cervix. To the best of our knowledge, Ludwin et al.¹⁶ is the only author who described the diagnostic criteria for cervical anomalies. In his experience the absence of a cleft on the ectocervix is the only diagnostic criterion for distinguishing between these two conditions. Vaginoscopy provides a close view of the cervix allowing an accurate assessment. The presence of a single cervix covered by squamous epithelium, the absence of a cervical cleft, a single external uterine orifice with the cervical septum apex covered by endocervical glandular epithelium, the non-continuity between the vaginal septum and the cervical anomaly, are reliable diagnostic parameters that allowed to accurately diagnose the presence of a single cervix with a cervical septum. The non-continuity between the vaginal septum and the cervical anomaly represents a valuable and easily identifiable landmark for the differential diagnosis; our hypothesis is that this absence of communication directly reflects the alteration in the resorption mechanism that determines the U2bC1V1 anomaly. The absence of this feature could, on the contrary, identify a deficit in the fusion mechanism that determines the U2bC2V1 malformation instead.

Table 2 summarises the hysteroscopic criteria for the differential diagnosis between the two conditions.

The two cervical anomalies differ not only in their pathogenetic mechanism (fusion defect in the case of

double cervix versus a reabsorption defect in the case of single cervix with cervical septum) but also for a different incidence.¹⁶ In fact, in our experience while double cervix is an uncommon anomaly, single cervix with cervical septum is even rarer, although some authors claim the exact opposite. This is clearly due to a lack of standardised diagnostic criteria.¹⁶

The two conditions differ also in the treatment. While there are no surgical indications for treating double cervix, the most recent scientific evidence, although based on limited data and accounting for low sample size, has shown that resection of the cervical septum in case of single cervix, seems to be a safe and effective procedure.¹³⁻¹⁵ In a multicenter randomised controlled trial, 28 patients diagnosed with complete uterine septum and cervical septum, without vaginal septum, who had a history of miscarriages or infertility underwent surgical treatment of these conditions. Patients were randomised into two groups according to receiving or not cervical septum incision. The results showed that incision of uterine septum associated with removal of the cervical septum makes the procedure safer, easier, and less complicated. No significant differences in the reproductive outcomes were found in the two groups. The caesarean section rate was higher in the group with preservation of the cervical septum.¹⁴ Also in our case, the simultaneous incision of the cervical septum and the complete uterine septum makes the procedure simpler, quicker and safer because the step in which the two hemicavities must be connected is avoided.¹⁹ This step corresponds to the most challenging phase of the procedure performed to treat complete uterine septum with double cervix, thus it is possible to create false paths along the myometrium and subsequent uterine perforation.

Regarding surgical technique, while US guidance is essential in the treatment of U2bC2V1 anomaly in order to connect the two uterine haemicavities, in U2bC1V1 patients this guide is not necessary since the incision starts at the vaginal septum apex and continues anterogradely since the uterine fundal interstitial line. The only application of US in this procedure is to assess the post-operative fundal myometrial thickness.

The strength of our technique is the possibility to obtain an accurate diagnosis in a single step by combining hysteroscopy and US. In addition, the use of a 15 Fr miniresectoscope makes our surgical technique safe, effective and minimally invasive, allowing us to discharge the patient 3 hours after the procedure. The limitation of

Table 2. Hysteroscopic criteria for the differential diagnosis between single cervix with cervical septum and double cervix.

| | Single cervix with cervical septum | Double cervix |
|---|---|----------------------|
| Number of external uterine orifice | One | Two |
| Cervical septum apex covered by endocervical glandular epithelium | Yes | No |
| Intercervical cleft | No | Yes |
| Continuity between the vaginal and the cervical septum | No | Yes |

our study is the fact that it is a report of a single case. Further studies are needed to evaluate the reproducibility of our technique and to assess the future obstetric outcomes.

Conclusion

The differential diagnosis between single cervix with cervical septum and double cervix is a crucial moment in the management of patients with complex genital anomalies in order to plan the type of surgical treatment. The combined approach using hysteroscopy and US simultaneously, makes it possible to obtain an accurate diagnosis avoiding diagnostic delays and multiple diagnostic steps.

Minimally invasive endoscopic treatment of a U2bC1V1 anomaly with a 15 Fr bipolar miniresectoscope is an effective and safe procedure, easier if compared to the treatment of U2bC2V1 anomaly, in which US guidance plays a fundamental role. Further studies are needed to evaluate the obstetric outcomes of these patients and to standardise the proposed technique.

Ethics

Informed Consent: Signed informed consent allowing the use of personal data was given by the patient.

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Footnotes

Authorship Contributions

Surgical and Medical Practices: U.C., Concept: U.C., G.S., G.G., Design: U.C., F.B., Data Collection or Processing: E.L.F., E.B., C.F., Analysis or Interpretation: U.C., F.B., G.G., Literature Search: E.L.F., E.B., C.F., Writing: U.C., F.B.

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Video 1. <https://vimeo.com/1007985390/c15bc447e0?share=copy>
