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Differentiating between a double cervix or cervical duplication and a complete septate uterus with longitudinal vaginal septum

To the Editor:

It was interesting to read the report of another rare case of a patient with a septate uterus with a double cervix and longitudinal vaginal septum presented by Celik and Mulayim [1].

The authors refer to other previous reports of exceptionally rare cases related to the defect, which is beyond the classification of congenital defects of the uterus commonly used by the American Society for Reproductive Medicine (ASRM) [2]. The first report of the simultaneous occurrence of a double cervix and a septate uterus [3] undermined the earlier concepts of embryogenesis [4].

This report, like other earlier reports on new cases or even a series of cases, raises our interest and encourages discussion on the accuracy of the definition and the naming of the so-called double cervix, and on its proper classification.

It is essential to distinguish between a double cervix and the more well-known septum of the cervical canal. The latter is defined as Class VA by the ASRM and is characterized by a complete uterine septum that very often is accompanied by a longitudinal vaginal septum.

By contrast, a double cervix is defined by the presence of two separate ectocervixes (portio vaginalis), which are separated by an intercervical cleft between the cervixes. This is analogous to the presence of an intercornual cleft in a bicornuate uterus [2,5]. The visualization of a double cervix may be hindered by the presence of a vaginal septum. However, it is easy to identify after the complete removal of the vaginal septum during surgery (Fig. 1). A typical double cervix is characteristic for Class III by ASRM, which refers to uterus dydelphys [2].

Two separate ectocervixes may indicate a fusion disorder, and not a resorption disorder, related to the septum during embryonic development. Such cases should be described as a double cervix or a duplicated cervix (Figs. 1B and 2C).

Unfortunately, the authors did not provide a photograph of the double cervix, preferably prior to and after the procedure. This photograph would have been particularly valuable, especially because the defect is so rare. The description of the cervical construction examined by endoscopy is not detailed, and unfortunately, the nuclear magnetic resonance (NMR) image only shows the upper part of the uterus, without the double cervix.

However, although the result of the dye test conducted during laparoscopy is described in detail, it is also typical of a complete vaginal septum, defined as Class VA by ASRM. As such, the result of the dye test is not a differentiating element and this should be emphasized to doctors who rarely encounter congenital uterine malformations. It should also be noted that the presence of two cervical canals in NMR or other imaging tests, such as hysterosalpingography or three-dimensional ultrasound, does not differentiate between a double uterine cervix and the partition of the cervical canal.

We routinely use advanced three-dimensional ultrasound diagnostics and specialize in the treatment of congenital uterine defects in our daily practice. Each year, multiple cases of subseptate uterus are diagnosed and approximately five to 12 patients with a complete septate uterus accompanying longitudinal vaginal septum are referred to us from all over the country. Although we regard the diagnosis and treatment of such cases to be relatively simple, they may be regarded as unusual by general gynecologists, who may be encountering such defects for the first time in their practice. All our patients with adverse obstetric history or infertility are treated with hysteroscopic metroplasty, often under intraoperative three-dimensional transrectal ultrasonography. In this procedure, the septum of the cervical canal is not preserved, but is removed. Our unpublished data so far indicate that the procedure does not increase the risk of cervical incompetence.

In our experience, in many cases of a complete uterine septum with vaginal septum, only the cervix and its outer opening are visible during a physical examination with the application of a vaginal speculum into one part of the vagina. The same picture is obtained on the opposite side of the vagina because the vaginal septum is stretched by the speculum. When the speculum is applied only to one part of the vagina, it is shifted either to the right or to the left, giving the impression that it is the vaginal part of the ectocervix. Therefore, it should be stressed that in many cases, the result of a vaginal speculum examination may mimic the presence of a double cervix. As such, it is advisable to use a double-handle speculum (two sets), rather than a single Cusco speculum. If, prior to the removal of the vaginal septum, the recess between the cervix and the vaginal partition is not visible, a diagnosis of a double

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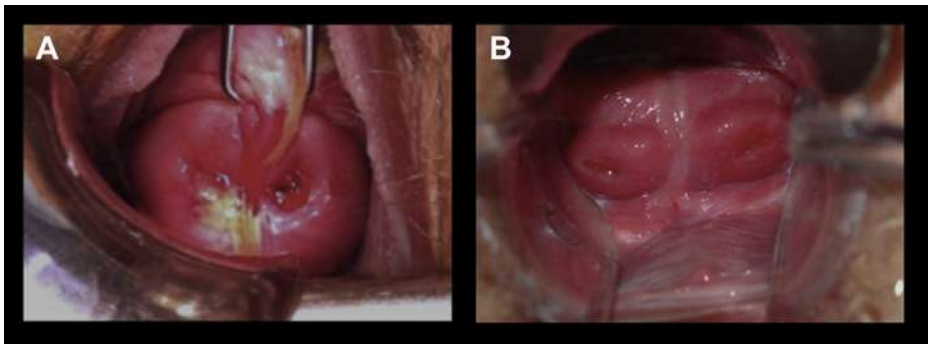

Fig. 1. (A) A single septate cervix and (B) double cervix with intercervical cleft and two separate ectocervixes is easy to identify after the complete removal of the vaginal septum.

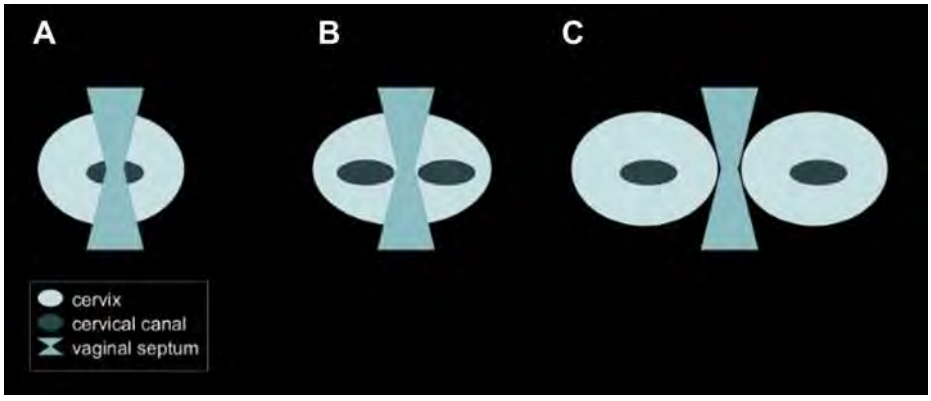

Fig. 2. Vaginal speculum examinations. (A and B) A single septate cervix with a longitudinal vaginal septum is a common form of complete septate uterus (Class VA by ASRM), whereas (C) a double (duplicated) cervix is very rare and connected with a septate uterus.

cervix should not yet be made. After the complete removal of the vaginal septum, two ectocervixes completely separated by an intercervical cleft should be properly visible if it is indeed a double cervix. If an intercervical cleft is absent, the case should be categorized as the well-known Class VA by ASRM, corresponding to a single cervix and septum of the cervical canal, or a septate cervix (Fig. 1A). These are evident as the two more or less distant external ostia on a single ectocervix (Fig. 2A and B). The distance between the external ostia, or the width of the lower part of the septum in the cervical canal, is not important, nor is it an element that differentiates a double cervix from a septate cervix. In a typical image of a complete uterine septum, the cervical septum is wider in its lower part (corresponding to the outer opening), tapers toward the inner opening, and reextends toward the upper part of the uterine cavity.

Unfortunately, some reports on new cases of patients with a complete septate uterus with double cervix lack the appropriate records and sufficiently detailed descriptions of the findings. Moreover, reports often relate to a series of cases of duplicated cervixes, which are in fact simply typical examples of complete uterine septum, defined by ASRM, and accompanied by longitudinal vaginal septum [6].

Therefore, we believe that the actual number of cases of properly verified double cervixes is unknown, and the incidence in the population is even more difficult to estimate. A detailed review and critical analysis of the cases of a double cervix that have been reported so far are needed. Attention should be paid to relevant records; in particular, photographs of the double cervix. These photographs are easy to take, especially during or after the procedure, if the vaginal septum has been removed.

A detailed distinction will enable us to differentiate between a complete septate uterus, which is categorized as Class VA by ASRM and is a typical, yet rarely observed condition in gynecologic practice, and a double cervix with a simultaneous complete vaginal septum, which is a more unique observation.

References

[1] Celik NY, Mulayim B. A mullerian anomaly "without classification": septate uterus with double cervix and longitudinal vaginal septum. Taiwan J Obstet Gynecol 2012;51:649–50.

[2] Buttram Jr VC, Gomel V, Siegler A, DeCherney A, Gibbons W, March C. The American Fertility Society classifications of adnexal adhesions, distal tubal occlusion, tubal occlusion secondary to tubal ligation, tubal pregnancies, Mullerian anomalies and intrauterine adhesions. Fertil Steril 1988;49:944–55.

[3] McBean J, Brumsted JR. Septate uterus with cervical duplication: a rare malformation. Fertil Steril 1994;62:415–7.

[4] Crosby WM, Hill EC. Embriology of the mullerian system: Review of present day theory. Obstet Gynecol 1962;20:507–15.

[5] Ludwin A, Pityński K, Ludwin I, Banas T, Knafel A. Two- and three-dimensional ultrasonography and sonohysterography versus hysteroscopy with laparoscopy in the differential diagnosis of septate,

bicornuate, and arcuate uteri. J Minim Invasive Gynecol 2013;20:90–9.
[6] Chen SQ, Deng N, Jiang HY, Li JB, Lu S, Yao SZ. Management and reproductive outcome of complete septate uterus with duplicated cervix and vaginal septum: review of 21 cases. Arch Gynecol Obstet 2013;287:709–14.

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