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Secretary General's Office

Dr Meena Samant, Secretary General, ISOPARB
21/D, Road No 10, Rajendranagar, Patna - 800016
Phone: +91 93341 05945 | E-mail: meenasamant@rediffmail.com

Indian Journal of Perinatology and Reproductive Biology

CD 55, Sector I, Salt Lake City, Kolkata 700 064
E-mail: ijoparb1978@gmail.com

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Editorial Office: CD-55, Salt Lake City
Sector-I, Kolkata 700064
Email: ijoparb1978@gmail.com

Contents

Editor's Choice

Learning to Work Remotely … 5
Prof (Dr) Hiralal Konar

Views & Reviews

Changing Trends in Management of Labor … … … … … … … … … … … … … … … … 8
Prof (Dr) Hiralal Konar

Case Report: Obstetrics

A Challenging Case of Takayasu Arteritis in Pregnancy with Successful Fetomaternal Outcome 12
Dr S Harinisri, Dr Dipika Roy, Dr Samaresh Malo, Dr Sharmistha Ganguly, Prof Tapan Kumar Maiti

Rare Case of Pregnancy in Left Non-Communicating Rudimentary Horn of Unicornuate Uterus … 15
Dr Shuchi Gupta

Pregnancy with Gaucher's Disease – A Case Report … … … … … … … … … … … … … … 18
Dr S Harinisri, Dr Sanjib S Mondal, Prof Tapan Kumar Maiti

Review Article: Gynecology

Screening for Congenital Hypothyroidism … … … … … … … … … … … … … … … … … 21
Dr. Meena Samant, Dr. Juhi Sisodia, Dr Divya Suman

Clinical Article: Gynecology

Experience of Conducting Cervical Cancer Screening During Covid-19 Pandemic … … … … 24
Dr Indira Palo, Dr Shweta Mohanty, Dr Payal Keswarpu, Dr Bharati Mishra

Case Report: Gynecology

Gossypiboma — A Surgeon's Nightmare … … … … … … … … … … … … … … … … … 28
Dr Amit Basu, Dr Deblina Kar

Swyer Syndrome in Siblings: A Rare Presentation… … … … … … … … … … … … … … 32
Sree Reshmi R J, Pooja D C, Dr Siddhartha Datta, Dr Shamim Khandekar

Instruction to Authors … 38

Editor's Choice

Learning to Work Remotely

The day of submitting this editor's choice in the pandemic of Covid-19, in India, 4,22,19,896 cases recovered amongst the total of 4,28,81,179 infected cases. 96,46,98,071 number of Indians received the Dose – 1 vaccines, 78,44,78998 numbers of populations received the second dose and 1,85,54,653 received the precautionary dose (GOI, COWIN). Keeping in mind the India's huge population, such coverage within this short spell of time, is no doubt a mammoth task.

We all thought, we will have a sigh of relief now. Unfortunately, this planet is still in unrest. The world is on war, rockets rain down on Ukraine's cities as initial talks ended without success.

During the COVID – 19 pandemic, many journals (international) decided not to publish any COVID – 19 specific articles due to rapidity of its developments, changes in the disease pathology and management options. IJOPARB published views, reviews and articles all throughout the period for the members to keep them updated and to stay safe. In our profession the impact of COVID – 19 pandemic continued to be very significant. The initial opinion was with the vaccine that could not be offered to a pregnant woman and even to a breast feeding mother. We all faced difficulties with this virus, the disease and its treatment. Initially physicians and the lactating mothers were worried as there was relative scarcity of formal studies with this issue specially with the drugs. At the same time many became very fearful with the vaccine. Surgical management was reduced significantly, longer waiting list was for patient's appointment. The article by Mullin et al[1] was very reassuring. Many women who were scared, restarted breastfeeding thereafter.

Amidst all the adversities, many of us could see some opportunities. In these pandemic years technology enhanced healthcare framework, helped us to provide patient care service remotely. It has now become the talk of the day. The terminology 'telemedicine' according to General Medical Council (GMC) is the provision of medical services and patient care from a distance using information and communication technology (ICT).[2,3]

The aim of telemedicine is to support decision making, training, medical education and to support health care on remote basis. Types of telemedicine modalities are different. In synchronous modality, a specialist is in consultation with a patient virtually where another lead clinician is often present with the patient. Whereas in the asynchronous modality, there is transmission of information such as digital images (ultrasound, histopathology) or video clips to a specialist of a tertiary care for review and opinion. Other modalities are: remote patient monitoring like sending individual patient data (BP, fetal movement record etc.) to the specialist for analysis. In obstetrics, virtual consultation can be obtained in the issue of FHR monitoring, blood pressure and fundal height etc.

Use of telemedicine as a modality in obstetrics is not a new one. Working in UK even in early '90s we had the experience of sharing information and communication for CTG tracing analysis. The pregnant women waiting at the community centre were consulted with CTG-tracings that we received through the fax. On the other end, the trained midwife and the GP were there to support the pregnant woman. However the record states that telemedicine was first described by Bohem and Haire in 1979.[4]

Currently GMC and the National Health Service in England, have made guidance for practicing telemedicine.[5] The key considerations for conducting remote consultation are divided into five main areas.[6] Planning of service needs: (a) a dedicated team with required equipment and technology. For this appropriate training is a must, (b) a confidential setting with confirmed patient identity. Minimum two identifiers are considered. (i) Date of birth and (ii) Address, (c) The health care team need to select patients suitable for remote consultation (e.g. low risk, chronic follow ups, no need for physical examination), (d) proper documentation as per consultation, the follow ups and agreed action plan and (e) The patient should be in a private well lit room. There are provisions for pre consultation questionnaires also.

Unlike traditional medical therapies, digital technology is evolving very fast. It is becoming an integral part of modern health care science. Mass production of health care apps, mobile platforms, advanced technology are to facilitate digitalized healthcare via remote communication system and consultation. Use of telemedicine in the discipline of obstetrics and gynecology is on the way. Opinions favor to develop patient initiated virtual follow up clinics.[7] Regular face to face scheduled clinics are to be there also. Arrangements are to be initiated for home monitoring devices (maternal blood pressure, blood glucose testing, FHR monitors) as discussed above.

It is expected that use of telemedicine is going to meet the challenges of rising numbers of outpatient appointments and to reduce the larger waiting lists. This is especially so in the busy hospital clinics like the government run medical colleges and the tertiary care centers in our country.

The Journal ISOPARB has become very popular and close to our members over the years. The journal shape, the attracting cover page, and get-up has undergone significant changes. The patterns of article presentation has been streamlined. Colored photographs in each issue are well appreciated. The journal is indexed since 2017, with much sincere support of our members. Moreover, members are happy to read the journal online depending on their available time.

"***Old order changeth yielding place to new ….***" - Lord Alfred Tennyson. As time passes by, we the existing team of executive body members of ISOPARB are to move on to the next position of the hierarchy. I am deeply indebted to all the members of ISOPARB, having the opportunity to serve the society over the last seven years. I thank the journal committee members each and all, for their kind support. Without their support, it would not have been possible for me to carry out this huge task.

I wish to thank all the members specially, Prof. Gita Ganguly Mukherjee, Prof. Sudip Chakraborty, Prof. A. Majhi, Editor Emeritus, Prof. S.N. Tripathy, Prof. Manju Gita Mishra, Past Presidents, Dr. Usha Sharma, President ISOPARB, Dr. Meena Samant, the Secretary General, ISOPARB, Dr. Picklu Chaudhuri, Prof & Head, Dr. Arindam Halder, Assoc Prof, Dr. Gita Basu Banerjee, Professor, Dr. R.P. Dey, Professor, Dr. Pallab Mistry, Assoc Professor, West Bengal University of Health Sciences for their support throughout the period.

Prof (Dr) Hiralal Konar
Editor-in-Chief
MBBS (Cal), MD (PGI), DNB, MNAMS, FACS (USA) FRCOG (London)
Member: Oncology Committee of AOFOG
FOGSI Representative to Asia Oceania Federation of Obstetricians and Gynaecologists (AOFOG)
Chairman, Indian College of Obstetricians and Gynaecologists (ICOG)
Prof. & Head, Dept. OB-Gyn, KPC Medical College and Hospital, Calcutta

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Views & Reviews

Changing Trends in Management of Labor

Prof (Dr) Hiralal Konar

"A LABOR which is unduly prolonged is likely to give rise to one or more of the three types of distress, namely maternal, fetal or "Obstetricians' distress". Of the three, the last may be easily the most dangerous!…. "Dawn should not rise twice upon the same labour" - Ian Donald. Practical Obstetrical Problems, Lloyd Luke, London, 1979.

Working as a registrar in one hospital of UK in late '90s, I had to conduct an audit for rising Cesarean Delivery (CD) rate from 11% to 13.6% for that year. We were three registrars working in that hospital. Audit revealed dysfunctional labor due to large baby was one of the lead causes.

Today, Cesarean Delivery (CD) rate has risen in most countries worldwide.[1] Factors for the rise of CD are many. Decreased use of vaginal operative deliveries, increased primary CD with low threshold of labor progress, failed induction of labor, advanced maternal age, increased maternal request and the litigation concerns are the few. WHO has proposed an incidence between 10% and 15% as a target to optimize maternal and perinatal health.[2] Questions arise how to determine an optimal CD rate keeping in mind the several variable clinical factors as mentioned above. Furthermore, it is to be decided whether we should primarily stress how to limit the CD rates or look into the maternal and perinatal health outcomes following child birth.

Management of labor, be it normal, dysfunctional or with cephalopelvic disproportion is an area of long debate and controversy. Philpott (1972) introduced the initial concept of early warning of abnormal labor by drawing a straight line representing the mean rate of progress of the slowest 10% of patients in the African population.[3] The lower limit was 1 cm per hour in the active phase. It was designated as 'alert line'. Later on a second line, the 'action line' was drawn initially four hours and later on changed to two hours and further changed to four hours to the right and parallel to the alert line.[3] With this it was possible to prevent dysfunctional labors. This view was widely accepted. Artificial rupture of membranes (ARM) is performed when the labor is confirmed and the head is four fifth or less is palpable abdominally. ARM is done to assess the state of the liquor or to augment labor when needed. When the labor progress remained to the left of the nomogram or less than two hour to the right, it is it is considered normal and no augmentation is needed. On the other hand, if the cervical dilatation strays more than two hours to the right of the nomogram, the labor is judged dysfunctional and contractions are stimulated with an intravenous infusion of syntocinon provided malpresentation or cephalopelvic disproportion are ruled out. Benefits are improved fetomaternal outcome and above all, reduction of cesarean delivery (CD) significantly.[4,3,5] These landmark studies[3] provided a clear and defined criteria for midwives working in peripheral clinics who needed to refer woman in labor to the higher facilities. These studies referred to the study of Harare Hospital, Zimbabwe (then Rhodesia). This result coincided with the reports of the National Maternity Hospital, Dublin with active management of labor. The sole objective of 'active management' was to achieve birth within a designated time frame using early amniotomy, with or without the oxytocin and one to one labor care.

In 1990s, it was endorsed by WHO as a useful tool for the management of labor. It was accepted globally.

However its utilization and correct completion rate were too low (31% and 3% respectively).

Alert line represents the slowest tenth centile of nulliparous women in labor. Since 2018, WHO initiated a process of revising the partograph keeping in mind the variability of an individual woman in the progress of labor and that ends in good maternal and perinatal outcomes.

Partograph is the graphical representation of the progress of labor in terms of women's cervical dilatation and descent of the fetus presenting part, against time (Philpott & Castle 1972).[3] Formal regular recording of important clinical parameters have been maintained covering the well-being of the woman and the baby.

Partograph has been used as the proper tool to make decision during labor with the aim of optimizing the time of referral and interventions.[6] The cervicograph in the partograph where cervical dilatation is plotted from 4 cm (earlier 3 cm). The acceptable cervical dilatation rate is 1 cm per hour. The alert line represented the slowest tenth centile of the nulliparous women in labor.[6] The action line is drawn at intervals of 4 hours (earlier 2 hours) to the right and parallel the alert line. The action line was meant to identify abnormal labor. This situation needed medical intervention that may suggest augmentation of labor or urgent delivery based on the underlying abnormality.

Currently several observational studies questioned the validity of one centimeter per hour rate to use as the benchmark for assessing the adequacy of labor progress. One systemic review[6] failed to assess the usefulness of the objective in optimizing referral of laboring women from rural or primary health care facilities to higher care units. One largest study in the review found a mild increase in the risk of adverse birth outcomes in slow labors compared to fast labor.[7] Importantly, the measures of identifying pregnant women at risk of developing complications through labor, delivery and immediate post partum are not perfect. It is true that perinatal outcomes are affected by many other reasons that are not related to cervical dilatation rates only.[8] The alert and action lines failed to identify women at increased risk of outcomes. Labor progression for women in normal labor may not be linear. The relevance of alert line is to allow reasonable time to the health care facilities where interventions such as augmentation or cesarean section cannot be performed and where referral facilities are difficult to organize to reach the higher level care centre. In sum, there is an urgent need for optimal benchmarks for assessing progress of labor and to make the decision when best to intervene to reduce adverse birth outcomes for the woman and her baby.

The advantage of the partograph in the AMOL is the simplicity of visual display of all the relevant events as recorded. It has its role in teaching and updating the labor care management. Active management of labor has been practiced at the National Maternity Hospital, Dublin for more than 50 years. The institute confirmed the diagnosis of spontaneous labor when cervix was effaced rather than by dilatation.

Progress of labor is strictly defined by the graphical record and interpretation of cervical dilatation and descent of the presenting part.[7,9,3,4] Sole objective was the detection of delay in labor at an early stage using the reference to cervical dilatation and descent of the presenting part plotting. Dysfunctional labor could be corrected before the onset of prolonged labor or obstructed labor. Dynamics of labor are more important than the mechanics. Friedman's sigmoid shaped curve was divided into latent phase until 3 cm, active phase at the rate of 1 cm/hour and the deceleration phase from 9 cm to full dilatation.

Among all the phases of labor, latent phase is an ill defined area. It is difficult to identify with certainty. It is understood and defined only in retrospect. Duration of latent phase of labor is variable. Overall in primigravida, it is 20 hours (average 8.6 hours) and 14 hours (average 5.3 hours) in multigravida. It is defined as the interval between the onset of labor pain until the cervix is 3 cm changed to 4 cm and now 5cm or 6 cm dilated. In primigravida, it corresponds to the process of cervical effacement. Friedman[10] stated that latent phase should not exceed 20 hours in a primigravida and not to exceed 14 hours in a multigravida. However it is sometimes difficult to distinguish between a prolonged latent phase from spurious labor or pre-labor phase. Therefore it is better to be conservative in approach of management.

National Maternity Hospital, Dublin practiced active management of labor in a different way. The diagnosis of spontaneous labor confirmed by effacement of the cervix rather than by dilatation. Debate arose in 1980 with the concept of active management of labor (AMOL) in lowering cesarean delivery (CD). National

Maternity Hospital in Dublin[11] (O'Driscoll et al 1988) and Parkland Memorial Hospital in Texas[12] were the main study centers to evaluate the merits of AMOL in lowering CD rates.

Current concept of partographic labor monitoring is beyond the limited boundaries of clinical assessment of uterine contractions and occasional vaginal examination. Labor Care Guide (WHO) has made several important changes. It emphasizes the need of evidence based practices and at the same time to maintain the woman's dignity, privacy and confidentiality. Following the Labor Care Guide, documentation is initiated once the active phase has been diagnosed. The terminology as alert and action lines, latent and active phases of labor are not used. Many of us feel, modern management of labor should embrace different ways of care and whether not the partogram is used or which rate of progress in labor is used to represent normal labor.[8] Key to labor management is situational accuracy knowing where one is making the labor progress. Laboring woman need individualized care.[13] Reason for rising rates of cesarean delivery (CD) are multiple as it is all over the world. Justifying the use of the partograph to lower CD rate remains a dilemma.

More recent data[14] from the Consortium of Safe Labor, revised the definition of normal labor. Active phase of labor is changed to at least 6 cm dilatation. Cervical change is now thought to be slower.[15]

The new WHO recommendations are designed for an individual woman's care during the course of labor. The new labor monitoring tool, "WHO Labor Care Guide"[16] directs clearly the method of labor monitoring, place of clinical interventions and above all, the main task is the respectful maternity care. WHO revised partograph, is thought to be a revolutionary step for the care of individualized woman in labor. The changes that have been introduced in the revised partograph and Labor Care Guide are: active phase is defined to start from 5 cm of cervical dilatation instead of 4 cm. Fixed rate limit of 1 cm/hour alert and action line in the active phase is replaced. It is guided by an evidence based time limit for each centimeter of cervical dilatation, derived from the 95th centiles of labor duration at different centimeter levels of women. It has been observed to result in normal perinatal outcome.[12,14] The cervical dilatation is recorded in the 'assessment' section and the steps are taken, in consultation with the woman. It is recorded in the 'plan' section. With this, iatrogenic cause of apparent poor progress of labor and at the same time unnecessary interventions are reduced.

The other major addition into the new care is the intensified monitoring of the second stage of labor. The strong uterine contractions, with the propulsive force combined with voluntary (expulsive) efforts make the second stage labor a more crucial time. Increased monitoring and vigilance are needed. The current "Labor Care Guide" emphasizes more attention for the mother and the baby.[17]

Supportive Care stresses the importance of clear recording of evidence based practices to improve woman's positive child birth experience and outcomes.

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A Challenging Case of Takayasu Arteritis in Pregnancy with Successful Fetomaternal Outcome

Dr S Harinisri,[1] Dr Dipika Roy,[2] Dr Samaresh Malo,[3] Dr Sharmistha Ganguly,[4] Prof Tapan Kumar Maiti[5]

Introduction

Takayasu arteritis is a chronic inflammatory arteritis affecting large vessels. It occurs before age 40 with upper extremity vascular involvement. MR angiography is the most important investigation and done without gadolinium contrast in pregnancy. Treatment is with steroids which is safe in pregnancy. Here, we are presenting a case of Takayasu arteritis in pregnancy which was carried till term with a successful fetomaternal outcome.

Case Report

A 26 year old G2P0+1 presented to NRSMCH outdoor at 32 weeks gestation being referred from a private practitioner as a case of Takayasu with echo report showing left atrium enlarged and probable left coronary artery aneurysm.. She had no complaints at that time. She gave history of ovulation induction with letrozole. On examination, her vitals were a pulse rate of 70/min on right upper limb which was low volume and 90/ min on left side which was high volume. Pressure was 110/70 mmHg on right side and 220/80 mmHg on left side. She had carotid artery bruit and bilateral renal artery bruit. Obstetric examination showed uterus size corresponding to period of amenorrhea, 32 weeks with regular heart sound of 136 bpm. Patient was admitted and was dealt with a multidisciplinary approach with opinions from cardiology, rheumatology and anaesthesiology.

Her blood pressure in all 4 limbs was monitored strictly twice daily and was maintained at 208/54 mmHg in left upper limb and 116/70mmHg in right upper limb and antihypertensive drugs dosage was adjusted at regular intervals to suit her needs. She was started on labetalol 100 mg three times a day increased to 400 mg three times a day and subsequently nifedepine 30 mg twice a day was added as pressure was not under control. Patient had no complaints of any premonitory symptoms of pre-eclampsia throughout pregnancy. Alternate day urine dipstick was done to rule out any new onset proteinuria and it was negative throughout pregnancy. Complete blood count, liver and renal function tests were repeated weekly and were normal. Takayasu specific imaging investigations were done and results are given in Figure 1. All these showed her to have Stage V disease. Ophthalmology examination was done and it revealed few dot haemorrhages in both eyes. Foetal monitoring was done with ultrasonography and colour Doppler every 3 weeks and showed normal growth and liquor

1. 3rd Year Post-Graduate Trainee, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata.
2. 3rd Year Post Graduate Trainee, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata.
3. VS, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata.
4. RMO cum Clinical Tutor, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata.
5. Professor, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata.

Corresponding author email: dipikasona83@gmail.com

Figure 1: Imaging investigations in antenatal period

MR Neck Angiography	Thickening in arch, right brachio cephalic artery, bilaterial common carotid artery Occlusion of left subclavian artery 40-50% stenosis left proximal mid common carotid artery No signal in vertebral artery Possiblity of Aorto arteritis
MR Renal Angiography	Diffuse mild disease in abdominal aorta Bilateral renal atery stenosis Occluded SMA 90-99% stenosis at celiac ostium
Repeat Echo	Concentric LVH LVEF 65% Grade 2 Diastolic Dysfunction Normal values No RWMA at rest
Colour Doppler	Narrowing in the lumen of bilateral renal arteries Acceleration time increased in bilateral sites RAR – 0.82 Diffuse wall thickening in bilateral common carotid arteries Palvus tardus waveform in bilateral intersegmental arteries

volume. She was put on Tab. Prednisolone 7.5 mg once a day and Tab. Aspirin 75 mg once a day from 32 weeks as per rheumatology advice.

Pregnancy was carried till 37 weeks gestation and since she was Stage V disease termination was by elective Lower segment caesarean section which was under combined spinal epidural as per anaesthesiology advice. Her intra op period was uneventful and her pressure was maintained at 236/92 mmHg. She delivered a healthy 2.250 kg baby girl. From anaesthesiologist side, she received dexamethasone 8 mg intravenous three times a day starting from intra operative period and continued postoperatively for 2 weeks.

Post op, patient was followed up in High dependency unit and her vitals were similar to antenatal period. Epidural top up was continued with infusion ropivacaine 0.15 % diluted in 50 ml NS at 5ml/hour for upto 60 hours post operatively. She was advised for follow up at cardiology and rheumatology outdoor post discharge and mother and baby was discharged in stable condition. She was discharged with labetalol and nicardia and Inj. Dexamethasone 6 mg intravenous three times a day for 2 weeks followed by oral prednisolone as per anaesthesiology advice.

Through telephonic conversation, we came to know that patient did not take any medicines as advised and did not come for follow up. One year after her discharge she developed cerebrovascular accident with weakness of the left side and at present she is recovering from that.

Discussion

Takayasu arteritis is a rare, chronic, inflammatory, progressive, idiopathic arteriopathy, affecting young women of reproductive age group, causing narrowing, occlusion and aneurysm of systemic and pulmonary arteries, aorta and its branches. Its incidence is reported to be 13 cases per million population with Asian predominance.[1]

Prognosis of Takayasu arteritis in pregnancy is worsened by comorbid severe renovascular hypertension, cardiac involvement or pulmonary hypertension. Involvement of abdominal aorta portends worse perinatal outcome. It can complicate pregnancy by causing pregnancy related hypertension, preeclampsia, abruption, congestive cardiac failure, progressive renal involvement, foetal growth restriction, preterm birth and foetal death. In our patient severe systolic hypertension was seen which was managed with a holistic approach with opinions from cardiologist and rheumatologist.

Clinical diagnosis is by using American College of rheumatology 1990 criteria where if 3 out of 6 criteria is fulfilled, the diagnosis is made. It includes age under 40 at disease onset, claudication of extremities, decreased brachial artery pulses, systolic blood pressure difference > 10 mmHg between arms, bruit over subclavian arteries or aorta and angiogram abnormalities.[2] In our patient, all these criteria were fulfilled. Imaging options in pregnancy include angiogram, echo and Doppler studies all of which were done in our patient.

Management in pregnancy is by routine antenatal check up with serial monitoring of blood pressure, renal function, cardiac status and preeclamptic screening. In our patient, these were followed. Foetal surveillance is by daily foetal movement count, foetal biometry, biophysical score and foetal Doppler. Cardiologist and rheumatologist opinion was taken and she was put on Prednisone and aspirin.

Vaginal delivery is usually preferred in these patients.[1] Induction of labour is not indicated and spontaneous onset of labour is awaited. Second stage of labour is cut short by instrumental delivery. Lower segment caesarean section is preferred from stage IIb and onwards of Numano classification to prevent cardiac decompensation due to increased blood volume and blood pressure observed during uterine contractions and increased cardiac output observed in labour. As our case was Numano Stage V, elective Lower segment caesarean section was performed.

Epidural anaesthesia provides stable hemodynamic and pain relief which was used in our patient.[1] It is associated with gradual onset of sympathetic block and decrease in pressure. There is smooth control of pressure in intraoperative and postoperative period. Regional anaesthesia is associated with sympathetic blockade and subsequent drop in pressure and compromised regional circulation due to stenosed arteries. There is risk of aortic dissection on table. In such a case patient presents with a sudden ripping pain, breathlessness or loss of consciousness. It is treated with beta blockers, narcotics, opiates, pericardiocentesis to relieve the pressure and urgent surgery.[3]

Conclusion

The optimal management of pregnant patients with this disease has not yet been defined. The course of the disease seems to be neither affected nor worsened by pregnancy. Although unfavourable foetal and maternal outcomes has been reported, our case had a favourable fetomaternal outcome. With an interdisciplinary collaboration, the prognosis of both can be improved.

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Case Report : Obstetrics

Rare Case of Pregnancy in Left Non-Communicating Rudimentary Horn of Unicornuate Uterus

Dr Shuchi Gupta

Abstract

Pregnancy in the rudimentary horn is rare and carries grave consequences for the mother and fetus. 90% of them present with intraperitoneal haemorrhage in the second trimester due to rupture of the horn.

A case report of unruptured pregnancy in a left rudimentary horn of unicornuate uterus at a gestational age of 30weeks 4 days. Laparotomy was done and the rudimentary horn excised along with left sided salpingectomy. The need for a high index of suspicion and the role of ultrasonography in the accurate diagnosis of such cases is highlighted.

Introduction

Pregnancy in a rudimentary horn of unicornuate uterus is rare. An incidence of 1 in 76,000 - 1,50,000 pregnancies is reported in the literature.[1] Unicornuate uterus is a congenital anomaly of the uterus and results from a non-developing Mullerian duct or agenesis of the Mullerian system.[2] It was first classified in 1979 by Buttram and Gibbons and further revised by the American Society of Reproductive Medicine in 1988.[3,4] It is a Type II classification that can be further sub classified into communicating, non communicating, no cavity and no horn.[2] In 83% of the cases the rudimentary horn has been found to be non-communicating.[5] In woman who previously delivered vaginally, this problem was difficult to be suspected.

Case Report

Mrs. LX 23 y G2 P1+0 was admitted to our hospital with 8 months pregnancy with absent fetal movements for 2 days. She had previous full term vaginal delivery at home. On examination - P/A Fundal height was 30 wks, oblique lie, liquor reduced, FHS absent, P/S - slight bleeding present, P/V- cervix closed, posterior, presenting part not felt. USG showed single intrauterine dead fetus of 30 weeks 4days with oblique lie, cephalic and oligohydroamnios. Induction failed and emergency LSCS was planned. On laparotomy 6-8 weeks size right unicornuate uterus with non-communicating left horn having 30-week pregnancy with bilateral small tubes. Still born female fetus was delivered out from left rudimentary pregnant horn. Placenta was thin and covering almost ¾ th of cavity & was adherent to uterine wall with no intervening deciduas. Umblical cord was short. Myometrium was very thin. Liquor was absent. On exploration, cavity of left horn was neither communicating with the cavity of right horn nor with the cervix, confirmed by P/V examination also. Right non pregnant horn was communicating with the cervix. Excision of the left horn along with tube done to prevent the risk of repeat pregnancy in this horn. HPE of tissue from right horn shows decidual cast with no chorionic villi.

Dr. Shuchi Gupta, Senior Consultant, Department of Obstetrics and Gynecology, Fatima Hospital, Lucknow.
Corresponding author email: drguptashuchi@gmail.com

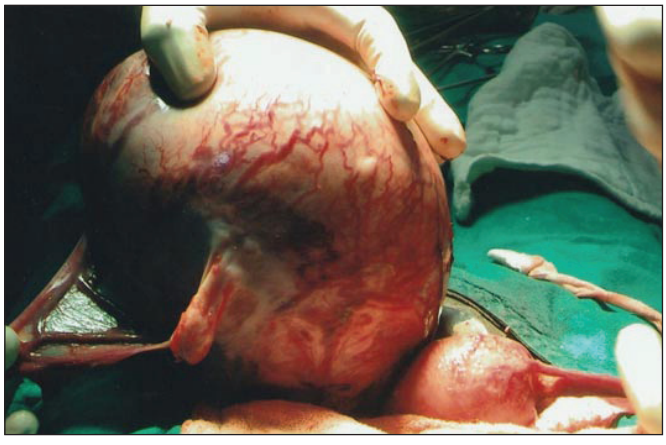

Fig. 1: Pregnancy in non-communicating rudimentary horn of unicornuate uterus.

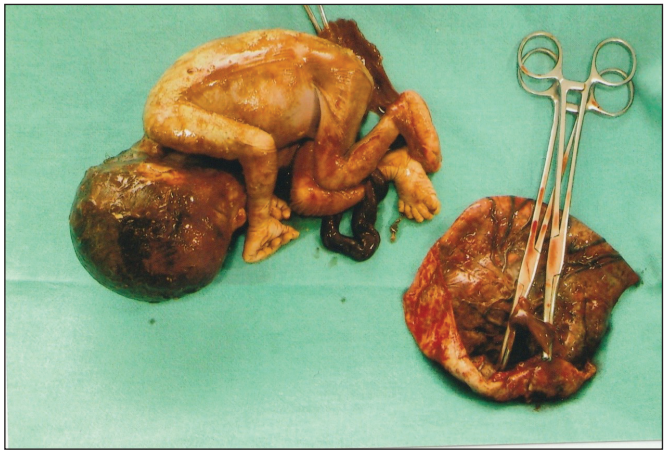

Fig. 2: Fetus and placenta.

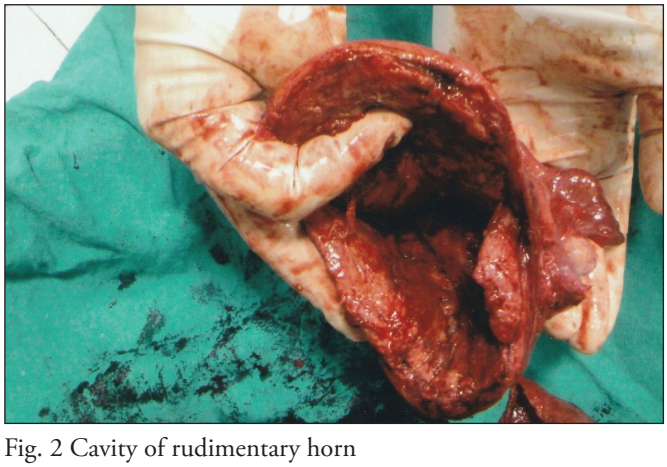

Fig. 2 Cavity of rudimentary horn

Her post operative period was uneventful. She was discharged on the 8th post-operative day and adviced follow-up after 6 weeks along with advice for IVP.

Discussion

Unicornuate uterus results from the failure in the development of one of the paramesonephric ducts either partially or completely. Partial development of one of the ducts gives rise to a rudimentary uterine horn. Pregnancy in a non-communicating rudimentary horn occurs probably by fertilization taking place due to transperitoneal migration of the sperm or the fertilized ovum, as evidenced in 8% of such cases, corpus luteum is found on the contralateral side to the rudimentary horn having the pregnancy.[1] Unicornuate uterus is most of the time an accidental finding which is usually asymptomatic until reproductive age. It presents as first trimester loss (24.3%), recurrent first trimester miscarriage (5–10%), a second trimester loss (25%), ectopic pregnancy (2.7%), or found during an infertility work-up.[2] In cases with successful pregnancy there is increased risk of preterm labour, abnormal fetal lie, intrauterine growth restriction preterm delivery (20.1%), intrauterine demise (10.5%) and (49.9%) preterm live birth up to 28–30 weeks of gestation.[6]

The usual and most dreaded complication is the massive intraperitoneal haemorrhage due to rupture of the horn which can be life threatening to the mother, resulting from a thin myometrium of the rudimentary horn, with the non-functional endometrium leading to adherent placenta as found in our case. The timing of rupture varies from 5 to 35 (avg 21.5) weeks depending on the horn musculature and its ability to hypertrophy and dilate. Few pregnancy cases with late or false diagnosis, which have progressed to 3rd trimester resulting in live births but among them neonatal survivability was only 6%.[7]

Early diagnosis is essential and challenging for the management as the consequences of rupture can cause significant mortality and morbidity to the mother & fetus. A careful ultrasound in the 1st trimester can diagnose pregnancy in the rudimentary horn. Tsafrir et al[8] has proposed set of criteria for diagnosis of pregnancy in the rudimentary horn: (1) A pseudo pattern of asymmetrical bicornuate uterus, (2) Absent visual continuity of tissue surrounding the gestational sac and the uterine cervix & (3) Presence of myometrial tissue surrounding the gestational sac. Sensitivity in detecting rudimentary horn uterus through ultrasound is only 30% and the condition is commonly missed.[9] Sensitivity decreases as the pregnancy advances, due to lack of definitive clinical criteria, in such cases MRI is very useful to confirm the diagnosis.

Interesting to note in our case was that induction had failed and incidental diagnosis of unicornuate uterus with pregnancy in non-communicating left rudimentary horn could be made peroperatively. The rudimentary horn was excised along with ipsilateral fallopian tube to reduce the risk of recurrent pregnancy in rudimentary horn in future and also to reduce the risk of dysmenorrhea or hematometra. As 31% of patients with mullerian anomalies will also have urinary tract anomalies with congenital absence of a kidney,[3] it is mandatory for this woman to have further assessment before attempting any future pregnancy.

In one report (Konar et al), three fetuses have been recovered on laparotomy from a ruptured gravid horn of a bicornuate uterus.[10]

Conclusion

Although pregnancy in a non-communicating rudimentary horn is uncommon, the diagnosis is always challenging.

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Case Report : Obstetrics

Pregnancy with Gaucher's Disease – A Case Report

Dr S Harinisri,[1] Dr Sanjib S Mondal,[2] Prof Tapan Kumar Maiti[3]

Introduction

Gaucher's disease is a lysosomal storage disorder caused by deficiency of beta glucosidase resulting in accumulation of its substrate glucosylceramide, leading to visceral, hematologic and skeletal manifestations. Here, we are presenting a case of pregnancy in a case of Gaucher's disease that was carried till term and to stress on the importance of multidisciplinary management in such cases.[1,2,3,4]

Case Report

A 27 year old female presented to NRSMCH outdoor as a diagnosed case of Gaucher's disease not on any treatment at 16 weeks gestation. She had no complaints at that time. She gave no history of illness till 23 years of age. She gave history of being on irregular treatment for hypothyroidism. On examination, her vitals were stable. Per abdomen examination revealed hepatomegaly 5cm below right costal margin and huge splenomegaly extending below umbilicus and uterus was just palpable. She was planned for a multidisciplinary management. Haematology opinion was taken and investigations were carried out. Bone marrow aspiration showed increased histiocytes with eccentric nuclei and crumpled tissue paper appearance of cytoplasm resembling Gaucher's cell with the impression being a storage disorder. Fibro-scan showed onset of fibrosis. Ultrasound whole abdomen showed a spleen of size 20.5cm and liver of size 16.6cm. Serum reports showed elevated ferritin of 287ng/ml, elevated folate of >23.60ng/ml and decreased Vitamin D of 18.74ng/ml. Mutation analysis of her husband confirmed that he was not a carrier. Patient received regular enzyme replacement therapy with glucocerebrosidase enzyme (Cerezyme) 60 IU/kg IV once in 2 weeks (Each 400 U Cerezyme enzyme vial reconstituted in 10.2ml sterile water and total 2640U reconstituted Cerezyme diluted in 100ml 0.9% NS and given IV over 2.5 hours. She was also on regular Injection VitB and VitD3 supplementation. Routine follow up with complete blood count, liver and renal function test was done (Table 1).

Regular antenatal check up was given. She was on levothyroxine 75microgram and carbohydrate restricted diet due to abnormal Thyroid stimulating hormone levels and deranged Oral glucose tolerance test. Anomaly scan was normal. Throughout her antenatal period, patient had no complaints and no bleeding manifestations. At 34 weeks gestation, patient was advised growth scan, and she brought the reports at 37 weeks gestation which revealed features suggestive of Foetal growth restriction with Cerebroplacental ratio<1. Patient was admitted and upon admission patient Non stress test was done which was non-reassuring (showed 5 decelerations in 20 minutes) and so was taken up for emergency

1. 3rd Year Post-Graduate Trainee, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata
2. RMO cum Clinical Tutor, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata
3. Professor, Department of Obstetrics and Gynaecology, NRS Medical College and Hospital, Kolkata

Corresponding author email: 10101993h@gmail.com

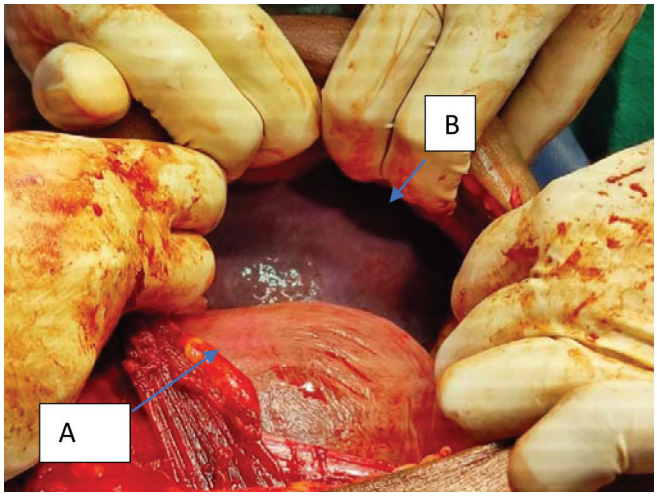

Figure 1: Intra operative findings; A – Uterus displaced downwards and to the right due to enlarged spleen; B – Spleen enlarged three times its normal size and seen just above uterus, displacing the bowel loops to the side

Lower segment caesarean section because of foetal distress. It was under general anaesthesia as her pre op platelet count was <80,000 (Table 1). Intra op findings revealed grossly enlarged spleen pushing uterus to the right with displaced bowel loops (Figure 1). A baby girl was delivered, weighing 2.010 kg.

Post op, patient was followed up in High dependency unit and she had no episodes of postpartum haemorrhage and her vitals were stable. As per anaesthesia advice, she received 1 whole blood, 1 Packed red cell, 2 Fresh frozen plasma and 2 platelets and patient's ECHO was done which was normal. As per haematology advice enzyme replacement therapy was stopped as she was lactating and they advised her for outdoor follow up 4 weeks after discharge. Both mother and baby were discharged in stable condition.

Discussion

Gaucher's disease is inherited as autosomal recessive. Type 1, usually presents with less severe symptoms, at more advanced age and is particularly amenable to enzyme replacement therapy. It's incidence is 1 in 50,000 – 1,00,000 worldwide.[1] In these patients reproductive age is commonly reached and childbearing is desired. These patients need appropriate prenatal diagnosis, counselling and careful obstetrical surveillance. Few data on the frequency of Gaucher disease during reproductive age and pregnancy can be found in literature[1] making our case unique.

Anaemia and thrombocytopenia may worsen during pregnancy, and excessive bleeding may complicate pregnancy, delivery and postpartum. Hepatosplenomegaly may interfere with normal foetal growth. Both these were seen in our patient and she also showed late onset FGR. Increased calcium demand in pregnancy increases the risk of bone crisis, osteopenia, osteonecrosis and fracture. There is also increased risk of infection and spontaneous abortion. The effects that pregnancy might have on the course of the disease is still unsolved.[2,4]

The diagnosis is performed by either determining the enzyme activity in peripheral blood leukocytes or through DNA based analysis.[3] Mutation sequencing in this patient showed Exon 9,c.1184>T variant and beta glucosidase assay was 0.67nmol/hr/ml. Enzyme replacement therapy is recommended during pregnancy to mitigate risks, especially bleeding. But it is yet to be shown whether its use has any adverse effect on fetal development. Cerezyme during pregnancy has demonstrated the ability to reduce the risk of miscarriage and this was used in our patient.

Table 1: Lab findings showing how the patient was monitored throughout pregnancy, worsening thrombocytopenia and persisting anemia, requiring blood replacement in the post operative period

Date	Hb (g/dl)	PCV (%)	Platelet (cells/ cumm)	TLC (cells/ cumm)	Urea (mg/dl)	Creatinine (mg/dl)	T.Bil/ Direct Bil (mg/dl)	SGPT (U/L)	SGOT (U/L)
16/12/20					18	0.7	1.4/0.7	14	20
14/1/21	8.1	25.2	48000	3300					
17/2/21	8.2	26.3	60000	2970					
22/4/21	9.3	31.1	90000	2600	15	0.7		32	28
1/6/21 (pre-op)	9.2	29.4	39000	4200	15	0.7	1.0/0.3	12	21
(at discharge)	10.3	31.5	60000	5600	16	0.5	1.2/0.6	16	17

The first antenatal appointment should include comprehensive examination of patient and multidisciplinary approach. In our patient too, this was followed. In addition to haematology opinion, surgeon's opinion was also taken who did not advice for any active surgical intervention from their side, an endocrinology opinion for her thyroid concerns and a general medicine opinion. Genetic counselling is recommended. Monitoring is adapted to the needs of individual patient based on disease status. Ferritin concentrations are often elevated as part of the sustained acute inflammatory response and does not indicate iron overload which was seen in this patient.[1]

During Caesarean section, it is advised to do a transverse Cohen incision or a Pfannensteil curved incision. Exploration of peritoneal cavity to be avoided as palpation of enlarged organs may precipitate bleeding. General anaesthesia is usually avoided due to risk of aspiration in the mother and neurological and respiratory depression in new-born but may be used if clotting parameter are altered in the mother or if she has severe spinal deformities.[1]

Conclusion

In our case there was no significant complication that occurred during pregnancy and delivery. Although thrombocytopenia was seen in antenatal period, patient showed no bleeding manifestations and required transfusion only in the postpartum period. If the disease is controlled, with proper therapy and monitoring, mothers are likely to have uncomplicated pregnancies and deliveries.

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Screening for Congenital Hypothyroidism

Dr. Meena Samant,[1] Dr. Juhi Sisodia,[2] Dr Divya Suman[3]

Congenital hypothyroidism (CH) is a treatable cause of intellectual disability in children, hence the importance of newborn screening for hypothyroidism.

First time newborn screening for a congenital disorder was done by Prof. Dr. Robert Guthrie in 1960, in USA for phenylketonuria.[1] Screening was done first time in 1965. This led to early diagnosis and treatment of hypothyroidism which in turn improved intellectual outcome in babies born with CH. This created a worldwide interest in screening for hypothyroidism. In India screening for CH was done first time at B. J. Wadia Hospital, Mumbai in 1982 using cord blood sample.[2]

Incidence of CH as obtained from neonatal thyroid screening programs ranges from 1:3000 to 1:4000 live births.[3] Cause of CH can be attributed to ethnicity, consanguinity, nutritional and environmental factors including iodine deficiency.

Types of CH based upon their cause can be classified into

1) Agenesis (22%-42%)
2) Ectopy (35%-42%)
3) Gland in place defect (24%-36%)

Clinical features of CH include mental retardation, developmental delay, delayed psychological development, congenital anomalies of heart, epilepsy, infantile cerebral palsy, hoarseness of voice, feeding problems, hypotonia, umbilical hernia, dry skin, jaundice. This causes a considerable financial and emotional burden on parents to manage such children. Hence there is a need of newborn screening program to detect congenital disorder as early as possible.

Wilson – Jungner in 1996 have given a criteria for selection of disorder in newborn screening program.[4] These are as follows:

1) The condition should be an important health problem.
2) Natural history of the condition should be well understood.
3) It should be detectable at an early age.
4) Treatment at an early stage should be beneficial.
5) Suitable test should be devised for early detection.
6) The test should be acceptable.
7) Intervals for repeating the test should be available.
8) Adequate health service provision should be made for the extra clinical workload resulting from screening.
9) The risk both physical and psychological should be less than the benefits.
10) The cost should be balanced against the benefits.

When to Screen

Primary TSH based CH screening is practical and cost effective. TSH surge in newborn starts 30 mins after birth, peaks at 24hrs and persists for 48-72 hrs. Thus cord blood is spared of this effect. So sample for screening should be taken from cord blood or postnatal day3 - day5 sample. This largely eliminates false positive result due to TSH surge.[5]

1. MD, DNB, MRCOG
2. DNB
3. MD (Obstetrics and Gynaecology)
Corresponding author email: meenasamant@rediffmail.com

The advantage of cord blood sample screening is that it is painless and can be done in early discharge cases. But its disadvantage is that it cannot be used for screening of other metabolic disorder which are dependent on feeding.

The advantage of postnatal sample is that it can be used to test other metabolic disorders dependent on feeding like galactosemia, phenylketonuria while its disadvantage is that it requires a longer stay in hospital or revisit to hospital if mother gets discharged early.

Method

Cord blood is taken in a plain vacutainer and sent to lab for testing. Alternatively postnatal sample can be collected and sent. Sample is collected by heel puncture on the plantar surface of the foot. Circles are drawn on filter paper which is directly applied to puncture area. Blood sample should completely fill the circles. It is then air dried for 4 hrs at room temperature and then sent to lab. The filter paper should be attached to a card which carries all the information about the baby.

Which test TSH or T4

An ideal screening test should be highly sensitive and specific. 3 main screening types used are

1) Primary T4 screening
2) Primary TSH testing
3) Simultaneous T4 and TSH measurement

Primary TSH based CH screening is more specific, practical and cost effective.[6] Main drawback of primary TSH screening is that it may miss infants with delayed rise of TSH which is seen in preterm babies due to immaturity of the hypothalamic-pitutary-thyroid axis. It also cannot detect cases of central CH, hypothyroxenemia and infants with thyroid TBG deficiency. Confirmatory test is done if TSH is greater than cut-off value.

Recommendations for recall testing are:[7]

1) TSH > 20 mIU/L is the cut-off for recall.
2) TSH > 40 mIU/L is recommended for defining screen positive cases for immediate recall for venous blood confirmatory test.
3) TSH from 20-40mIU/L should have a second TSH screen at 7-10 days of age. TSH in this range can often be false positive or there may be transient hypothyroidism. Transient CH is common in premature infants in iodine deficient areas.

Case of central CH can be identified by primary T4 testing. There is higher chance of false positive result in case of primary T4 testing. Also it fails to detect newborn with compensated form of CH (where T4 levels are normal and TSH levels are high) which is commonly seen in ectopic thyroid, the most common cause of CH.

Abnormal values i.e. T4<6.5 microgram/dl or TSH >20 mIU/L should always be confirmed by a repeat testing of venous blood sampling.[9] Age related TSH cut off (>34 mIU/L) is recommended for samples taken between 24-48 hrs of age.

Overall primary TSH assay is the recommended method for screening.

TSH Cut Off

For TSH levels from 20 – 40 mIU/L, a repeat filter paper sample or a repeat serum sample should be taken at 7-10 days of age. This will allow the neonatal factors causing a false positive result to settle down. If the repeat TSH is > 20 mIU/L for age < 2 wks or > 10 mIU/L for age >2 wks , confirmatory test by fT4 and TSH should be done.[8]

For TSH > 40 mIU/L, newborn should be immediately recalled. Venous blood sample is sent for fT4 and TSH testing by chemiluminescence or ELISA assay.

Treatment with levothyroxine is started in following condition –

1) fT4 < 12pmol/L or <1.1 ng/dl, irrespective of TSH value.
2) fT4 < 15 pmol/ L or < 1.17 ng/dl with venous TSH >20 mIU/L if age is <2wks and >10 mIU/L if age is >2 wks.
3) Normal T4/fT4 with persistently elevated TSH > 10 mIU/L at age > 3 wks.

In newborn with TSH >= 80 mIU/L, therapy should be started immediately without waiting for the result of confirmatory test. All the preterm and low birth weight infants should undergo screening at 48-72 hrs of age.

Imaging is done for assessing the severity and cause of CH. Imaging should never cause a delay in initiation

of treatment. It should just be used to assess the severity and cause of disease. The thyroid gland should be imaged using either scintigraphy or ultrasonography or both.

Treatment

Levothyroxine is the drug of choice for treatment of hypothyroidism. It should be started as soon as possible and not later than 2 wks after birth. The dose is adjusted between 10-15 microgram/kg per day.

Follow up test should be done after 1-2 wks of start of treatment. Subsequent evaluation should take place every 2 wks until TSH value reaches normal. Then every 1-3 months until the age of 1 year. Then every 2-4 months until the age of 3 years. Thereafter every 3-12 months until growth is complete.

Conclusion

Hence we see that there is a definite need to include newborn screening in the public health program like immunization program. There is a considerable financial and emotional burden on the parents of child who develops the complication of congenital hypothyroidism. Screening the newborn for congenital hypothyroidism is the most cost effective way to prevent it.

One such platform to introduce newborn screening in national program is Rashtriya Bal Swasthya Karyakram (RBSK) which is launched by Ministry of Health and Family Welfare, Government of India under National Rural Health Mission. It involves screening of children from birth to 18 years of age for 32 common health conditions. Children who screen positive are provided with further follow up and treatment. At present screening of Congenital Hypothyroidism is optional for states and UTs. They may include it depending on epidemiological situation and availability of testing.

Now that an initiative has been started, we are hopeful that screening for Congenital Hypothyroidism will soon be made mandatory in national health program.

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Clinical Article : Gynecology

Experience of Conducting Cervical Cancer Screening During Covid-19 Pandemic

Dr Indira Palo,[1] Dr Shweta Mohanty,[1] Dr Payal Keswarpu,[2] Dr Bharati Mishra[1]

Abstract

Objective: Cervical cancer contributes to a significant number of cancer cases every year specially in the developing countries. With COVID-19 pandemic screening services were paused for long periods. Innovative tests and healthcare delivery can help restart screening services to reach the goal set by WHO to eliminate cervical cancer.

Methods: We had planned for camp-based screening, however with the pandemic situation we had to change plans. Women visiting the outpatient department were sent to a dedicated facility for screening. Screening was done using Pap smear followed by ready to use VIA test Kit. Biopsy was done if VIA was positive or if indicated by PAP smear.

Results: Majority of the patients in our study were aged between 31-40 years of age. Vaginal discharge was the most common presenting complaint. In our study accuracy of VIA was at 91%.

Conclusion: We could offer screening services in our hospital maintaining social distance. The VIA Kit was helpful as we always had the necessary items to conduct the test faster. It reduced the exposure time in the hospital. In addition, adopting VIA as screening procedure helped us in reducing hospital visits for the women by providing instant results and decision on next steps.

Introduction

Every year a total of 569847 new cases of cervical cancer are detected globally with approximately 311365 deaths.[1] About 85% of these deaths occurred in low and middle-income countries.[2] In India there are about 75000 new cases detected annually.[3] The high mortality rate from cervical cancer globally can be reduced through a comprehensive approach that includes primary prevention, effective screening, and treatment programs.[2] Effective screening is a powerful tool to reduce the burden of mortality and morbidity of cervical cancer.

However, with the COVID 19 pandemic preventive services like cancer screening were severely affected. This situation has caused a significant delay in people seeking early care. On one hand, the healthcare system was stretched beyond its capacity and providing

1. Department of Obstetrics and Gynecology, M.K.C.G. Medical Hospital, Berhampur, Ganjam, Odisha
2. Westchester KnowledgeWorks, Umiya Business Park, Kadubeesanahalli, Bangalore, Karnataka

Corresponding author email: #

emergency care was also a challenge. On the other hand, having a gathering of people to conduct a camp-based screening was not possible for the fear of spread of infection.

While our initial plan was to restart cervical cancer screening services in our center through a camp-based approach, we had to defer this till the peak of pandemic was over. Even then it was not possible to organize camps in traditional sense with hundreds of women being examined in single day. Hence, we looked at organizing this in our hospital daily where all women in the age group of 30-65 years attending the OPD were sent for screening to a designated facility in the campus. The objective was to provide preventive care while maintaining social distance.

The present study aims to look towards one such simple measure of a readily available VIA Kit for screening of cervical cancer. The objective was to get an understanding on conducting cervical cancer screening in an outpatient center by a gynecologist using naked eye VIA method using a disposable readily available VIA Kit. In addition to this the secondary objective was to obtain feedback on VIA based kit testing in the existing healthcare infrastructure.

Materials & Methods

This was a cross sectional study involving a total of 100 women who were recruited in the out-patient unit of Gynecology department of medical college hospital. Institutional Ethical Committee approval was obtained for the study and patient consent was taken.[4] The recruited women were aged between 30 – 65 years of age with no previous history of hysterectomy and presented to the gynecology outpatient unit with various complaints.

Brief clinical history was obtained followed by naked eye inspection of cervix and vagina. Pap smear sample was collected which was followed by visual inspection with acetic acid using a disposable kit. The cerVIA™ Kit developed by Dalrada Health consisted of a bivalve speculum, gloves, saline solution for cleaning the secretions, cotton buds, pre-diluted 5% acetic acid, waterproof drape to be placed under the buttocks which are needed to perform VIA test on the cervix. Biopsy was done for all the cases which were either VIA positive or Pap smear showed any epithelial abnormality and as part of case management protocol of the institute.

All cases were examined by a post graduate student pursing education in Dept of Obstetrics and Gynecology. VIA results were documented as positive or negative and Pap smear results were documented as per Bethesda classification.

Results

A total of 100 women were screened with Pap smear and VIA using the cerVIA™ kit. The age wise distribution, the clinical symptoms and signs of the subjects are tabulated as follows in Table 1. Majority of the patients were aged between 31-40 years of age and the group between 60-65 years of age had the least no of cases.

Table 1: Characteristics of patients in the study

	Characteristic	Sample size
Age group	31-40	38
	41-50	31
	51-60	28
	61-65	3
SYMPTOMS		
Asymptomatic		55
Discharge per vaginum	Present	35
Abnormal bleeding	Present	7
Something coming out of vagina		3

Comparison between Pap and VIA

The comparison between Pap and VIA results is as follows in Table 2

Table 2: Comparison of VIA and Pap

	Pap Positive	Pap Negative
VIA Positive	9	9
VIA Negative	0	82

(VIA=visual inspection with acetic acid; PAP= Papanicolaou test)

All the 9 cases which were positive by both VIA and Papsmear were confirmed to be malignant by biopsy. VIA was 100% specific while sensitivity was 50% and accuracy was at 91%. Out of 100 cases 55 were asymptomatic, 35 cases white discharge and rest 7 had abnormal uterine bleeding as associated findings. Seven out of ten cases with abnormal uterine bleeding

showed acetowhite changes on VIA and Papsmear showed evidence for intraepithelial malignancy. Remaining three cases had prolapse of uterus.

Discussion

COVID-19 pandemic has affected more than 200 countries with more than 210 million cases worldwide. India reported 33 million cases of COVID 19. This pandemic bought about many changes in our society and healthcare system. Countries had complete lockdown to different levels of travel restrictions imposed.

Example of one such restriction was on 19th May 2020, due to the COVID-19 pandemic impact, the Government of India suspended the door-to-door screening of people above 30 years of age as part of the national program based on the risk associated oral cavity examination., the early detection of cancer services will primarily be restricted now to out-patient settings in primary, secondary, and tertiary care centers. till the peak of pandemic was over.

While our initial plan was to restart cervical cancer screening services in our center through a camp-based approach, we had to defer this based on GOI guideline. Our hospital being a tertiary care centers caters to a population around 100km radius. During lockdown period patients could not travel to seek hospitals services unless it was an emergency and when the restrictions were lifted there was fearing of contracting infection and loss of work wage which had just resumed. This showed a drastic decline in healthcare services availed.

According to a study done by hospitals participating in National Cancer Grid there was a more than 60% reduction in outpatients in India.[6] According to a study by Bakouny et al cancer screening rates declined by 60 to 80% compared to pre pandemic screening rates.[7] This was across all cancers like breast, cervix, lung, prostate, and colon. The consequences of this are likely to be witnessed in the coming years such as delayed diagnosis and advanced stage at presentation.

Several approaches are being discussed to provide continued services using innovative approaches. For example, teleconsultation services utilization increased by more than 500% during the pandemic, home test kits were developed to provide easy access to diagnostic services and care closer to home. One such innovation is the VIA Kit which enables quick examination compared to standard preparation time of getting all necessary instruments, disposables and reagents prepared. This will help in less time spent in a healthcare facility. In addition, there is no risk of cross infection between patients as all items are single use for one patient.

This study of Pap Smear and VIA was done in OPD setting in a tertiary care hospital, while VIA is usually done in peripheral health centers. In our study we used a ready-to use "kit" instead of preparing 5% acetic acid solution each time and procuring the other items like glove, speculum etc. separately. The VIA kit was very handy in conducting the test quickly and easily. The concentration of the acetic acid solution was fixed thereby reducing the variability of the results observed during the study. This could further lead to use of automated image analysis in the future.

The examination was conducted by resident doctors in training. Accuracy of VIA was 91% in our study and sensitivity was 50% which is reported in literature. However, we feel this can improve with sufficient training to distinguish positive and false positive case and with practice.

In summary we could offer screening services in our hospital without crowding and maintaining social distance. The VIA Kit was helpful as we always had the necessary items instead of waiting for availability of 5% acetic acid solution, sterilization etc. We had reduced work force during pandemic for routine healthcare services and this helped in saving time and resources. It reduced the exposure time of the women in the hospital. In addition, adopting VIA as screening procedure helped us in reducing hospital visits for the women by providing instant results and decision on next steps.

Acknowledgements

Dr Roja Ramani, Dept of Pharmacology, MKCG Medical College for guiding through the document preparation process.

Conflict of Interest

We declare no conflict of interest in the conduct of the study.

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ABBREVIATIONS

- WHO- World Health Organization
- VIA- Visual inspection with Acetic Acid
- OPD – Outpatient department

Case Report : Gynecology

Gossypiboma — A Surgeon's Nightmare

Dr Amit Basu,[1] Dr Deblina Kar[2]

Abstract

Surgical material most commonly made of cotton called gossypiboma are sometimes inadvertently left behind in the body at the time of surgical closure. The implications are grave for both the patient and the surgeons. There are some risk factors and conditions of surgery which contributes to the occurrence of this mishap and those conditions are potentially preventable. Through this case report we aim to re – empasize the phenomenon of gossypiboma and highlight its consequences and stress on its possible prevention.

Introduction

Gossypiboma or textiloma is used to denote specifically a mass of cotton material, usually gauze, sponges and towels left in the body cavity at the end of an operation. The word gossypiboma derives from two sources: the Latin word "gossypium" meaning textile or cotton, and the Swahili word "boma" meaning place of concealment.[1,2] Gossypiboma has virtually been reported after all kinds of operations including intrathoracic, orthopaedics, intraspinal, and neurological and even after breast surgeries, but most common after an abdominal or gynaecological surgery. Emergency surgery, sudden change in plan of an operation on the table, obesity, missing proper preoperative and post operative sponge count, profuse bleeding when large number of gauze and swabs are to be used to achieve haemostasis and provide a clear field view.

1. MBBS, DGO, FICOG, Additional Chief Health Director, Senior Consultant Obstetrician & Gynaecologist. B R Singh Hospital, Eastern Railways, Kolkata
2. MBBS, MS in OBG, FMAS

Corresponding author email: drdeblinakar@gmail.com

Prevalence of gossypiboma is difficult to document accurately due to under reporting of cases probably attributed to the fear of litigation.[7] Though both sexes are affected, women are at increased risk especially during obstetric and gynaecological operations.

Diagnosis of gossypiboma is often delayed because of its nonspecific presenting features and it may take even years to appear. Often it's associated with severe morbidity and rarely mortality.[7,8]

When a patient with history of previous surgery presents with complains in form of abdominal pains, nausea, vomiting, diarrhoea, abdominal mass, weight loss, malnutrition and features of intestinal obstruction or malabsorption syndrome ,a suspicion of gossipyboma should always be kept in mind.

Diagnosis is often by computerised tomography, though X-rays and ultrasound have also helped in detection where the cotton mass carries radio opaque marker.

Case Presentation

An emaciated lady, 33 yrs P3+0 (previous 2 Lscs) presented to our Gynaecology emergency of B R Singh Hospital, Kolkata with complaints fever and

profuse foul smelling discharge from a opening in her recent midline laparotomy incision And vaginal discharge as well.

Past History

She had elective caesarean section on 02/08/2019 at Suburban Nursing Home at (37 weeks+) and was discharged on the 7th day after stitch removal.

At the time of discharge she reported to her doctor that she was not feeling well and had felt a swelling in her abdomen. She returned after 2 days with several bouts of vomiting, fever and a swollen and tender lower abdominal scar of LSCS and copious purulent vaginal pus discharge. She was admitted and 2 units of whole blood was transfused by her obstetrician. The abdominal scar was explored at bedside to drain out pus and daily wound dressing were done for the next 10 days. She also gave history of some instrumental vaginal manipulation to drain out pus vaginally. No ultrasound or radiological imaging of abdomen was done at this point. When she did not recuperate with conservative management she was advised to attend a Govt Medical College in Kolkata. Meanwhile she visited another private practitioner who advised a CT scan of abdomen. She did a CT scan of abdomen & got admitted to a Medical College & Hospital in Kolkata on 30/08/2019. Her CT scan report (28/08/2019) showed a bulky uterus and ovaries were not visualised, lower abdomen showed a heterogeneous lesion with thick capsule with entrapped air within in the right side of the pelvis and right Lumbar region with surrounding mildly distended gut. Hence Gossypibioma could not be ruled out. Her attending doctors did an exploratory laparotomy with a midline incision under general anaesthesia on 30/08/2019.

Operative Notes

There was dense adhesion present in abdomen and pelvis. Adhesiolysis was done and several pus pockets were seen around intestine. Pus was drained and sent for culture sensitivity. Gut was examined & found to be normal. An abdominal drain was kept in situ. The midline longitudinal incision was sutured with PDS (p-dioxanone) & skin closed with Ethilon (Nylon sutures).

In her post op period she had nausea, weakness with no sense of general wellbeing. She could eat only small portions of normal diet daily with severe loss of appetite. Post operatively dressing of her previous LSCS wound was done daily until it healed (without secondary sutures).She was discharged on 24/09/2019 (25 days after Laparotomy). First post-operative follow-up on 10/10/19 was uneventful apart from the fact that she complained of generalised weakness. At her second follow-up on 07/11/19 she complained of pus discharge from laparotomy wound. On examination purulent discharge from two points of midline vertical laparotomy incision was noted. Wound dressing done, & pus sent for culture and she was prescribed oral antibiotics. At her third follow-up on 28/11/19 she was still found to have purulent discharge from wound and now from vagina as well (Almost 3 months after Laparotomy). The patient at this point had lost faith in the course of treatment and did not appear for further follow up. She had continued dressing of wound at home throughout the month of December 2019. She was prostrated, grossly emaciated and unable to perform any household chores to the point that her mother in law used to take care of the children/ new born. Finally on exacerbation of her symptoms and appearance of fever again she got herself admitted to B R Singh Hospital, Kolkata on 04/01/2020 at 7pm (Saturday).

On admission patient had a very poor general condition, was pale, weak and had a foul smell emanating from her. Her BMI was 18, pulse rate 86/min, blood pressure was 90/60. Investigations showed Hb 7.4 gm%, TC 9900, RFT and LFT were within normal range. On local examination of abdomen the transverse (LSCS) incision was healthy. There were two small openings in the longitudinal (Laparotomy) incision were from there was discharge of copious foul smelling pus, also copious discharge of pus from Vagina.

Dressing was done and pus was sent for culture sensitivity. We put her on Injection Meropenem & Injection Metronidazole along with other supportive medications. She was posted for ultrasound of whole abdomen the following day. Pus Culture sensitivity report shows heavy growth of E. coli highly sensitive to Meropenem. On the next day during bedside dressing of the wound, gaping of midline longitudinal incision was stretched with an artery forceps to facilitate drainage of pus. A whitish structure was seen & was gently pulled out by the artery forceps. The structure turned out to be an intact mop trying to make its

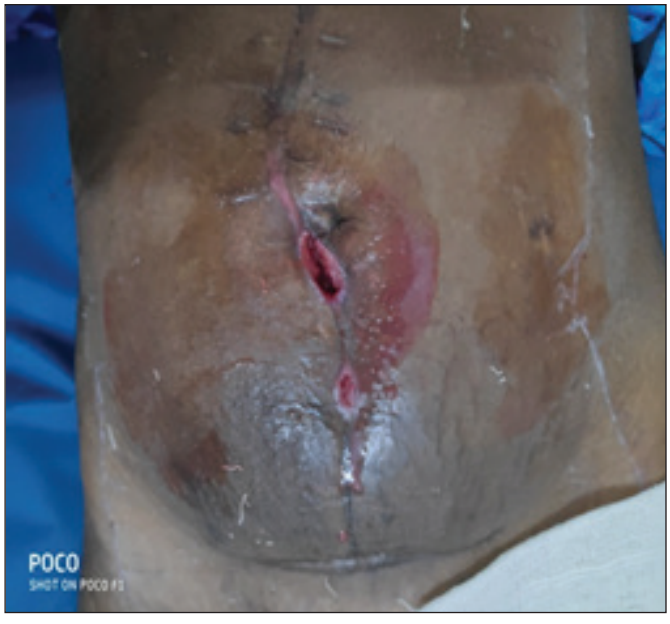

Fig 1: Small pus point was extended to facilitate drainage of pus

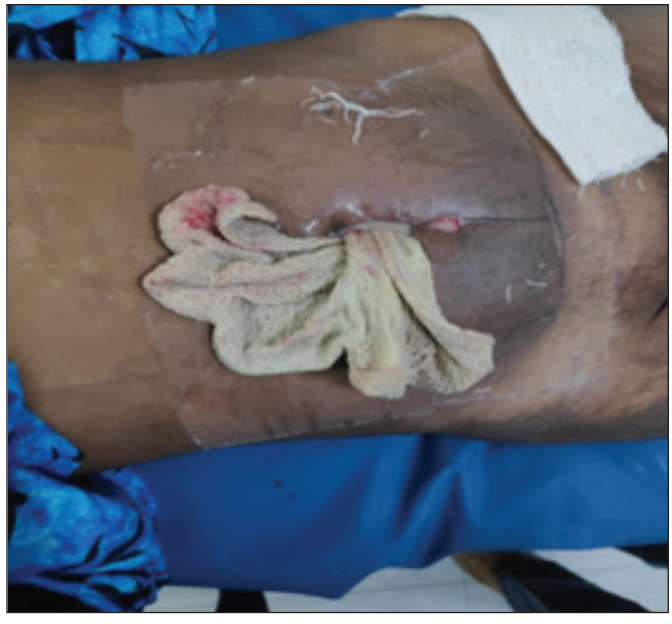

Fig 2: After a few dressings a piece of foreign body seen. Artery forceps was used to pull it out

Fig 3: Entire mop came out through the small rent

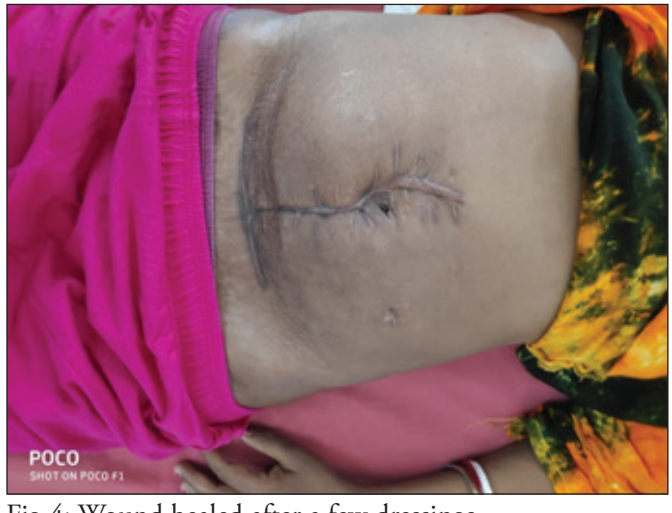

Fig 4: Wound healed after a few dressings

way out through the abdominal layers. The wound dressing done with betadine lotion and antibiotic ointment. Dressing was continued daily.

Further two units blood were transfused along with injection MVI, probiotics. Betadine vaginal pessary were given per vaginally daily. After 5 days the wound healed completely. She regained her appetite was visibly well. Before discharge her TC was 7400 & Hb 12.2gm%. She came for follow up after 4 weeks and was found to be doing very well.

Discussion

Gossypiboma is a condition rarely chanced upon.[1,6] This rarity may also reflect under reporting due to fear of litigation[4,7] The technical competence, skills and awareness of the surgeon and the theatre nursing staff are important. Emergency surgeries are at risk of Gossypiboma.

Gossypiboma has the potential to harm the patient profusely as a retained piece of cotton material evokes two different types of reaction in body. There is an exudative reaction which leads to the formation of abscesses, and there is also a fibrotic reaction which leads to adhesions and mass lesions.[3] This leads to severe morbidity as well as mortality. A history of previous surgery is mandatory for the diagnosis of gossypiboma at whatever site.[11] However the symptoms may be non-specific and mild and may be

overlooked for months or even for years.[8] The interval between originating surgery and manifestations may range from 11days to 28 yrs in a study by Garry and Agarwal.[7]

Diagnosis is by CT scan which shows brightly echogenic, well defined structure in a cystic mass.[9] Air bubbles and calcifications may be seen, leading to confusion with an abscess.[12,13] Diagnosis will be enhanced by plain X-rays only if the gauze or towel has radio-opaque marker. Ultrasonography, magnetic resonance imaging and upper gastrointestinal contrast radiographs have been valuable. Treatment for gossypiboma is surgical exploration, though always not required. Spontaneous migration of intraluminal retained gauzes can occur leading to expulsion of the mass during defecation . Intra uterine masses may be extruded through vagina, cervix. Laparoscopic methods may be used to remove left over intra-abdominal gauze.[5,15] The legal implications of gossypiboma are high. Gossypiboma may be misdiagnosed as a malignant tumour leading to unnecessary invasive investigations and extirpative surgery which may be disabling.[10]

Laparotomy in the midst of adhesions, abscesses, intestinal fistulae and intestinal obstruction can lead to morbidities and even death.[7]

Conclusion

Gossypiboma is a dangerous but easily preventable surgical condition. A properly ensured preoperative and post operative mop count should be done and doubly checked. Mops with radiopaque markers should be preferred. Avoid using small swab inside abdomen. When in don't hesitate getting a postoperative X-ray or CT scan.

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Swyer Syndrome in Siblings: A Rare Presentation

Sree Reshmi R J,[1] Pooja D C,[2] Dr Siddhartha Datta,[3] Dr Shamim Khandekar[4]

Abstract

Objective: Swyer syndrome in siblings is a rare entity. The risk for gonadal malignancies is more. Swyer syndrome running in families is a rare event.

Case Presentation: We are presenting this rare entity involving two affected siblings born to a nonconsanguineous couple. A 19 year old female presented with primary amenorrhea. The clinical findings, laboratory, radiological investigations and genetic study revealed Swyer syndrome. On follow up, we found that the elder sibling has similar presentation.

Conclusion: Early diagnosis and prophylactic gonadectomy plays an important role as chances of gonadoblastoma in these cases are high and increases with age. Psychological support and social rehabilitation should also be included in management.

Introduction

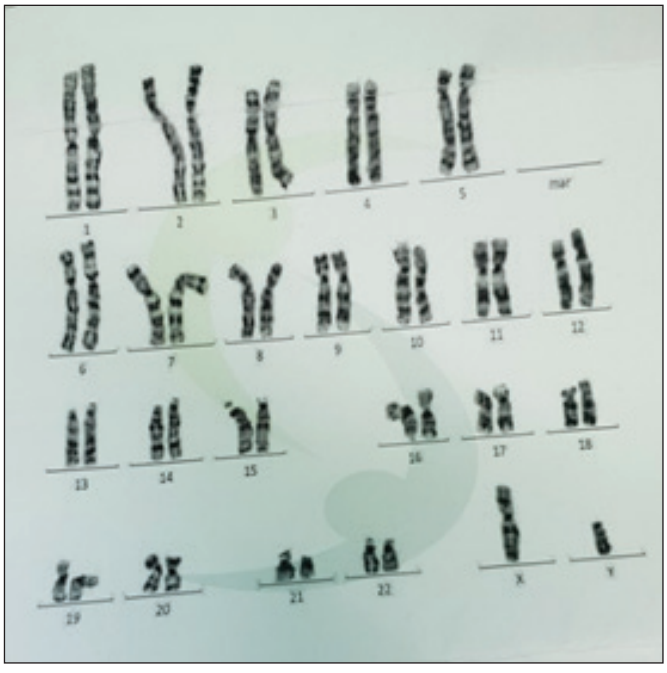
Swyer syndrome is a pure gonadal dysgenesis associated with a 46XY karyotype commonly presenting with primary amenorrhea in a phenotypic female. Inspite of having a Y chromosome, complete testicular dysgenesis is associated with a complete lack of androgenization of the external genitalia and persistent mullarian structures due to insufficient AMH production.[1] Swyer syndrome running in families is a rare event and few such scenarios were reported in the literature. It can have X linked, Y linked or autosomal inheritance.[2] Two affected siblings born to a non-consanguineous couple, presented with primary amenorrhea in the outpatient unit of North Bengal Medical College.

1. Post graduate trainee MS Obstetrics and Gynaecology, North Bengal Medical College, Darjeeling
2. Post graduate trainee MD Radiodiagnosis, North Bengal Medical College, Darjeeling
3. Professor Department of Obstetrics and Gynaecology, North Bengal Medical College, Darjeeling.
4. Assistant professor, Department of Obstretics and Gynaecology, North Bengal Medical College, Darjeeling.

Corresponding author email: reshmiraveendran940@gmail.com

Case Presentation

A 19 year old phenotypically female patient presented to our outpatient unit with primary amenorrhea. On examination her height was 157cm, weight 48 kg, arm span 159 cm and upper segment 82 cm. Tanner stage 2 breast development. Pubic hair is tanner stage 3. On examining the breast primary areola was found (B2 Stage). External genitalia was feminine, vestibule formed, vagina patent and lined by pink rugose mucosa. No clitoromegaly noticed. No palpable swelling in inguinal or labial region. Pelvic ultrasonography showed hypoplastic uterus (31mm*7mm*8mm) and absent ovaries. No ovaries were found even in magnetic resonance imaging. Laboratory findings were consistent with hypergonadotrophic hypogonadism (Table 1). Karyotyping was done and it showed 46XY genotypic pattern. Testosterone level was



Karyotyping showing 46XY pattern

within normal limit. Patient was started on combined estrogen and progesterone preparation and she started getting withdrawal bleeding every month thereafter. Serial ultrasound was done and it showed increase in the uterine size (Table 2).

Table 1

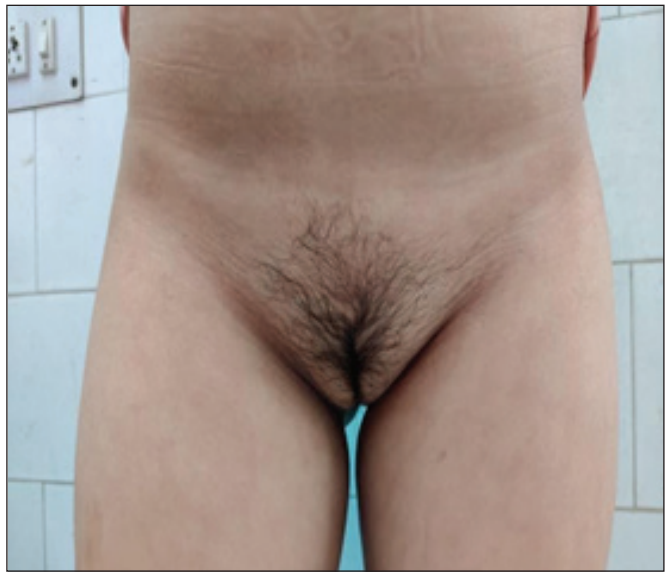
Test	level
FSH	18.7mIU/mi
LH	11.7mIU/ml
TSH	2.4mIU/ml
Testosterone	0.34ng/ml

Laparoscopy showed small uterus,cervix, normal tubes and streak ovaries. Pouch of douglas was normal. LDH was 103mIU/ml (normal 89-200), AFP, HCG were also normal. Since there was no evidence of any tumour gonadectomy was not done.

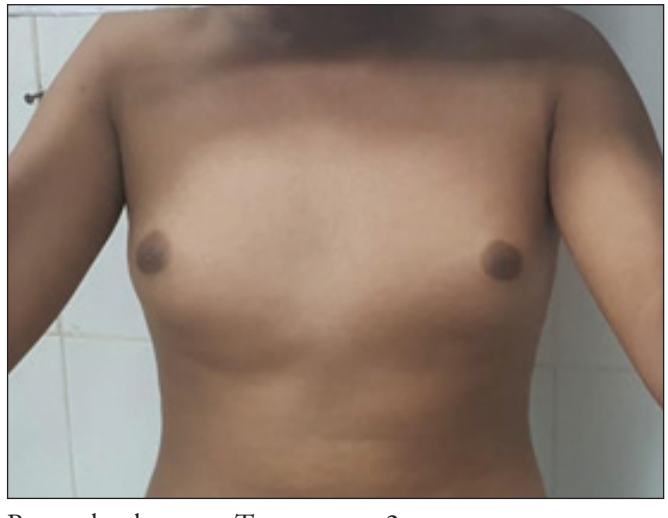
Table 2

Date	Size of uterus
14/11/2019	31x7x8mm
28/11/2019	40x20x10mm
31/1/2020	48x22x11mm
8/2/2020	52x23x12mm
21/3/2020	51x25x23mm
25/4/2020	53x27x30mm

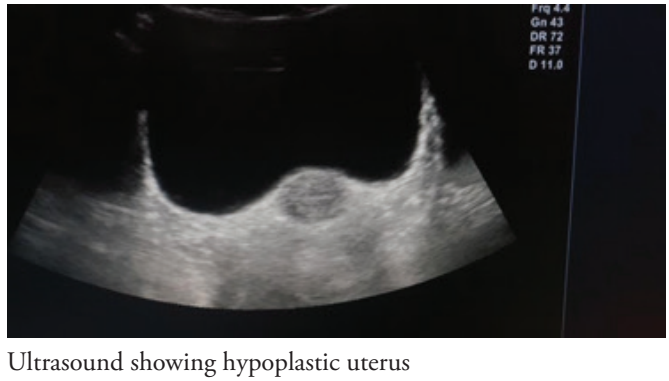
Her elder sister also had similar complaints and was investigated. She was found to have similar findings with internal genital organs, streak ovaries and 46XY



Pubic hair development Tanner stage 3

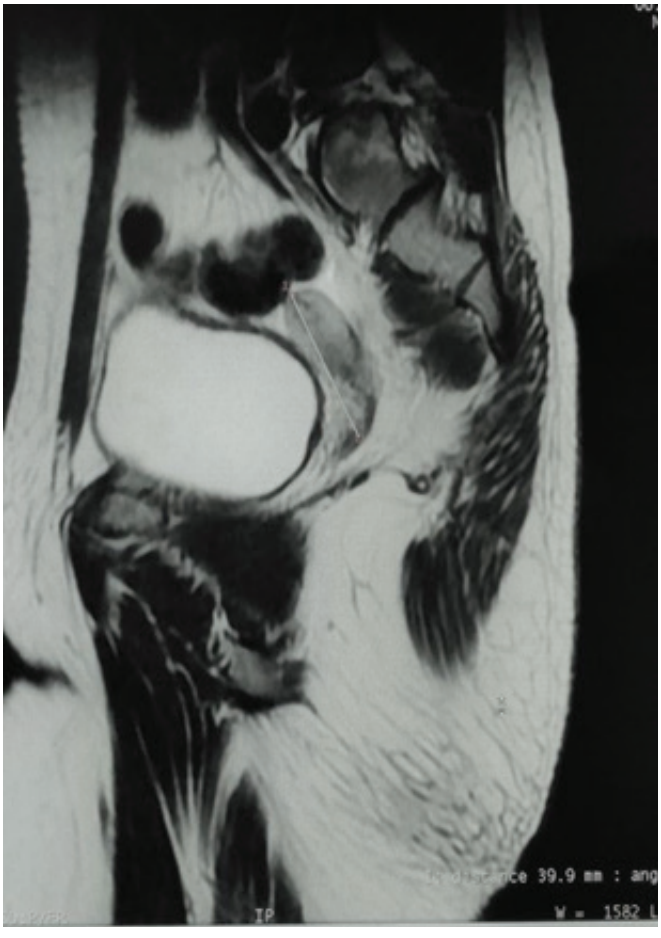


Breast development Tanner stage 2

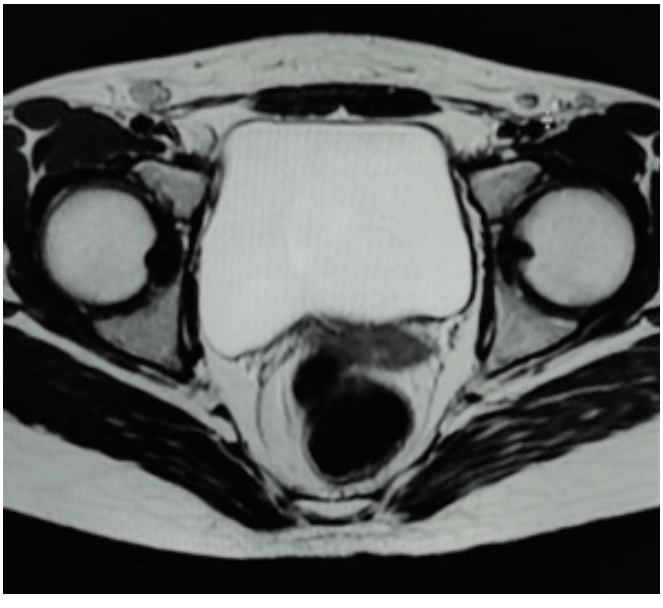


Ultrasound showing hypoplastic uterus

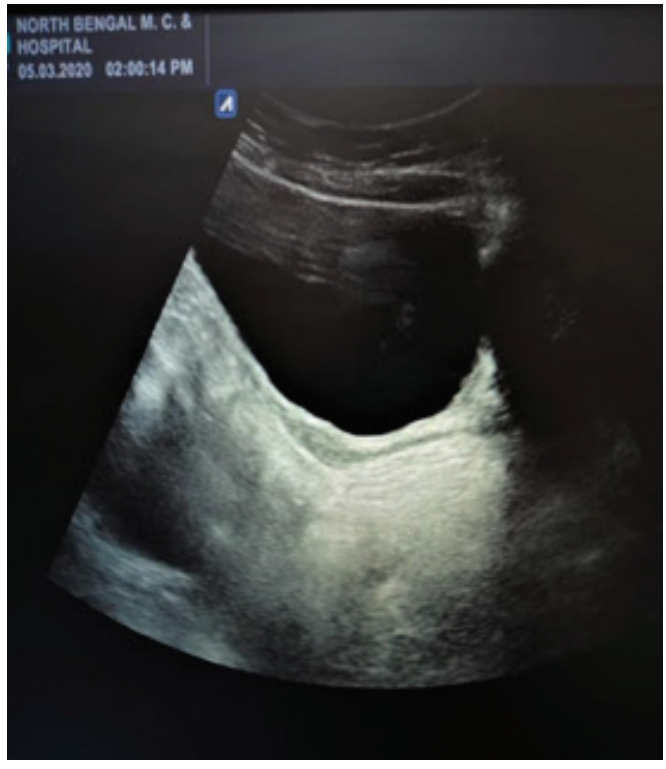
karyotype. LDH was 112, AFP & HCG were also normal. She did not have any swelling in the ovaries and hence prophylactic gonadectomy was not done. Being tea garden workers, both are alive and well at the time of reporting.



Sagittal section MRI showing hypoplastic uterus



Axial section MRI showing hypoplastic uterus



Sagittal section of ultrasound image showing hypoplastic uterus

Discussion

Phenotypic female with unambiguously female genitalia, normal mullerian structures (uterus, tubes, vagina), primary amenorrhoea and 46XY karyotype were first described by Swyer.[3] They have streak gonads. Mutations in SRY gene Yp11.[3] happen in 15-20%,[4] although mutations in desert hedgehog gene (DHH) 12q 13.1, NR5A1 9q33, CBX2 17q25, and 9p24.[3] have also been reported.[5] NROB1(DAX1) mutation involving Xp21.[3] may act antagonistically to SRY and result in sex reversal with adrenal hypoplasia.[6] NROB1 gene mutation is inherited in an X-linked pattern, hence from mother, while SRY mutation is exclusively inherited from father.[7] Paternal germ-line mosaicisms for SRY missense mutation may produce two populations of sperms with one normal and another carrying mutations leading to sex reversal in one sibling and XY normal in another.[8] DHH-autosomal recessive, require two mutated copies one from each parent, to develop Swyer syndrome. Consanguineous parents have a higher chance to overtly express a recessive condition.[9]

In a XY fetus, development of testes at 8 weeks requires action of SRY gene. With defective SRY, TDF is not formed, the indifferent gonads fail to differentiate into testes, and hence, no AMH or testosterone.[10] Wolffian ducts fail to develop and by default Mullerian system persist.[10] Because of lack of DHT, urogenital sinus fails to virilize, labio-scrotal swellings do not fuse resulting in female external genitalia.[11] Without

AMH, the Mullerian ducts develop into normal internal female genital organs (uterus, tubes, cervix, vagina).[12] Inability of the streak gonads to produce estrogens and androgens appropriately, secondary sex characters are stunted.[13] Limited adrenal androgen exposure result in sparse pubic hair.[13]

Hypergonadotropic primary amenorrhoea with female phenotype and 46XY karyotype, may also indicate androgen insensitivity syndrome, but presence of uterus, tubes, vagina, excludes the possibility.

Incidence for this condition varies between 1 in 20,000 to 1 in 80,000, and in sisters is rarer, indeed.[14]

The diagnosis is based on clinical, radiological and genetic evaluation. It should be differentiated from other causes of primary amenorrhea such as Turners syndrome, XY/XO mosaicism, XX gonadal gysgenesis and Kallmann syndrome.[15] Diagnosis of Turner syndrome can be ruled out by clinical examination. Patient with Turner syndrome may have short neck with webbed appearance, low hair line, low set ears and shield chest, karyotypically 45X0 with streak gonads.[16] Mixed gonadal dysgenesis may present with ambiguous genitalia, further radiological imaging may reveal one side testes and other side ovary.[17] Presentation of Kallmann syndrome is primary amenorrhea with anosmia.[18] Here the genotype is 46XX.

Swyer syndrome is associated with SRY gene mutations. Most of the mutations occur in the HMG box.[19]

Dysgenetic gonads have a tendency to develop into a pre malignant or malignant changes (gonadoblastoma, dysgerminoma, embryonal carcinoma) than streak gonads especially if they are intra-abdominal. The significant risk is 20 to 30%.[20] Gonadectomy even prophylactic, is indicated soon after the diagnosis. Extensive search for rudimentary gonads is needed.

Hormone replacement therapy can be given as in this case it showed improvement in uterine development.

The gender of upbringing in the reported cases was female. Although infertile, clients may become pregnant and carry to term with donor egg

Long term prognosis of Swyer syndrome is good once the gonadectomy is done. In patients with streak gonads regular follow up is advised.

Conclusion

The treatment of Swyer syndrome is multi-disciplinary. The patient is provided psychological support. This case is relevant as it is a rare and both sisters were affected. Early diagnosis would allow for conservative treatment which may help to reduce the emotional trauma and improve the patient outcome. Prophylactic gonadectomy is suggested to reduce the risk of gonadoblastoma.

Declaration of Patient Consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands the name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed. Financial Support and Sponsorship - Nil. Conflict of Interest - None stated.

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