THE JOURNAL OF OBSTETRICS AND GYNECOLOGY OF INDIA

The Journal of Obstetrics and Gynecology of India (September–October 2015) 65(5):346–349
DOI 10.1007/s13224-015-0740-7

INSTRUMENTATION AND TECHNIQUES

Two-Stage Repair of Complex Utero Cervico Vaginal Anomaly with Fish-Mouth Technique and Full-Thickness Skin Graft

Vinoo Balakrishnan[1] · **Manesh Senan**[2]

Received: 3 March 2015 / Accepted: 1 July 2015 / Published online: 8 September 2015

About the Reviewer

Dr. Vinoo Balakrishnan is currently working as senior consultant in the Department of Gynaecology, with special interest in minimally invasive surgery at Kerala Institute of Medical Science (KIMS), Trivandrum, Kerala. He is graduated from Trivandrum Medical college and did his MD from the same college in 1991. He worked as Registrar and clinical tutor at Cork University Hospital, Republic of Ireland, for 9 years while he took his MRCPI and MRCOG from Ireland and UK, respectively. He had four articles to his credit. FRCOG was conferred in 2014 for his contributions to this speciality. Since 2002, he has been attached to Mullingar Regional Hospital and Kerry General Hospital where he works as a consultant. He organizes basic laparoscopy workshop in KIMS Hospital, Trivandrum every year.

Dr. Vinoo Balakrishnan is senior consultant in Department of Gynecology and Dr.Manesh Senan in Department of plastic surgery and reconstructive, Kerala Institute of Medical Sciences, Kerala, India.

Electronic supplementary material The online version of this article (doi:10.1007/s13224-015-0740-7) contains supplementary material, which is available to authorized users.

✉ Vinoo Balakrishnan
vinu9977@hotmail.com

Manesh Senan
maneshsenan@yahoo.com

[1] Department of Gynecology, Kerala Institute of Medical Sciences, Anayara.P.O., Trivandrum, Kerala 695029, India

[2] Department of Plastic Surgery, Kerala Institute of Medical Sciences, Anayara.P.O., Trivandrum, Kerala 695029, India

Abstract Description of successful correction of complex Mullerian anomaly, in stages using Full thickness skin graft, prior to the onset of menarche.

Keywords Primary amenorrhea · Complex mullerian anomaly · Fish mouth technique · Full thickness graft

Introduction

Vertical fusion anomalies (uterocervical malformation and vaginal agenesis) are uncommon [1] with a prevalence rate of 6.7 %; when present along with transverse fusion anomaly (hemi uterus), it is even more rare, requiring the

need for complex reconstruction techniques. Coupled with the fact that the patient is living in an orthodox social setup where normal sexual development with menstrual cycles is required for woman to get married, this can pose a serious dilemma for the treating doctor, especially when convention has been to do hysterectomy.

Case Report

A 13-year-old girl who had normal female karyotype presented to local gynecology department since she had not attained menarche. She did not have any monthly cyclical abdominal pain. The body contours, breasts, and distribution of axillary and pubic hair were of normal female pattern. The clitoris, the labia majora, minora, and urethra were all normal. No vaginal introitus or dimple could be found on careful examination of the mucous membrane from urethra to anus.

Investigations

Blood biochemistry showed that her Follicle Stimulating hormone (FSH) and Luteinizing Hormone (LH) levels were in prepubertal zone. Initial ultrasound examination of abdomen and pelvis showed the absence of vagina and cervix. Uterus was smaller, located more toward right lateral wall. Both ovaries were normal. No renal anomalies were detected in that scan. The endometrium was very thin. MRI confirmed the findings of USS but also showed a small nubbin-like structure close to the left ovary.

Treatment plan consisted of a monthly Ultrasound scan (USS) to determine the thickness of endometrium and surgical correction, if there is an increase in thickness. Meanwhile, she agreed to undergo Franks method of creating an artificial vagina, without operation, by daily pushing the graduated tubes against the perineum. USS done on the fourth month showed normal endometrial thickness, but there was a small endometrioma adjacent to the right ovary on USS. Hence, it was decided to go ahead with a diagnostic laparoscopy, to drain the endometrioma and confirm the USS findings.

A diagnostic laparoscopy was done under general anesthesia. It showed that the upper abdomen was normal. Left ovary and left fallopian tube were also normal. A rudimentary uterine horn was detected on the left side which seemed to be non-functioning and non-communicating. On the right side was a relatively enlarged uterine horn, with normal right round ligament and normal right Fallopian tube. A transverse thin band of tissue was seen running from one uterine horn to the other between bladder and rectum. The uterine horn on right side had a tapering end pointed toward bladder area. A small endometrioma of dimension 2×3 cm was seen in the right ovarian fossa, which was enucleated, and bipolar cauterization of the endometrioma was done. There was no hematosalpinx on the right tube.

In the light of the findings, a complete and frank discussion with the parents was done especially concerning the doubtful reproductive capabilities even after multiple complex reconstruction procedures were done, which could have complications. Nevertheless, the parents were affirmative in their opinion that reconstruction was their only hope for the future social well-being of the child since, in many places in India, normal periods are a sign of positive reproductive health and a necessity for an arranged marriage.

Treatment

Since the patient did not find the Franks method much useful, she was admitted for surgical vaginal reconstruction. After routine pre-operative preparation, under spinal epidural anesthesia, a transverse incision was made between urethra and the rectum after inserting a Foley catheter into bladder. The space between urethra and rectum was dissected carefully by a combination of sharp and blunt dissection. A small amount of bleeding was controlled by pressure. The space was gradually enlarged to admit two, then three, and finally four fingers, and then a vaginal mold was inserted. A full-thickness skin graft of about 10 cm in length and 4 cm in width from both the inguinal areas was taken. The graft was sewn length-wise and placed over the mold, inside out. Then, the mold and graft were inserted and held in place by suturing labia majora over it. Post-operative period was normal, except that she was asked to be in bed for a week with catheter in situ, and soft diet was given for first week. The donor site of full-thickness graft was closed primarily. On the 6th post-operative day, the mold was removed and she was given a dilator to be used on daily basis, to prevent the collapse of this artificial space. She was advised to take progesterone orally to suppress her menstrual function.

Four months later, she was brought back for the final phase of reconstructive surgery. She had an overnight fasting before day of surgery. The surgical team comprised the author and an experienced plastic surgeon who had performed several vaginal reconstructive surgeries before. A single dose of 1500 mg of Cefazolin with 100 ml of 500 mg Metronidazole was given 30 min before the operation for anti-microbial prophylaxis. Under general anesthesia, the patient was placed in the dorsal lithotomy position. However, in order to allow the combined abdomino-perineal approach, the lower extremities were placed

in stirrups, positioned with hips abducted 45° and flexed 45°. After catheterization of the bladder, the abdominal and vulvar areas were cleaned and draped. The operation started with a pfannenstiel incision. On exploration, the findings were the same as before. The left tube with a rudimentary bulb on the left side was excised. The right uterus was mobilized to midline by dividing the round ligament. The tapered lower end of the uterus was resected until normal looking endometrium was seen. The uterine size was much smaller than the normal. At this juncture, the lower part of the uterus was split antero-posteriorly with stay sutures on both lips.

The second surgeon advanced a dilator through the previously constructed neovagina which was well healed and epithelized. The dilator was palpated between rectum and bladder and the area was opened transversely. Hemostasis was attained. Foley catheter was inserted through the vagina and into the uterine cavity. Since the lumen of the uterus was much smaller, only a 7F pediatric catheter with 2 ml of saline could be accommodated. Gentle traction of this Foley brought the uterus toward the vagina, and the uterus was stitched by Fish-mouth technique to the neovagina by 2.0 PDS on all four corners. This allowed to increase the lumen at the site of anastomoses and also reduce the chances of stricture. Supporting sutures were inserted. After pelvic lavage, abdomen was closed.

In the post-operative period, the plan was to keep the Foley in the vagina for 2 weeks, but it came out accidently on the fifth day. Apart from antibiotic coverage, she was on LMWH and was asked to continue on with progesterone to suppress her menstruation. She was discharged home on 7th post-operative day and advised to use vaginal dilator and continue on progesterone. After 6 weeks, progesterone was stopped and she had normal menstruation. Since then she had 6 cycles which were scanty in amount and painless. The two subsequent USS performed 3 months apart showed no evidence of any hematometra or endometriosis.

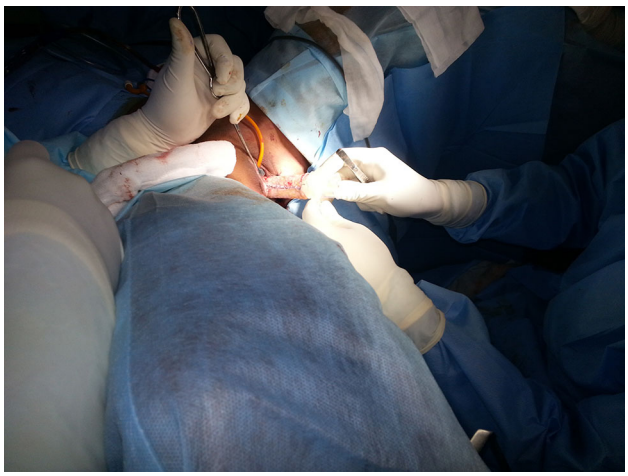

Fig. 1 Creation of neovagina

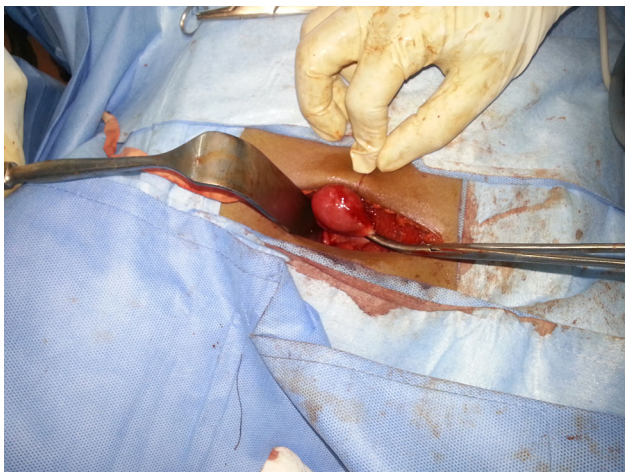

Fig. 2 Right non communicating uterine horn

Discussion

Fallopian tube, uterus, cervix, and the upper two-third of vagina develop from a pair of Mullerian ducts. The ovaries and the lower third of vagina develop from primitive Yolk sac and sino vaginal bulbs, respectively [2].

Three phases such as organogenesis, fusion, and resorption need to occur for the normal Mullerian duct development.

Our case had defects both in organogenesis and fusion leading to complex Mullerian duct anomaly (MDA). Treatment and fertility outcome differs widely between anomalies, and hence, a proper classification of MDA is vital. The most common classification system is developed by the American society of Reproductive Medicine. This index case falls into Class II but with a subtype (ii) non-cavitary (non-functional) rudimentary horn on the left side and a subtype (iv) Cavitary non-communicating rudimentary horn on the right side. This subtype (iv) Cavitary non-communicating rudimentary horn on the right side was the one that produced right-sided endometrioma of the right ovary.

Management of women with this malformation remains confusing. Total hysterectomy is recommended when canalization procedures fail or are impossible [3]. In this case, the parents were persistent for reconstruction on account of their concern for the future social well-being of their child in spite of informing them that childbearing may not be possible.

Most cases present with cyclical abdominal pain and absent periods along with hematometra, which require urgent surgical procedures for drainage and or reconstruction [4]. Fortunately in our patient who presented

early, we could follow up and suppress her menarche until final procedure was complete (Figs. 1, 2).

In the literature, although more than 10 surgical procedures for neovaginal reconstruction [5] have been described so far, the best has not yet been identified. Mc Indoe technique—split-thickness graft—is generally used which requires frequent dilatation or sexual intercourse to maintain the patency. Hence, this procedure is done close to period of sexual activity in adult population. In this particular case, considering the age of the patient, we have used full-thickness grafts which not only require less dilatation of up to 3–6 months, but also tend to adjust better for the growth of the perineum and maintain better moisture on account of some of sweat glands.

Simultaneous creation of neovagina and uterovaginal fistula can be associated with complications even with a normal lumen uterus. Graft maceration, sloughing, stenosis, and detachment are seen mostly when combined procedure is done, i.e., when both the neovagina creation and attachment of the uterus to the newly created vagina are performed. Here, we did it in stages, so when the uterus was brought to the upper end of the vagina we had already created, the well-healed and epithelized vaginal walls were available for holding the sutures.

When both vagina and cervix are absent and a functioning uterine corpus is present, it is difficult to obtain a satisfactory fistulous tract through which menstruation can occur. Conservative surgical treatment of uterine cervical atresia, mainly canalization techniques, is at high risk of secondary stenosis of the cervix, up to 40–60 % [6]. Open anastomoses through combined abdomino-perineal approach and the Vinoo–Manesh fish-mouth technique with vertical and horizontal slits help create a wider anastomotic region with less chances of stricture in even a small uterus so much that stent may not be required for more than 4–5 days.

Moreover, some authors conclude that the chance of subsequent pregnancies is unlikely, particularly in association with vaginal aplasia. Consequently, hysterectomy was recommended as the first line of management by many authors. When a patient is diagnosed to have complex mullerian anomaly, she and her family should be counseled in detail before surgery. The surgical methods, interval between the stages, and the possible complications including bladder and rectal injury should be addressed. Both the patient and her family are to be made aware of the future fertility, the possibility of stenosis, and the consequent issues.

Conclusion

We report this case to highlight the difficulties and challenges encountered in the management of complex mullerian duct anomalies. Although there had been a few reports of successful reconstruction, the management of these cases still remains a challenge in gynecology. This case is the one which was diagnosed prior to menarche and treated in two stages: Full-thickness graft was used for the creation of neovagina and Vinoo–Manesh fish-mouth technique for uterovaginal anastomoses, which can be beneficial in the cases of narrow uterine lumens like in this case. The goals of the reconstructive surgery are to provide a conduit for menstrual flow, to relieve pain, and to preserve reproductive potential.

Compliance with Ethical Standards

Conflict of interest Dr. Vinoo Balakrishnan and Dr. Manesh Senan declare that they have no conflict of interest.

References

1. Roberts CP, Roche JA, et al. Surgical methods in the treatment of Congenital anomalies of Uterine cervix. Curr Opin Obstet Gynaecol. 2011;23(251):257–1042.
2. Chandler TM, Machan LS, Cooperberg PL, et al. Mullerian duct anomalies: from diagnosis to intervention. Br J Radiol. 2009; 82(984):1034–42.
3. Acien P, Acien M. FerrerMS: complex malformation of the female Genital tract. New Types and revision of classification. Hum. Reproduction. 2004;19:2377–84.
4. Beksac MS, Salman MC, et al. A new technique of surgical treatment of vaginal agenesis using combined Abdomino-Perineal approach; Case reports in Medicine. Volume 2011(Article ID 120175).
5. Nichols DH, Randall CL. Creation of a neovagina: from vaginal surgery. 3rd ed. New York: Appleton; 1989.
6. Hafeez M, Haleem S. Functioning uterus with absent cervix, short vagina and transeverse septum. J Coll Physicians Surg Pak. 2008;19(60):386–8.