

MEDICAL SCIENCE

To Cite:

Goyal N, Agrawal M, Dewani D, Reddy M, Halani D. Tackling infertility in a case of bicornuate bicollis uterus with longitudinal vaginal septum: An arduous clinical challenge. *Medical Science* 2023; 27: e169ms2816. doi: <https://doi.org/10.54905/disssi/v27i133/e169ms2816>

Authors' Affiliation:

¹Post Graduate Resident, Department of Obstetrics and Gynaecology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be university), Wardha, Maharashtra, India

²Professor, Department of Obstetrics and Gynaecology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be university), Wardha, Maharashtra, India

³Associate Professor, Department of Obstetrics and Gynaecology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be university), Wardha, Maharashtra, India

Contact List

Nidhi Goyal	goyal.nidhi96@gmail.com
Manjusha Agrawal	badnemanjusha@gmail.com
Deepika Dewani	deepika.d3@gmail.com
Manila Reddy	manilareddyeti444@gmail.com
Dhruva Halani	Dhruva.halani@gmail.com

ORCID List

Nidhi Goyal	0000-0002-5458-2814
Manjusha Agrawal	0000-0003-2378-2925
Deepika Dewani	0000-0002-2165-0593
Manila Reddy	0000-0001-9204-7727
Dhruva Halani	0000-0003-4989-3324

*Corresponding author

Professor, Department of Obstetrics and Gynaecology, Jawaharlal Nehru Medical College, Datta Meghe Institute of Higher Education and Research (Deemed to be university), Wardha, Maharashtra, India
Email: badnemanju@gmail.com
ORCID: 0000-0003-2378-2925

Peer-Review History

Received: 07 January 2023
Reviewed & Revised: 10/January/2023 to 16/March/2023
Accepted: 20 March 2023
Published: 27 March 2023

Peer-review Method

External peer-review was done through double-blind method.

Medical Science
pISSN 2321-7359; eISSN 2321-7367

This open access article is distributed under [Creative Commons Attribution License 4.0 \(CC BY\)](https://creativecommons.org/licenses/by/4.0/).

Tackling infertility in a case of bicornuate bicollis uterus with longitudinal vaginal septum: An arduous clinical challenge

Nidhi Goyal¹, Manjusha Agrawal^{2*}, Deepika Dewani³,
Manila Reddy¹, Dhruva Halani¹

ABSTRACT

Bicornuate bicollis uterus is a congenital Mullerian anomaly which is comparatively rare. It is associated with about 25% of women having early miscarriage and infertility. The incidence of the bicornuate uterus along with vaginal anomalies is infrequent and unknown. We are presenting an unusual case of 25-year-old female with long-term primary infertility that was diagnosed with Bicornuate Bicollis uterus along with longitudinal vaginal septum. She underwent resection of vaginal septum and fallopian tubal recanalisation for tubal blockage, after which the patient conceived spontaneously and delivered a healthy male child later. Although these uterine anomalies are uncommon, they can still have profound medical and psychological implications.

Keywords: Bicornuate bicollis uterus, Vaginal septum, Infertility, Uterine anomaly

1. INTRODUCTION

Mullerian anomalies or uterine malformations result from an abnormal paramesonephric duct fusion during early embryonic life. The incidence of uterine malformations among the general population is about 7%-8% (Parmar and Tomar, 2014). Bicornuate uterus results from the abnormal fusion of lower segments of paired Mullerian ducts and amounts to more than 10% of all Mullerian anomalies (Sugi et al., 2021). A bicornuate uterus can either have a single (unicollis) or a dual cervix (bicollis), based on the length of the duplication of the ducts (Stearns and Al-Khabbaz, 2018).

Few cases of bicornuate bicollis uterus have been reported earlier; with principal complain of primary infertility. Challenges increased in our case when the bicornuate bicollis uterus was complicated because of the presence of a longitudinal vaginal septum as it contributed to the magnitude of infertility. Mullerian anomalies need to be cautiously dealt as they lead to infertility and complications during the course of evaluation of pregnancy, such as recurrent pregnancy loss, malpresentation, preterm labour, IUGR,

abnormal placentation, postpartum haemorrhage and perinatal mortality (Dohbit et al., 2017).

2. CASE DESCRIPTION

Patient information

A 25-year-old nulligravida female, married with six years of primary infertility came to OBGY OPD with complaints of cyclical cramp-like pain in the lower abdomen. The pain was localized predominantly in the hypogastric region radiating to the back and relieved by analgesics and antispasmodics. She also complained of dyspareunia frequently with poor sexual gratification in both partners. There was no history of fever and no urinary or bowel complaints. The patient had a regular menstrual cycle with menarche at the age of 13 years. She had been married for the past six years. Despite of regular intercourse associated with dyspareunia; she was unable to conceive because of poor sexual gratification in both partners.

Clinical examination

Her vitals were within normal limits. BMI: 20 kg/m², no visible thyroid swelling. She had tanner stage V secondary sexual characteristics. Abdominal examination revealed mild tenderness in the lower abdomen. On local examination; labia, fourchette and mons pubis appeared normal and had a perforated hymen. On separation of labia (Figure 1), a longitudinal vaginal septum between the anterior and posterior vaginal wall was seen dividing the vagina into two separate unobstructed cavities of different sizes. On difficult per speculum examination, two vagina and two Cervices were visualized, which were normal and healthy.



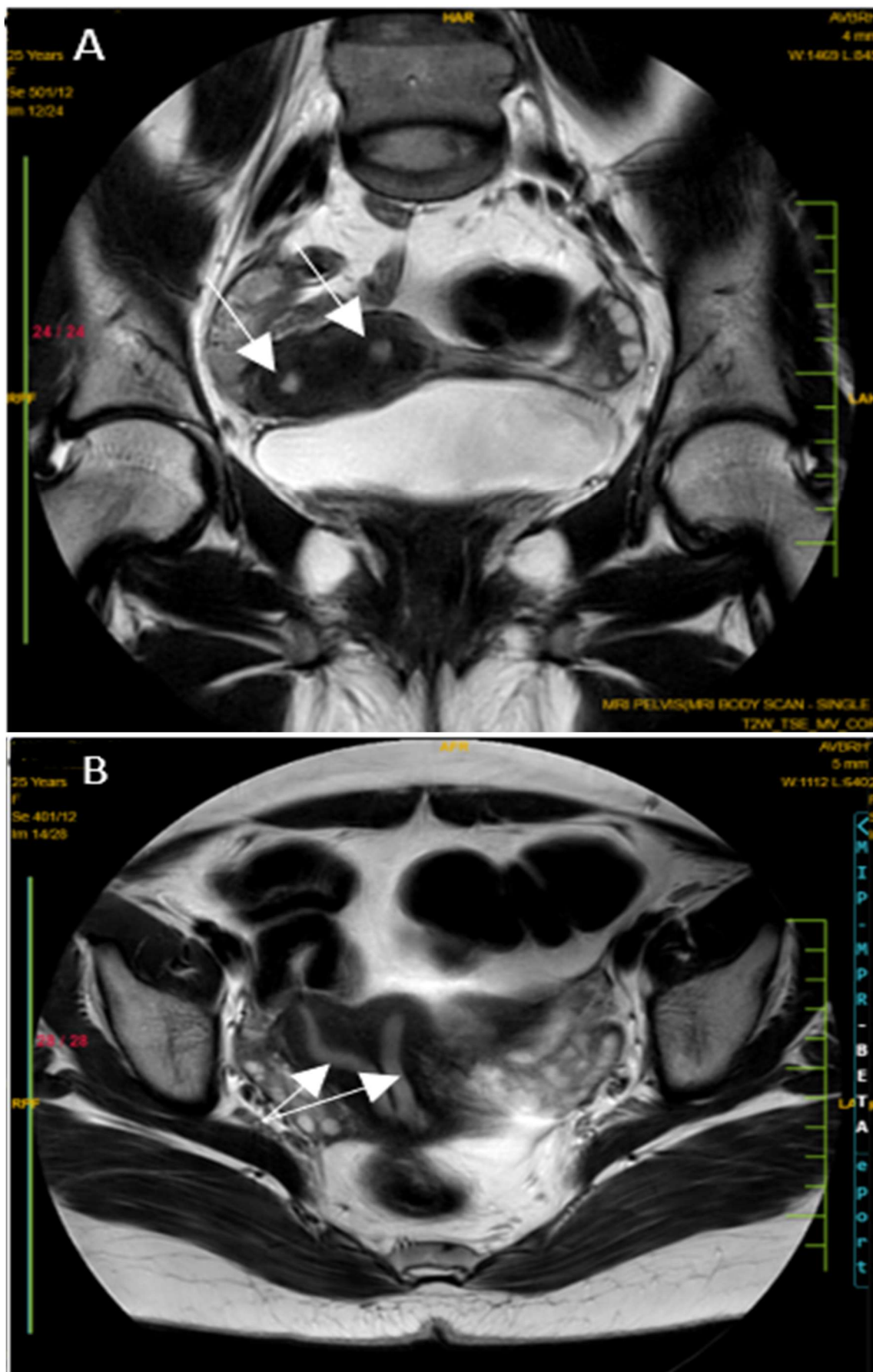
Figure 1 Longitudinal vaginal septum on per speculum examination (arrow)

On Per vaginal examination done under anaesthesia, cervices could not be reached. Her routine blood investigations, including complete blood count, thyroid profile, Anti-mullerian hormone, serum Prolactin and electrolytes were within normal limits. Husband's semen analysis was normal.

Diagnostic assessment

Ultrasonography revealed uterus bifurcation cranially with intervening tissue consistent with normal myometrium suggesting a bicornuate uterus. No renal anomaly could be seen on USG. Bicornuate Bicollis uterus was confirmed on MRI. Following are the images of T2WI MRI where two uterine cavities can be seen in coronal plane (Figure 2(A)), axial plane (Figure 2(B)) and in axial fat suppressed MRI (Figure 2(D)) with bulky peripherally arranged cysts in the ovaries (yellow arrowheads) suggesting polycystic ovarian changes. Two cervical cavities can also be seen (Figure 2(C)).

On diagnostic Hysterolaparoscopy, two cornua were visualized. The left uterus (cornu) had a single fallopian tube and ovary. The left uterus was normal in size but left ovary and fallopian tube appeared distended with a taut fimbrial end. The right uterus (cornu) also had a single fallopian tube and ovary (Figure 3). Dense adhesions were present between rectum and posterior surface of bladder (Figure 4).



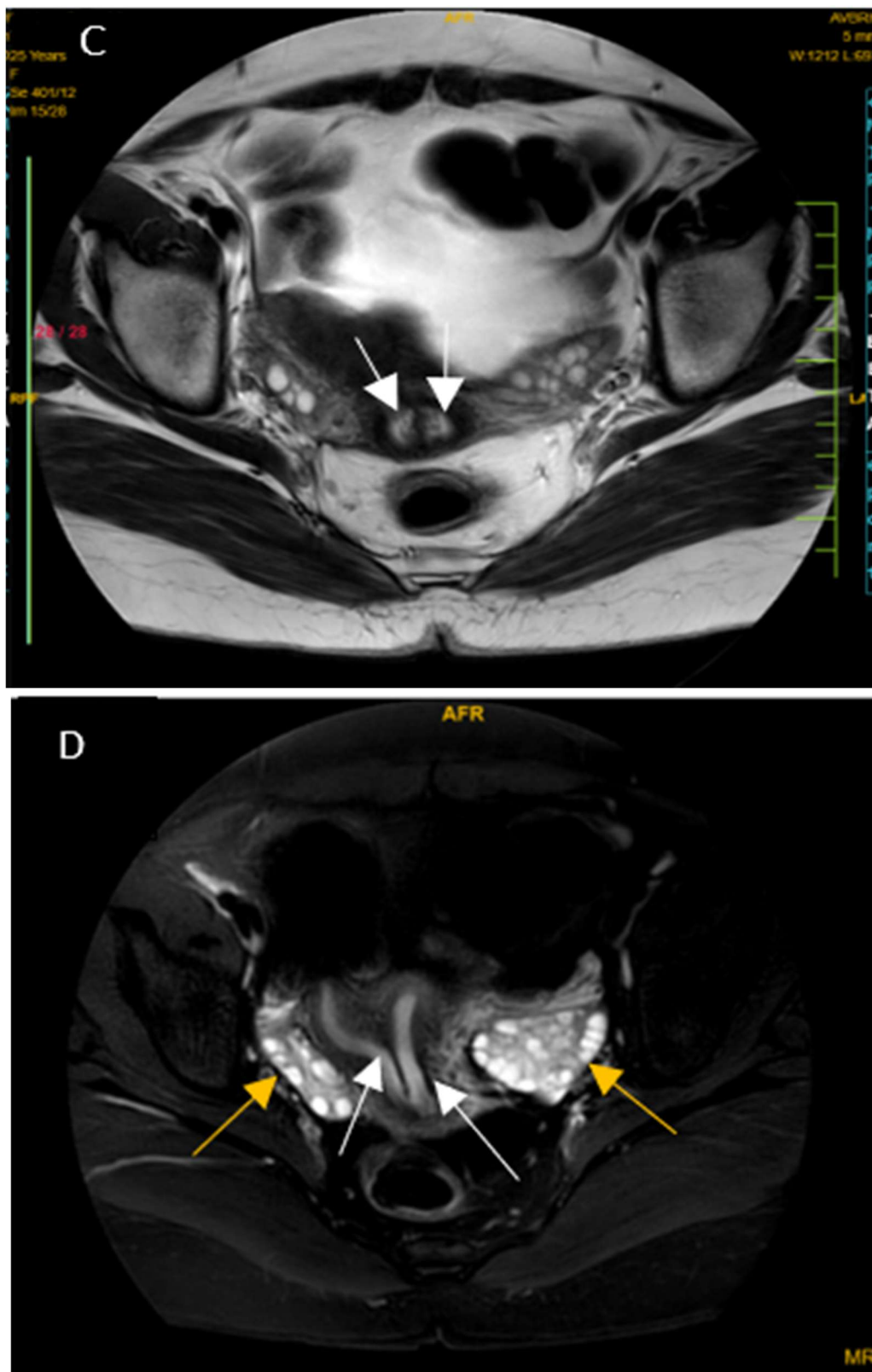


Figure 2 MRI images, A: Coronal T2WI MRI showing two uterine cavities (arrowheads), B: Axial T2WI MRI showing two uterine cavities (arrowheads), C: Axial T2WI MRI showing two cervical cavities (arrowheads), D: Axial T2WI Fat suppressed MRI, showing two uterine cavities (white arrowheads) with bulky peripherally arranged cysts in the ovaries (yellow arrowheads) suggesting polycystic ovarian changes

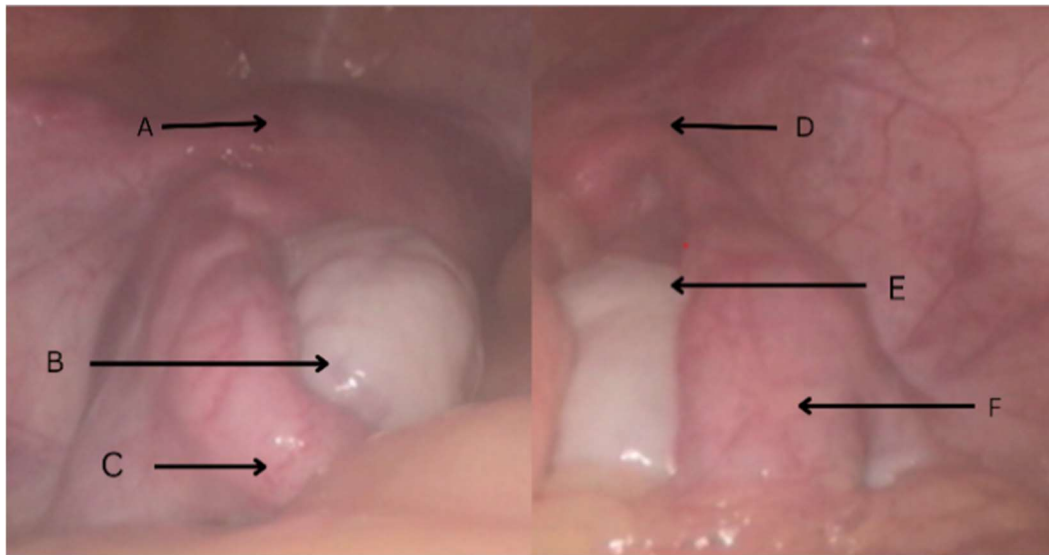


Figure 3 Two cornua along with two fallopian tubes and ovaries. (A: Left Cornu, B: Left sided Ovary, C: Left sided Fallopian Tube, D: Right Cornu, E: Right sided Ovary, F: Right sided Fallopian Tube) as seen on laparoscopy

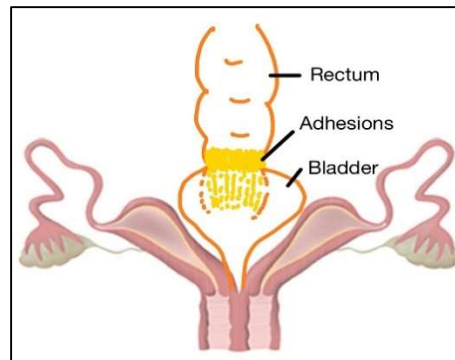


Figure 4 Intraoperative description of bicornuate bicollis uterus with adhesions between rectum and posterior wall of bladder

Hysteroscopically, both uterine cavities were visualized. Both had normal uterine walls with single ostia in each one of them. Flimsy adhesions were present over the left uterine ostia. Adhesiolysis was done to widen the ostial opening. Chromopertubation was suggestive of bilateral tubal blockage. Longitudinal midline fibrous vaginal septum was visualised during vaginoscopy (Figure 5). Resection of the vaginal septum from the posterior vaginal wall was done. Two Kocher forceps were applied at both the ends of septum to prevent blood loss. Metzenbaum scissors were used to excise the septum. Hemostatic sutures were taken using 3-0 absorbable sutures. Later fallopian tube recanalization was done to improve the odds of future fertility.

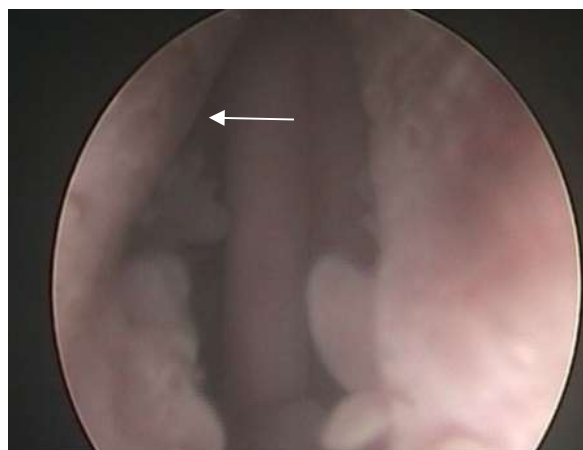


Figure 5 Longitudinal midline fibrous vaginal septum visualised (arrowhead) during vaginoscopy which was resected

Follow up and Outcome

The patient was advised for conception by natural method for 3 months. If unable to conceive by natural method, she was counselled that she could undergo assisted reproductive technology also. She came in gyne OPD after 3 months with a history of 2 months of amenorrhea. She conceived spontaneously and subsequently delivered a healthy male child weighing 2.3 kg at 36 weeks of gestation which was uneventful.

3. DISCUSSION

Bicornuate bicollis uterus is congenital uterine anomaly due to abnormal lateral fusion of paramesonephric duct during early embryonic life. Bicornuate uterus belongs to class III of mullerian duct anomalies according to ASRM. Diagnosis of bicornuate and bicollis uterus based on USG and MRI is done using CUME classification (Figure 6) (Practice Committee of the American Society for Reproductive Medicine, 2016).

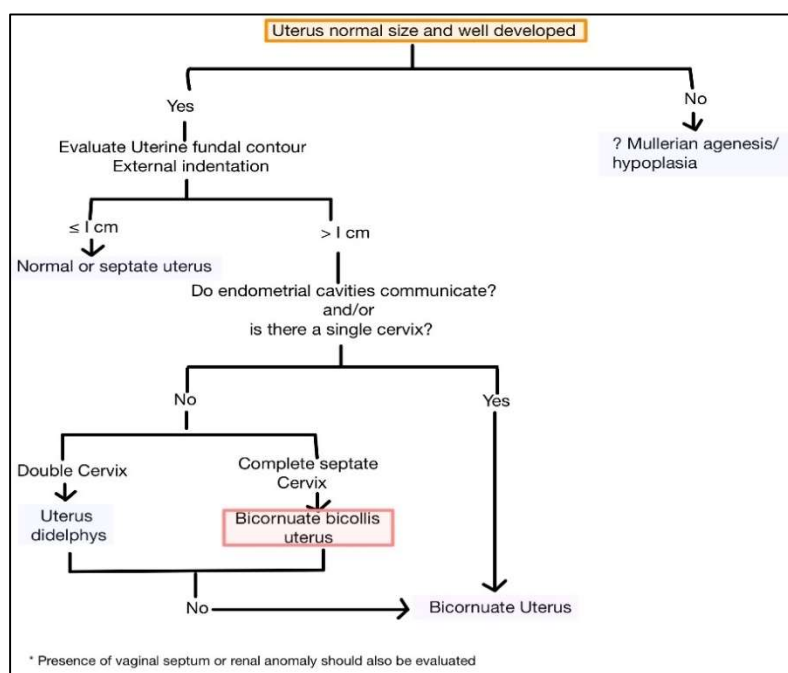


Figure 6 Algorithm for diagnosis of Bicornuate Bicollis uterus with longitudinal vaginal septum as per CUME classification

These anomalies are associated with numerous undesired outcomes such as infertility, recurrent pregnancy loss, malpresentations, preterm labour, IUGR, abnormal placentation, postpartum hemorrhage and perinatal mortality (Ravikanth, 2017). Even after above-enumerated complications, successful spontaneous pregnancy in a bicornuate bicollis uterus is still attainable.

Chu and colleagues did a similar study where longitudinal vaginal septum was resected using a surgical stapler. The procedure went uneventful with minimal blood loss. After two years patient reported back to the OPD with male partner dyspareunia because of protruding stapler pins at the septal margin of the vagina. Even though our method of resection was traditional, it has better outcomes in reinstituting the genital tract anatomy with minimal complications. This conventional method was preferred also because it was economically feasible by the patient. It is essential on the part of clinician to rule out other possible causes of infertility in such cases before counseling for assisted reproductive technology (Borgohain and Srivastava, 2017).

4. CONCLUSION

Pregnancy with bicornuate uterus in itself is associated with perinatal mortality and morbidity. In this patient, two cervices (bicollis) and longitudinal vaginal septum added to the long series of known complications such as infertility, recurrent pregnancy loss, malpresentations, preterm labour, IUGR, abnormal placentation, postpartum hemorrhage. Our patient had long duration of marriage with poor sexual gratification and dyspareunia due to longitudinal vaginal septum. Diagnosis was delayed for 6 years due to patient's unawareness and inhibition of expressing her sexual issues of dyspareunia and poor sexual gratification. As per the patient her inability to reproduce was unwelcomed in her society which had masses of implications on her mental health. Our

minimal surgical intervention helped the patient and husband to complete their family and recoup their society status. While such abnormalities are extremely rare, it is imperative for us clinicians to be able to appropriately manage the affected patients to improve their psychological, sexual and reproductive outcomes.

Ethical approval

Not applicable.

Informed consent

Written consent was obtained from the patient.

Funding

This study has not received any external funding.

Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

REFERENCES AND NOTES

1. Borgohain D, Srivastava S. Pregnancy in bicornuate uterus. *Int J Reprod Contracept Obstet Gynecol* 2017; 7(1):346–348. doi: 10.18203/2320-1770.ijrcog20175874
2. Dohbit JS, Meka E, Tochie JN, Kamla I, Mwadjie D, Foumane P. A case report of bicornis bicollis uterus with unilateral cervical atresia: An unusual aetiology of chronic debilitating pelvic pain in a Cameroonian teenager. *BMC Womens Health* 2017; 17(1):39. doi: 10.1186/s12905-017-0396-9
3. Parmar M, Tomar S. Bicornuate Uterus: Infertility Treatment and Pregnancy Continuation without Cerclage: An Unusual Case. *Open J Obstet Gynecol* 2014; 4(15):981–985. doi: 10.4236/ojog.2014.415138
4. Practice Committee of the American Society for Reproductive Medicine. Electronic address: ASRM@asrm.org and Practice Committee of the American Society for Reproductive Medicine. Uterine septum: A guideline. *Fertil Steril* 2016; 106(3):530–540. doi: 10.1016/j.fertnstert.2016.05.014
5. Ravikanth R. Bicornuate Uterus with Pregnancy. *J Basic Clin Reprod Sci* 2017; 6(2). <https://www.jbcrrs.org/abstract/bicornuate-uterus-with-pregnancy-4079.html>
6. Stearns K, Al-Khabbaz A. Bicornuate Bicollis Uterus with Obstruction of the Lower Uterine Segment and Cervical Prolapse Complicating Pregnancy. *Case Rep Obstet Gynecol* 2018; 2018:8910976. doi: 10.1155/2018/8910976
7. Sugi MD, Penna R, Jha P, Pöder L, Behr SC, Courtier J, Mok-Lin E, Rabban JT, Choi HH. Müllerian Duct Anomalies: Role in Fertility and Pregnancy. *Radiographics* 2021; 41(6):1857–1875. doi: 10.1148/rg.2021210022