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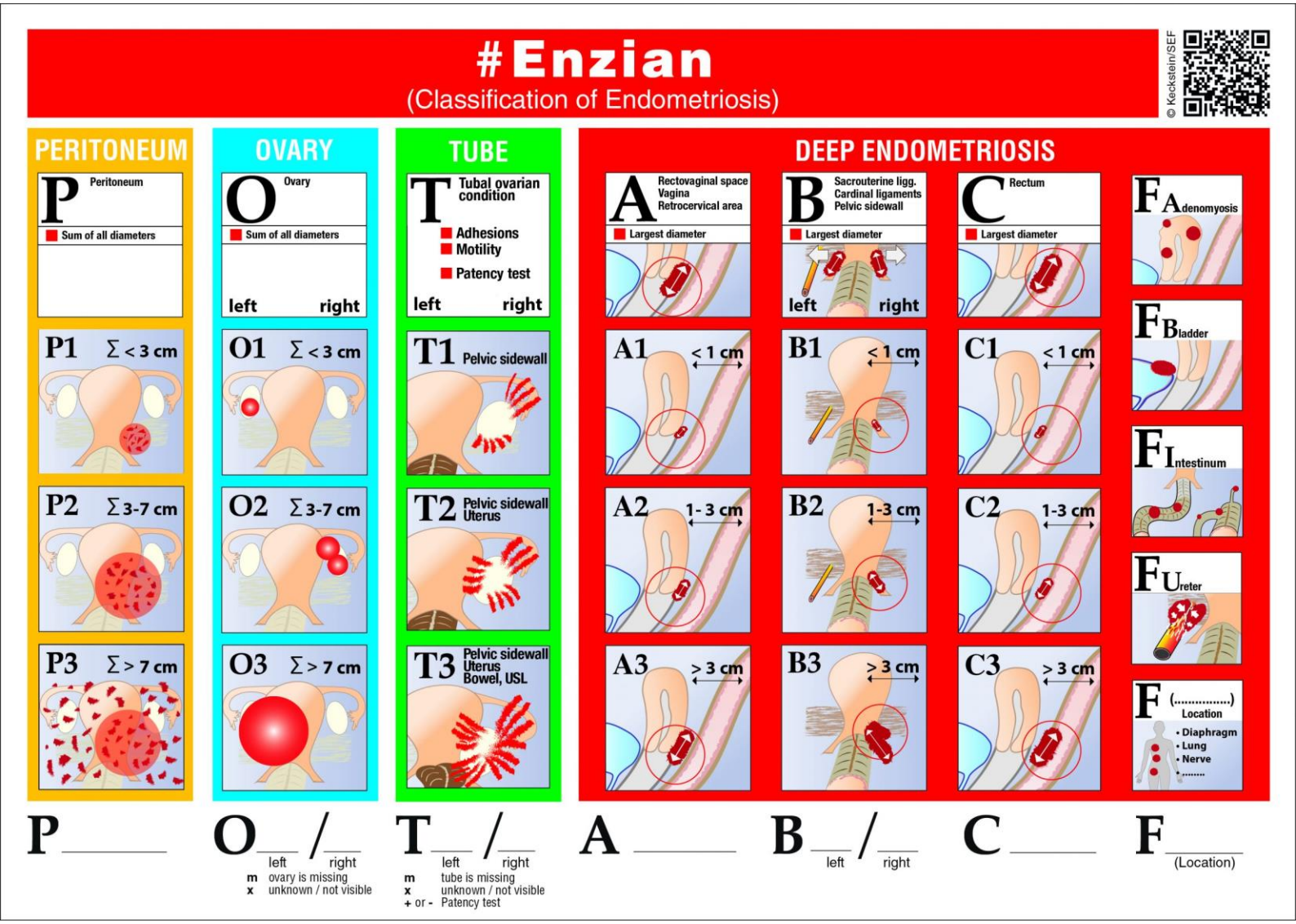

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The Titlepage shows the new extended **ENZIAN** Score for Endometriosis classification

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After a year of COVID19, the medical societies are fighting for funding.

On the one hand, the COVID19 crisis brought a so-called digitalization boost, but on the other hand it hit the traditional funding of the societies hard. At the beginning everyone wanted to show that they are still active and there were countless free teaching opportunities. The conference technology made it possible to register 500, 1000 or 5000 colleagues for conferences. The industry was also initially enthusiastic about the large number of participants at one event. With the latter, however, meanwhile a hangover mood did take over. The lack of personal contact at online events has apparently had a negative impact on the Return of Investment (ROI) for the industry. The large companies in particular complain about it, while smaller ones are happy about the greater exposure. With the new technology, live surgeries can also be financed by small companies and exciting workshops can be designed. For Societies like ISGE, however, all of this becomes a major challenge. The membership fees are adapted to the financial possibilities of our members and their countries of origin. The accreditation, another pillar of the financing, is severely restricted by COVID19, so that new course formats have to be found. For organizers like AAGL or ESGE, congresses are usually an additional source of income, for ISGE these only played a small role in the financing. Meanwhile, the competition on the web is so great that it is hardly possible to cover the costs. Numerous providers conduct courses online and issue certificates, which is ultimately very problematic. For accreditation and certification, colleagues should contact the professional associations that offer structured and Evidence-based education. ISGE is continuously working on an optimized training of our members, here we want to use the use of IT technologies in order to be able to offer excellent and affordable surgical education globally. Support us and become a member or a sponsor.

Sincerely

Guenter Noé

Incoming President ISGE 2021

THE INTERNATIONAL SOCIETY FOR GYNECOLOGIC ENDOSCOPY
PROMOTING VAGINAL SURGERY

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Improved reproductive outcomes in women with adenomyosis undergoing in vitro fertilisation following long term GnRH agonist downregulation: A case series

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Abstract

Background: Adenomyosis is a common condition that is often associated with poor reproductive outcome. Repeated implantation failure in in vitro fertilization (IVF) cycles can result from impaired implantation caused by adenomyosis. The evidence regarding the role of long-term suppression (LTS) with gonadotropin-releasing hormone agonists (GNRHa) prior to embryo transfer (ET) in cases of adenomyosis is limited and conflicting.
Aim: The aim of this study was to assess the efficacy of long-term GNRHa therapy on livebirth rates in women with adenomyosis and infertility.
Design: The following case series includes 15 women with infertility and known adenomyosis undergoing in vitro fertilisation (IVF) that underwent at least three months downregulation with GNRHa prior to ET. Outcomes were compared to previous cycles performed without LTS.
Results: LTS with GNRHa was given in 16 cases (94.1%) prior to ET. In one case (5.9%) 6 months suppression was given. The majority of these patients had previous unsuccessful IVF cycles prior to LTS protocol with GnHRa. 17 embryo transfers (16 frozen, 1 fresh from donor) following LTS protocol resulted in 10 liveborn deliveries at term (58.8%) vs. a live birth rate of only 7.7% per ET without LTS (26 embryo transfers resulting in 2 live births; P<0.0001). Only three women who underwent long term GnRHa downregulation had no successful embryo transfers.
Conclusion: Our successful outcomes support the use of LTS with GnRHa to improve reproductive outcomes.

Keywords: Adenomyosis, infertility, in vitro fertilisation, gonadotropin releasing hormone agonist

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Introduction

Adenomyosis is a benign estrogen dependent disease whereby ectopic endometrial glands or stroma are found within the myometrium, surrounded by myometrial smooth muscle cell hyperplasia and hypertrophy [1-3]. It can be diffuse or focal depending on extent of uterine spread [4, 5]. Women with adenomyosis typically present with dysmenorrhea, heavy menstrual bleeding or infertility, however a large proportion can be asymptomatic [3]. Therefore, true prevalence is unknown, with large variations between 5-70% cited within the literature [3, 6, 7]. Previously thought to be a disease associated with older multiparous women, delayed fertility and advancements in imaging techniques have led to an increased diagnosis in younger women of reproductive age being investigated for infertility [8]. Adenomyosis has been associated with poor reproductive prognoses [9, 10]. Multiple factors have been implicated including impaired implantation and utero-tubal disruption [11, 12]. Of particular interest is the impact of poor endometrial receptivity on implantation, which has been suggested to be more significant than embryo quality [13]. Increasing evidence is emerging to support the role of long-term GnRHa downregulation in improved reproductive outcomes, especially when used in conjunction with IVF or intracytoplasmic sperm injection [7, 14-16]. The following case series details successful pregnancy outcomes following GnRHa down regulation prior to embryo transfer (ET), further supporting the existing evidence and theorises its main mechanism of action is by improving endometrial receptivity.

Materials and methods

This is a retrospective case series that includes 15 patients diagnosed with adenomyosis and treated with long acting GnRHa prior to their IVF treatment. These patients were chosen from a population of patients attending a private IVF clinic. All women included required an ultrasound diagnosis of adenomyosis. Patients were treated with at least 3 months of Goserelin Acetate Implant (Zoladex®) as part of the IVF protocol prior to ET. The study was reviewed by the Epworth Healthcare ethical review board and registered as a quality assurance study. Data was retrospectively collated from patient files and included patient demographics, previous obstetric history, prior fertility treatment and outcomes as well as significant past medical history. Data were analysed to assess the effect of long-term GnRHa downregulation on reproductive outcomes. Other factors impacting fertility were also recorded for each patient (See Table 1).

Data analysis was performed using SPSS Statistics, version 11. Descriptive characteristics of data are presented as median and interquartile range. Test of statistical significance for categorical variables was done using Pearson's chi-square test and T-Test for non-categorical variables. Statistical significance was set at a p value <0.05.

Results

received long acting GnRHa down regulation were reviewed. The average age of the patients was 37.5 years (range 28-48, SD 6.5). Most of the patients had concurrent diagnosis of endometriosis (13 patients, 86.7%) that was also treated prior to the commencement of the current long-acting suppression (LTS) protocol, none of the patients had surgery immediately prior to the commencement of the LTS protocol. All of the endometriosis surgeries that were performed occurred prior to cycles with regular ET without LTS. Diffuse adenomyosis was

reported in 14 of the women (93.3%) while one patient had focal adenomyosis. All patients were diagnosed using ultrasound scans that were performed by a certified obstetrics and gynecology ultrasound specialists. Further data regarding patient characteristics can be found in Table 1.

Three months of long-term suppression with Zoladex was given in 16 cases (94.1%) prior to ET (Table 2). In one case (5.9%) 6 months suppression was given due to significant abnormal uterine bleeding. Frozen embryos were transferred in 16 (94.1%) patients after long term suppression (LTS) and a fresh embryo was transferred in one case (5.9%). Embryo transfers without LTS protocol included 15(57.7%) frozen embryos and 11 (42.3%) fresh embryos.

The majority of these patients had previous unsuccessful IVF cycles prior to LTS protocol with GnRHa down regulation. All patients had a single embryo transfer in all cycles. 17 embryo transfers (16 frozen, 1 fresh from donor) following LTS protocol resulted in 10 liveborn deliveries at term (58.8%) vs. a live birth rate of only 7.7% per ET without long term down regulation (26 embryo transfers resulting in 2 live births), this was statistically significant (P<0.0001). Only three women who underwent long term GnRHa downregulation had no successful embryo transfers. Total pregnancy rates were significantly better as well in the long-term suppression group (58.8% vs 19.2%, P=0.008 excluding chemical pregnancies, or 64.7% vs 26.9%, P=0.014 including chemical pregnancies).

Although most patients in this study had concurrent endometriosis, none of the LTS ET cycles were performed immediately following surgery for endometriosis. Furthermore, even though 9 patients underwent excision of endometriosis prior to other cycles, none of the patients had a live birth following the surgery (0%) while 6 of them (66.7%) had a live birth following LTS protocol. This superiority of LTS in cases of adenomyosis compared with endometriosis surgery was statistically significant (p=0.003)

Discussion

The pathogenesis of adenomyosis is not completely understood.[2] Adenomyosis is associated with a higher prevalence of recurrent pregnancy loss, failed assisted-reproductive treatment (ART) and poorer IVF reproductive outcomes [16-19]. Adenomyosis is suspected to impact fertility through a range of molecular mechanisms resulting in recurrent implantation failure (Figure 1). Adenomyosis related junctional zone disturbance causes dysperistalsis, impairing sperm transport and blastocyst implantation [12, 20, 21]. Hypoestrogenism, found in women with adenomyosis, perpetuates this dysperistalsis [22].

Impaired endometrial receptivity is also associated with implantation failure [23]. Adenomyosis has been associated with reduced endometrial receptivity markers, such as integrin ß3 and leukaemia-inhibiting factor, HOXA10 and HOXA11, which play critical roles in implantation as well as endometrial growth, differentiation and decidualisation[24, 25]. Pinopodes are morphological markers of endometrial receptivity seen on the endometrial surface at the time of implantation [25]. Decreased numbers and poorly formed pinopodes have been seen in human and mice studies with adenomyosis [25-27].

There however remains conflicting reviews in current literature regarding the impact of adenomyosis on infertility, with several authors concluding that cases with concurrent

endometriosis confound and limit available evidence [16, 22, 28]. A 2014 meta-analysis found a 28% reduction in likelihood of clinical pregnancy in women with adenomyosis undergoing IVF or ICSI and suggests screening for adenomyosis prior to ART [15]. A retrospective cohort study of 213 women showed a significantly decreased rate of viable clinical pregnancies in women with adenomyosis undergoing IVF with GnRH antagonist for ovarian stimulation [29]. Furthermore, Dueholm's review found an overall reduction in pregnancy rate with adenomyosis (RR 0.63) and an increased risk of miscarriage [20]. In contrast, a case-control retrospective study of 49 women with adenomyosis having oocyte donation showed no significant differences in implantation rates [30]. Benaglia's [31] prospective cohort study also showed no significant difference in clinical or ongoing pregnancy rates in women with adenomyosis undergoing IVF. Similar findings have been found in other studies [32, 33].

Overall, the heterogeneity of studies investigating adenomyosis and infertility make it difficult to compare results. There are no high quality studies, with those published being limited by their retrospective nature, differing diagnostic criteria, small sample sizes, differing ages and concurrent endometriosis [7, 20, 34, 35]. There is also a lack of studies looking at the effect of adenomyosis on natural conception [20].

There are no current guidelines specific to the management of adenomyosis, especially for those seeking fertility assistance [36]. Medical treatment is limited in patients that wish to conceive and hysterectomy is not a valid option for these patients.[6] Growing desire for fertility preservation has seen an increase in cytoreductive surgeries performed and development of safer surgical techniques [37]. Whilst surgical excision of adenomyosis improves symptoms and fertility outcomes they are not without risk, including uterine adhesions and pregnancy complications such as uterine rupture [37, 38].

It is thought that GnRHa improves fertility in women with adenomyosis through reduction in hyper estrogenic states both indirectly through to hypothalamic-pituitary axis and directly at the level of the tissues by normalising the endometrial over-expression of aromatase cytochrome P450 that occurs in adenomyosis [39]. GnRHa has also been shown to have an antiproliferative and apoptotic effect on endometrial cells in vitro and Khan[40] identified suppressed pathologic lesions in women with adenomyosis. GnRHa therapy may improve endometrial receptivity by reducing the extent of basal endometrium dislocation that occurs in adenomyosis [41]. A retrospective cohort study by Bao et al[26] also found an increase in endometrial receptivity markers following long-acting GnRHa protocol and significant increase in clinical pregnancy rates in women with decreased ovarian reserve.

Over the last decade, four retrospective cohort studies have evaluated the use of GnRHa downregulation on fertility and IVF outcomes in adenomyosis[41-44]. Niu [41] compared clinical pregnancy rates in the first IVF cycle post treatment with GnRHa and HRT versus HRT alone. They found that one month of GnRHa downregulation doubled implantation and ongoing clinical pregnancy rates. Small case series further support the benefit of GnRHa downregulation prior to IVF [45, 46]. However following 2-3 months of monthly goserelin, Park [44] found no statistical difference in clinical pregnancy rates. A small RCT found no significant

difference in outcomes[47]. Data regarding the effect of long term down regulation with GnRHa is limited [48].

We believe that adenomyosis can severely impair fertility and that our results further support this. Our cases were chosen for intervention following repeated failed IVF cycles, thought to be a result of their severe adenomyosis. Almost all of these patients had recurrent implantation failures prior to treatment with long term GnRHa downregulation. Our results show significant highly successful fertility outcomes post three months of GnRHa therapy prior to embryo transfer supporting its use in management of infertility. This differs from other evidence where successful cases with GnRHa downregulation were seen only following cytoreductive surgery [49-51]. Use of GnRHa therapy alone would likely be associated with less risks than in combination with surgical management, however whilst the women in our case series tolerated GnRHa therapy well (all successfully completing a minimum of three-months treatment), the side effects from a hypooestrogenic state may not be tolerated by all. Tolerance may be regime dependant and currently there remain large discrepancies in GnRHa downregulation regimes [41, 42, 44]. Our study has several limitations being retrospective in nature with a small sample size and high concurrent endometriosis rate. There was also selection bias as most patients had recurrent implantation failures in multiple cycles, this is also accentuated as the patients are their own controls. However, this significantly high success rate in a population that had very poor outcomes prior to this treatment is very promising. Acknowledging that outcomes will always be better after introduction of an intervention, we believe it supports further research to explore the impact of adenomyosis and GnRHa on implantation.

1.5 CONCLUSION

Our cases show promising results and support the use of long term GnRHa downregulation prior to frozen embryo transfer. The increased rate of pregnancy seen post GnRHa downregulation in this cohort may be attributed to increased endometrial receptivity promoted by GnRHa.

Furthermore, whilst we believe that GnRHa downregulation is a promising therapy for women with adenomyosis and infertility, further evidence is still required to assess its impact on pregnancy rates.

Case	Age	Time of infertility	Ovarian Reserve	Diffuse or focal adenomyosis	Wall thickness: Anterior (A) Posterior (P)	Other Factors Impacting Recurrent Implantation Failure
1	32	18 months	Normal AMH	Diffuse	A: 12mm P: 16mm	Excised stage 3 endometriosis Patent tubes Regular periods
2	28	24 months	Normal AMH	Diffuse	A: 10mm P: 16mm	Endometriosis: 35mm bowel lesion and 30mm endometrioma requiring excision
3	39	24 months	Normal AMH	Diffuse	A: 17mm P: 11mm	Mild endometriosis
4	39	24 months	Normal AMH	Diffuse	A:13mm P: 15mm	No
5	35	25 months	Normal AMH	Diffuse	A: 8mm P: 11mm	Excised moderate endometriosis
6	37	12 months	Normal AMH	Diffuse	A:13mm P:14mm	Multiple excisions of endometriosis
7	35	18 months Secondary infertility	Normal AMH	Diffuse	A: 7mm P: 10mm	Inc BMI – gastric sleeve Hydrosalpinx (required unilateral salpingo-ophorectomy) Endometriosis
8	47	No infertility No partner	Donor oocytes	Diffuse	A: 8mm P: 15mm	Hydrosalpinx (required unilateral salpingectomy) Excised endometriosis
9	46	4 years Secondary infertility	Normal AMH	Diffuse	A:11mm P:14mm	Excised extensive endometriosis
10	39	12 months Secondary infertility	Normal AMH	Diffuse	A:11mm P:12mm	Rheumatoid Arthritis Multiple excisions of endometriosis Myomectomy (10cm fibroid)
11	30	12 months	Normal AMH	Diffuse	A: 7mm P: 14mm	Excised stage 3 endometriosis Positive testing for NKC
12	30	14 months	Normal AMH	Focal	P: 4cm adenomyoma Posterior uterus above level of cervix	Factor V Leiden Moderate endometriosis
13	32	16 months	Normal AMH	Diffuse	A:18mm with 3cm full thickness bladder nodule P: 12mm	Endometriosis
14	48	6 years	Normal AMH	Diffuse	A: 18mm P: 18mm	Endometriosis
15	36	24 months	Normal AMH	Diffuse	624.3ml in total (too gross to measure) with repeat measurement post 6 months of Goserelin Acetate Implant – volume shrunk to 175 ml	Laparotomy for possible fibroid, diagnosed with adenomyosis. Goserelin Acetate Implant for 5 years with add back therapy, ceased 5 years prior to IVF

Table 1. Patient demographics

Case	Past Obstetric History	Number of Eggs Retrieved	Number of embryos	Embryo Quality	Embryo Transfer	Embryo Transfer Regime	GnRH agonist regime prior to ET	Outcome
1	Nil	12	4	Day 5	1st	No PGS Fresh ET	-	No pregnancy
					2nd	No PGS Frozen ET	-	No pregnancy
					3rd	No PGS Frozen ET	-	No pregnancy
					4th	No PGS Frozen ET	3 months Goserelin Acetate Implant	Vaginal delivery Liveborn at term
2	Nil	10	2	Day 5	1st	No PGS Fresh ET	-	No pregnancy
					2nd	No PGS Frozen ET	-	No pregnancy
		10	2	Day 5	3rd	No PGS Fresh ET	-	No pregnancy
					4th	No PGS Frozen ET	-	No pregnancy
		7	2	Day 5	5th	No PGS Fresh ET	-	Vaginal delivery Liveborn at term
					6th 12 months later	No PGS Frozen ET	3 months Goserelin Acetate Implant	Vaginal delivery Liveborn at term
3	Nil			Day 5	1st	No PGS Frozen ET	3 months Goserelin Acetate Implant	Caesarean Liveborn at term
4	Nil	12	8	Day 5	1st	PGS on 5 embryos – 3 euploid Frozen ET	3 months Goserelin Acetate Implant	Caesarean Liveborn at term
5	Nil	10	5	Day 5	1st	No PGS	-	No pregnancy

						Frozen ET		
					2nd	No PGS Frozen ET	-	No pregnancy
		10	6	Day 5	3rd	No PGS Frozen ET	3 months Goserelin Acetate Implant	Vaginal delivery Liveborn at term
6	Nil	11	6	Day 5	1st	No PGS Fresh ET	-	No pregnancy
					2nd	No PGS Frozen ET	-	No pregnancy
		21	9	Day 5	3rd	PGS on 3 embryos – 2 euploid Frozen ET	3 months Goserelin Acetate Implant	Vaginal delivery Liveborn at term
7	1 NVD at term	14	13	Day 5	1st	No PGS Fresh ET	-	Biochemical pregnancy
					2nd	PGS on 9 embryos – 5 euploid Frozen ET	3 months Goserelin Acetate Implant	No pregnancy
					3rd	PGS as above Frozen ET	3 months Goserelin Acetate Implant	No pregnancy
8	Nil	-	-	-	1st	No PGS Fresh ET (from donor)	3 months Goserelin Acetate Implant	Caesarean Liveborn at term
9	1 NVD at term	-	-	-	1st	No PGS Frozen ET	3 months Goserelin Acetate Implant	Biochemical pregnancy
10	1 NVD at term	8	6	Day 5	1st	No PGS Fresh ET	-	No pregnancy
					2nd	No PGS	-	8 weeks miscarriage

						Fresh ET		
		6	3	Day 5 Poor quality not used	-	-	-	-
		7	4	Day 5	3rd	PGS on 1 embryo – euploid Frozen ET	-	8 weeks miscarriage
		19	9	Day 5	4th	PGS on 4 embryos - 1 euploid Frozen ET	3 months Goserelin Acetate Implant	No pregnancy
11	Nil	12	7	Day 5	1st	No PGS Frozen ET	-	8 weeks miscarriage
					2nd	No PGS Frozen ET	-	No pregnancy *Nb went to another unit following this and had 10 unsuccessful ETs
		-	2 left	-	3rd with this unit	PGS on 2 embryos – 2 euploid Frozen ET	3 months Goserelin Acetate Implant	No pregnancy
					4th with this unit	PGS as above Frozen ET	3 months Goserelin Acetate Implant	Vaginal delivery Liveborn at term
12	Nil	8	6	Day 5	1st	No PGS Fresh ET	-	No pregnancy
					2nd	No PGS Frozen ET *FVL heterozygous diagnosed – commenced enoxaparin	-	No pregnancy

						from time of ET		
					3rd	No PGS Frozen ET	-	No pregnancy
					4th	No PGS Frozen ET	-	Vaginal delivery Liveborn at term
		8	4	Day 5 Only 2 good embryos	1st	No PGS Fresh ET	-	Biochemical pregnancy
					2nd	No PGS Frozen ET	-	No pregnancy
		12	5	Day 5 Only 4 good embryos	3rd	No PGS Fresh ET	-	No pregnancy
					4th	No PGS Frozen ET	-	No pregnancy
					5th	No PGS Frozen ET	3 months Goserelin Acetate Implant	No pregnancy
13	Nil		6		1st	PGS Frozen ET	3 months Goserelin Acetate Implant	Caesarean Liveborn at term
14	Nil		1		1st	Frozen ET	3 months Goserelin Acetate Implant	Not pregnant
15	Nil		4		1st	PGS Frozen ET	6 months Goserelin Acetate Implant	Caesarean Liveborn at term

Table 2. Summary of case studies following GnRH agonist downregulation

The above table outlines the case series of seventeen pregnancies undergoing GnRH agonist down-regulation for adenomyosis prior to IVF.

Definitions: ET- endometrial thickness, Zoladex – goserelin acetate, PGS – Pre-implantation Genetic Screening

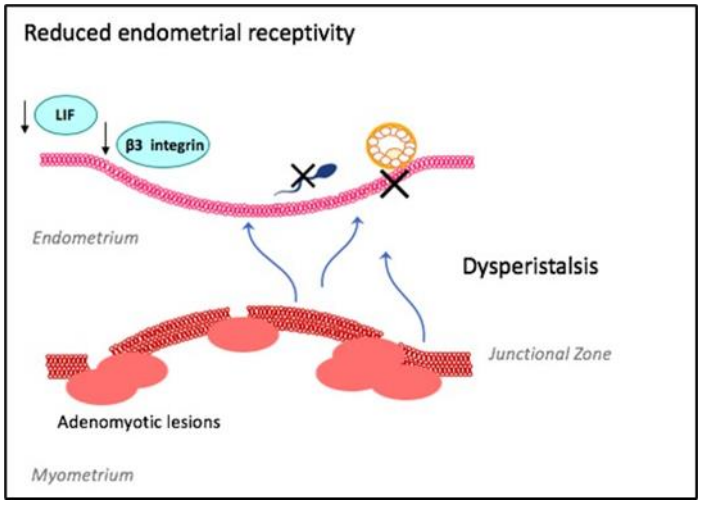

Figure 1: Suspected role of adenomyosis on fertility

The above figure outlines the key principles by which adenomyosis is thought to contribute to infertility. Adenomyotic lesions disrupt a proliferated junctional zone between the endometrium and myometrium, leading to uterine dysperistalsis and impaired sperm transport and blastocyst implantation. This is worsened by a hyperestrogenic state and reduced markers of endometrial receptivity that are critical to implantation.

LIF – leukaemia-inhibiting factor

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Laparoscopic ischial spine colpopexy: a new approach and first single center experience

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Abstract:

The laparoscopic treatment of prolapse is dominated by Sacral colpopexy, the latter is known as current Gold Standard". Various new approaches have been described in the last years with different fixation areas and different combinations. The ischial ligament is used successfully via vaginal route since a long time. We have developed a laparoscopic approach to use the structure for apical repair without mesh. This paper describes the technique and the first single center data for this new surgical technique.

Keywords: vault prolapse; laparoscopy, ischial spine,

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Introduction:

As mesh Issues caused by vaginal approaches occurred in the last two decades, more laparoscopic procedures were developed and published[1-3].Most of them were mesh based using long mesh arms (Dubuisson) or the pectineal ligament (Noé). There is a long tradition for abdominal fixation (laparotomy and vaginal fixation either [4, 5]. The ischial ligament is successfully used uni- or bi-lateral since decades but has not been approached abdominally yet. Due to mesh discussion, we have adopted the thread based vaginal technique to an abdominal approach. According to Delancy there is evidence that the apex of the vagina (level 1 by delaneys classification) is at the level of S2 vertebra or at the level of ischial spine. Also, the mid portion of the vagina (level 2 Delaney) derives a lot of support from the white line over the obturator fascia. Both the ischial spine and the white line are anatomically bellow the obturator nerve. With the advent of high-definition cameras, these areas can be precisely dissected and thereby we have a concept of repairing the prolapse to correct the anatomical position. Since the braided polyester suture stimulates a reasonable amount of fibrosis, we use it to replace the lack of collagenization at the vaginal end.

Surgical approach and technique

Trocar position

1) A 10 mm optic trocar is placed in the midline one inch above the umbilicus.

2) One 5 mm trocar is placed above and lateral to the left anterior superior iliac spine for the surgeon's left hand.

3) A second 5 mm trocar is added 2 inches to the left and two cm above the optical 10 mm trocar for the surgeon's right hand.

4) A third 5mm trocar is placed just above the MacBurneys point on the right side for the assistant.

5) A Fourth 5mm trocar is placed two inches below and to the right of the umbilicus. This is exclusively used for the safe and comfortable passage of the 40 mm 1/2 circle taper cut heavy needle, attached to the number 2 braided polyester suture.

STEP 1: Dissection to approach the ischial bone

A 10 mm 50-degree scope is passed through a supraumbilical canula. The retroperitoneum is entered by cutting the round ligament on the right side and incising the peritoneum cranially parallel to the right infundibulopelvic ligament (right ovarian vessels) and caudally to meet the uterovesical fold.

The para rectal space (Latzko's space) is developed between the uterine and internal iliac artery (lateral border) and ureter (medial border).

Further dissection is carried out at a level caudal to the obturator nerve just lateral to the internal iliac artery and the umbilical artery.

The dissection is to be carried out with great caution taking very small steps and sometimes even excising small bits of the fat (in view of the vast variations in the anatomy as mentioned above), until the ischial bone is reached.

Further careful dissection of this part of the ischial bone anteriorly and parallel to the obturator nerve is commenced.

One may come across small branches of the obturator artery and vein or even the corona mortis which may have to be cauterized and cut to prevent soiling of the area.

At this level the concerned strip of the ischial bone with its periosteal thickening is bordered by Obturator muscle anteriorly, the lumbosacral trunk, internal iliac vein and internal iliac artery posteriorly and the obturator nerve cranially as well as the greater sciatic notch caudally.

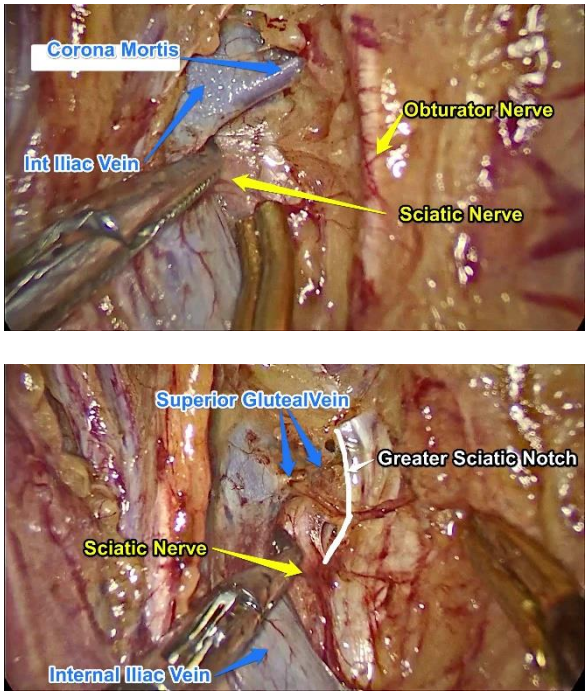

Fig. 1: Dissection area to approach the ischial spine

STEP2

A 40 mm 1/2 circle taper cut heavy needle attached to the number 2 braided polyester suture of 75 cm length is taken.

The needle is passed directly through the anterior abdominal wall above the right anterior superior iliac spine, thus ensuring that only the requisite suture material is intra-abdominal to prevent it from entangling and facilitating handling of the long single strand which will be used for further surgery.

The needle is then passed through the thickened portion of the periosteum above the ischial bone previously exposed using a laparoscopic needle holder passed through the fourth 5mm port described above.

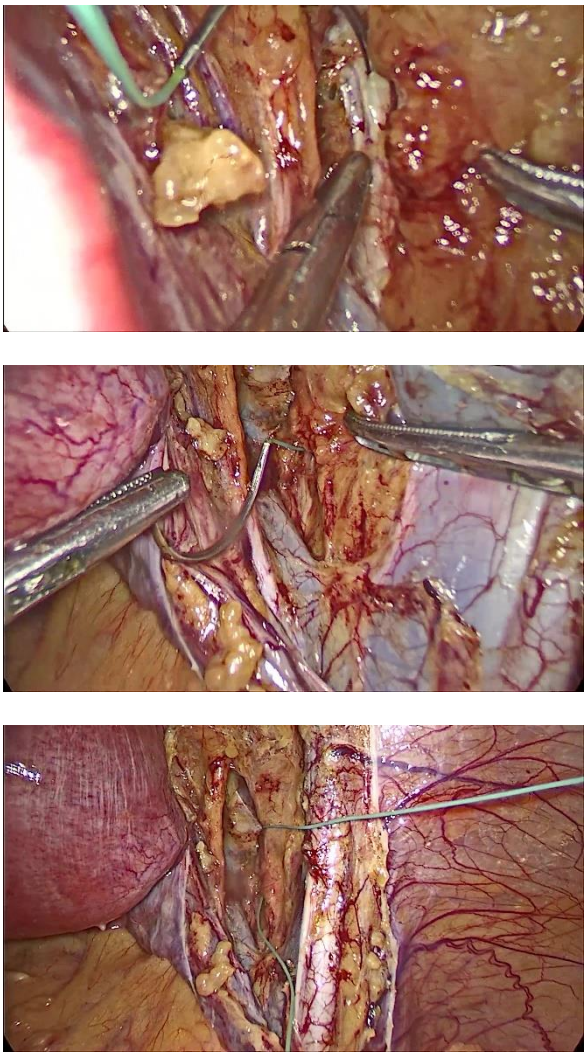

Fig. 2: Needle and thread placement

STEP 3

The 10 mm 50-degree scope is replaced with a 10mm 30-degree scope. A total hysterectomy is achieved if the patient is perimenopausal as it facilitates the further surgery. The vaginal vault is

sutured in two layers to reduce the extrusion of the poly-filament polyester suture.

STEP4

A 1.5-inch broad malleable copper retractor is introduced into the vagina from bellow. This facilitates the further dissection and suturing.

The vesico vaginal space is dissected out up to the trigone of the bladder. The peritoneum on the posterior vaginal wall is cut above the fat and the rectovaginal space is dissected out up to the lowermost part of the prolapse (not up to the levator ani) as guided by the assistant's finger behind the malleable retractor.

STEP5

The aforementioned needle on polyester which is already passed through the thickened periosteum of the ischial bone is now passed by multiple bites through the fascia on the anterior vaginal wall (plication of fascia) taking care not to enter the vagina avoiding full thickness bites.

The suture material is deliberately kept on the right side of the operative field to avoid entangling. Bites are taken in a parallel manner to facilitate laparoscopic suturing. The polyester suture is pulled through to leave at least about 30 to 35 cm to do a similar plication of the posterior vaginal wall fascia.

The suture on the anterior vaginal wall is now pulled to tighten the plicated area thus correcting the cystocoele. The needle is now held and passed in a manner similar to the anterior vaginal wall, posteriorly starting from fascia over the lowermost part of the prolapsed posterior vaginal wall (as guided by the assistant's finger placed posteriorly behind the malleable retractor).

Sequential bites are taken at multiple points in the fascia over the posterior vaginal wall (taking care not to entangle the suture) thus plicating the posterior vaginal wall and tightened effectively correcting the rectocele and enterocoele.

Special care must be taken at the area of the posterior fornix not to take full thickness bites as that part of the vagina is likely to be the thinnest.

A single bite is now taken in the anterior vaginal wall fascia (So that the subsequent knot lies anterior to the vaginal vault suture line and the suspension bridge from the ischial bone remains well above the right ureter) and 3 to 4 knots are tied to the afferent suture coming from the ischial bone so that even if the polyester suture is extruded, the suspension from the ischial bone remains in place.

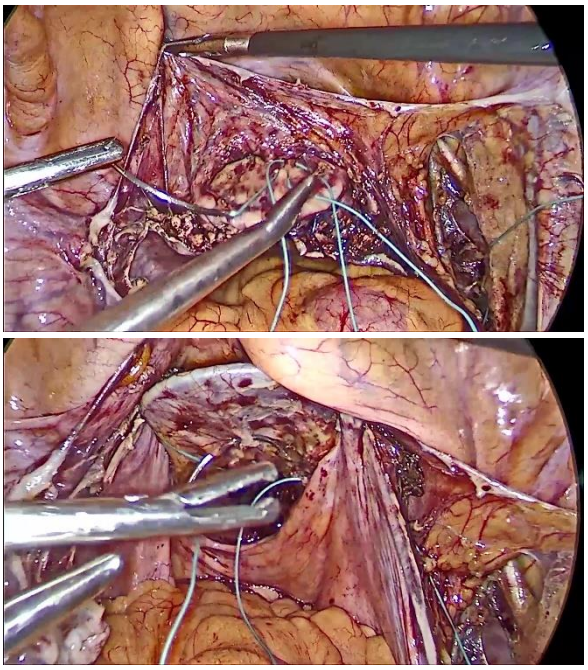

Fig. 3 Attachment to the vault after vaginal dissection

STEP 6

Now the suture is tied securely to the end coming into the abdomen (The suture end passing into the ischial bone) thus binging the vaginal apex to the S2 level. The excess suture from both ends and the needle are cut and removed from the abdomen.

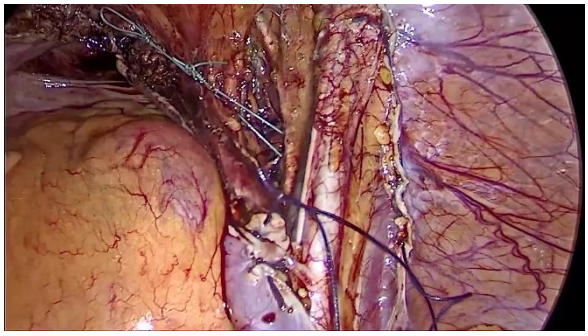

Fig. 5: Tightening of the thread to stabilize the apex

STEP 7

The entire peritoneum which has been opened from bellow the round ligament on the right side to the round ligament on left side is closed with 1-0 polyglycollic acid (Vicryl) in such a manner that the whole polyester suture and suspension system lies retroperitoneal.

This must be done meticulously as the polyester incites severe fibrosis and if exposed causes severe intestinal adhesions.

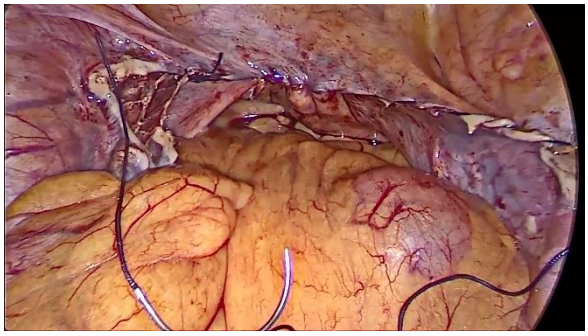

Fig.6: Closure of the peritoneum

Material and Method

We have followed up the patients one week, six months, one year and two years after surgery. The surgery was performed in 46 cases over a period of 3 years.

Table 1 shows the age of the patients and time taken for the surgery.

Descriptive Statistics: Age and Time for Surgery						
	N	Range	Min	Max	Mean	SD
Age (years)	46	31	39	70	48.89	8.295
Time for surgery (mins)	46	60	135	195	148.80	10.175

Table 1: Patient age and surgical time

The distribution of the stages of prolapse and defects is listed in Table 2.

Prolapse	**Frequency**	**Percent (%)**
Prolapse Grade 2	16	34.8
Prolapse Grade 3	26	56.5
Total Prolapse (Grade 4)	4	8.7
Total	46	100.0
Cystocele	23	50.0
Rectocele	24	52.2

Table 2: Distribution of defects

According to further symptoms we combined the ischial colpopexy with Burch colposuspension or lateral defect repair. The patients were assessed preoperatively for Stress urinary incontinence and those with genuine stress incontinence underwent Burch colposuspension. Those with lateral wall defects were further examined on table after the primary surgery (Ischial colpopexy). Suitable candidates further underwent a paravaginal repair.

Procedure	Frequency	Percent (%)
BURCH		
Yes	7	15.2
No	39	84.8
Total	46	100.0
Paravaginal		
Yes	8	17.4
No	38	82.6
Total	46	100.0

Table 3: Distribution of concomitant surgery

Results:

Complication:

In 4.3% (2) of cases two major complications occurred (Obturator vein injury). These could be managed intra-operatively without any consequential damage. Smaller complications such as wound infections or urinary tract infections occurred very rarely.

Follow-UP:

In the first (one week) follow no recurrence was measured. One patient complained about pain and one about constipation (Table 4). After 6 months still no recurrence was seen but one major bowel obstruction. By reviewing the surgical video, we realized that the peritoneum was not sutured back properly.

After one year 8.8% of the patients had an adverse outcome. One due to relapse and 3 due to exposure of the polyester thread with pain and leucorrhea.

One year later no additional recurrence occurred but one more thread exposure, finally 8.8% after 2 years while the recurrence rate was only 2.2%.

Follow–up @ 1 week	**Frequency**	**Percent (%)**
Pain	1	2.2
Constipation	1	2.2
Recurrence	0	0.0
None	45	97.8
Total	46	100.0
Follow–up @ 6 months	**Frequency**	**Percent (%)**
Pain	0	0
Bowel obstruction	1	2.2
Recurrence	0	0
None	44	95.6

Total	46	100.0

Table 4: complication and results one week and 6 months after surgery

Follow–up @ 1 year	**Frequency**	**Percent (%)**
Pain, leukorrhoea with extrusion of ethibond in vagina	3	6.6
Recurrence	1	2.2
None	42	84.6
Total	46	100.0
The stitch was cut flush at the vaginal vault in OPD, following which pain and leukorrhea disappeared and prolapse did not recur.		
Follow–up @ 2 year	**Frequency**	**Percent (%)**
Pain, leukorrhoea with extrusion of ethibond in vagina	1	2.2
Recurrence	0	0
None	45	97.8
Total	46	100.0
The stitch was cut flush at the vaginal vault in OPD, following which pain and leukorrhea disappeared and prolapse did not recur.		

Table 4: complication and results one week and 6 months after surgery

The patient with bowel obstruction received a second laparoscopy 2 months after the first surgery where it was found that loops of ileum were adherent to the exposed polyester suture at the site of the knot. The patient with suture exposure were treated by excising the exposed thread without recurrence.

Discussion:

Laparoscopic ischial colpopexy is a new approach for prolapse repair. The laparoscopic ischial colpopexy is a new approach to prolapse correction. The ligament has been used in vaginal surgery to fix the apex for decades. However, the laparoscopic approach has not yet been described. The technique was carried out early on and was described in the literature in 1981[6]. 2001 Lantzsch et al. Described urinary tract infection (n = 16, 8.0%), temporary irritation of the sciatic nerve (n = 15, 7.5%), temporary partial ureteral obstruction (n = 11, 5.5%) and less blood loss than 400 ml (n = 7) 3.5%) as operative complications (n = 200)[7]. In comparison, our operative complication rate is very low and must therefore compete with today's procedures such as sacropexy, pectopexy and lateral suspension. Compared to the procedures mentioned, our complication rate remains low. This is of course the small number of cases and the implementation by a very trained surgeon is of limited significance[8-11]. Lantzsch et al. reported that 119 of 123 patients were completely healed with no evidence of urinary incontinence or prolapse. At the follow-up examination, 4 patients (3.25%) had a recurrent vaginal vault prolapse. Recurrent cystoceles, rectoceles, enteroceles were found in 10 cases (8.1%), (0.8%), (0.8%). The follow-up period in this study was 4.8 years on average. In

comparison to the data from Lantzsch and the data from comparable laparoscopic studies, our success rate in the examined collective is very satisfactory. However, since we wanted to work without mesh, the high exposure rate of 8.8% must be viewed critically.

Limitations of the technique:

The thread material is supposed to produce good fibrosis, but appears to favor exposure due to its strong local irritation. Therefore, we have to look for alternatives regarding the material. The suturing technique appears to be effective in correcting even a combined prolapse. However, the proximity to the ureter, pelvic floor nerves and lymphatic tissue requires a high level of expertise from the surgeon, which will make it more difficult to spread.

Conclusion:

The laparoscopic ischial colpopexy can expand the portfolio of laparoscopic techniques for correcting pelvic floor defects. The early experiences with the technology show the feasibility but also the limitations. The material and safety of the technology still has to be examined on a larger scale before further statements can be made. Polyester sutures seem to be of risk to be used close to the vagina.

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Identical ovarian and deep pelvic endometriosis with colorectal involvement in monozygotic twins: a case report and review of the literature

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Abstract:

Endometriosis is a common benign gynecologic disease characterised by the presence of ectopic endometrial tissue outside the uterus. We present a brief review on the genetic factors underlying endometriosis, followed by a case report on concordant anatomical distribution of deep infiltrating endometriosis (DIE) in a pair of monozygotic (MZ) twins. To our knowledge, this is one of the first reported cases of DIE in MZ twins. The remarkable concordance and resemblance of deep disease involving the same anatomical sites, ovaries, pelvic floor and rectosigmoid colon in our MZ twins reiterates the role and impact of genetic factors in the pathogenesis of endometriosis.

Keywords: Deep infiltrating endometriosis, sigmoid resection, monozygotic twins, genetics.

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Introduction

Endometriosis is a polygenic multifactorial disease. Incidence of deep infiltrating endometriosis (DIE) involving the GI tract is estimated at 8-12% (1, 2) and commonly involves the rectosigmoid colon. Endometriosis is one of the most common benign gynecologic diseases. It causes pelvic pain and subfertility, and is characterised by the presence of ectopic endometrial tissue outside the uterus (3). Endometriosis is under-diagnosed and associated with a mean latency of 6.7 years from onset of symptoms to definitive diagnosis (4).

Most estimates of prevalence are made on the basis of surgical cases or small samples, and are highly selective, ranging between 5% and 10% in women of reproductive age, and up to 50% among infertile women (5-7). Endometriosis has an important socio-economic impact, because it greatly lowers quality of life for a significant portion of the population, and is responsible for substantial health expenditure, diagnosis, treatment, and loss of economic performance. The cost of endometriosis to the US health care system was $69.4 billion in 2009 (8).

Despite 150 years of hypothesis-driven research, the cause of endometriosis remains uncertain. Therapeutic options are therefore limited, often lacking unanimous consensus. However, there is mounting evidence that endometriosis is a complex multifactorial disease, with both genetic and environmental components contributing to disease susceptibility (7).

In 1980, Simpson et al. published the first formal genetic study of endometriosis (9). Studying 123 probands with histologically proven endometriosis, they found that 5.9% of female siblings over the age of 18 years had endometriosis; the mothers were affected in 8.1% of cases. However, only 1% of the patients' husband's first-degree relatives (controls) had the disease. Women with an affected sibling or parent were more likely to have a severe form of endometriosis (10). Severe endometriosis was present in 61% of probands who had an affected first-degree relative, whereas it was only present in 23% of the affected probands with no affected first-degree relatives.

One recent meta-analysis combining results from a genome-wide association study and replication studies showed that of the nine loci found to be associated with endometriosis in at least one of the studies, six remained statistically significant genome-wide, and two showed borderline statistically significant genome-wide association with moderate/severe disease (11).

In an Australian twin-based study, a twofold increase in endometriosis risk in monozygotic (MZ) compared with dizygotic (DZ) twin pairs was reported (7), which suggests that the genetic component contributing to phenotypic variability in endometriosis is about 52%.

These data imply that endometriosis is a complex genetic trait, and indicate that a number of genes interact with each other to form disease susceptibility, with the phenotype emerging in the presence of environmental risk factors which in themselves account for 53% of disease liability. Environmental chemicals, as well as food, have been discussed as possible contributing factors (12-14). However, there is no existing evidence as to the nature of this environmental contribution. Only one study to date has used quantitative analysis to examine the contribution of genetic and environmental factors to endometriosis, using a small twin sample (7). A larger twin sample is expected to provide further clarification on the role of genetic and environmental factors (12-14).

In this report, we present a case of deep infiltrating endometriosis (DIE) in a pair of monozygotic (MZ) twins.

Materials and Methodes

A pair of monozygotic twins was referred to our office within the same year, mainly due to severe chronic pelvic pain and infertility, with the following pertinent clinical information.

Twin A

A 31-year-old nulliparous woman was admitted to Farmanieh Hospital (Tehran, Iran) complaining of heavy menstrual blood loss, progressive chronic severe pelvic pain and dyspareunia for the past six years, painful defecation with passage of narrow, occasionally blood-tinged stool, and history of failed hormonal medical treatment on and off during the past six years. The patient had been infertile for the past two years. She had a history of hypothyroidism, thalassemia minor, cervical spinal cord tumour surgery, rhinoplasty and eye surgery.

Transvaginal ultrasound detected a 25 x 22 mm cyst in the right ovary and two heterogeneous hypoechoic foci (32 x 21 mm and 29 x 18 mm respectively) in the left ovary; these observations were compatible with endometrioma. Colonoscopy results were negative. Haemoglobin level was 11.4 g/dL, serum CA 125 level was 60.67 U/mL, serum CA 19-9 level was 6.4 U/mL, and AMH level was 5.7 ng/mL.

Intravenous pyelogram (IVP) results were negative. Magnetic Resonance Imaging (MRI) revealed two T1 foci in both ovaries (25 x 15 mm and 20 x 10 mm high respectively), which was suggestive of a hemorrhagic cyst or, more probably, a dermoid cyst. No other pathology was noted, including for the intestinal tract.

Twin B

A 31-year-old nulliparous woman was admitted to Farmanieh Hospital (Tehran, Iran) complaining of menorrhagia, progressive severe pelvic pain with rectal radiation for six years, painful defecation with occasional blood-tinged stool, severe dyspareunia for the past two years and failed hormonal medical treatment for the past six years. She had a history of hypothyroidism, thalassemia minor, mitral valve prolapse rhinoplasty and eye surgery.

Two transvaginal pelvic ultrasounds revealed a small anterior wall myoma; there were no other findings. Haemoglobin level was 10.8 g/dL, serum CA 125 level was 21 U/mL, serum CA 19-9 level was 9.4 U/mL, and AMG level was 7.4 ng/mL.

IVP results were within normal limits. MRI of the pelvis detected three small myomas (10-15 mm) at the anterior uterine wall and a 20 mm follicle in the left ovary. There were no other findings, including for the bowel. Colonoscopy results were within normal limits.

A double-contrast barium enema detected segmental luminal narrowing with upward displacement of the sigmoid area, with mucosal thinning. These findings suggested extrinsic pressure, mostly due to endometriosis, which seemed to have involved the posterior wall of the sigmoid colon.

Results

Case reports

Twin A

A laparoscopic segmental sigmoid resection was performed, with staple re-anastomosis, resection of pelvic floor DIE, and resection of a bilateral endometrioma. The procedure took 152 minutes. The patient was discharged after four days.

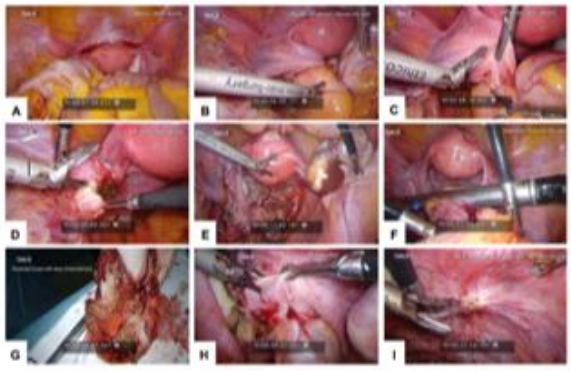

Figure 1. Laparoscopic images for Twin A. (A) Anterior uterine myoma; (B) attached left adnexa to stenotic RS colon; (C) left adnexal colonic adhesion; (D) left ovarian endometrioma; (E) right ovarian endometrioma; (F) segmental resection of RS colon; (G) resected bowel with deep endometriosis; (H) DIE pelvic floor; and (I) DIE at peritoneal site of invagination.

Twin B

Twin B's initial procedure was performed in the same month as Twin A's procedure. Laparoscopic adhesiolysis was performed, with resection of pelvic floor DIE, resection of ovarian endometrioma, and shaving of rectal deep endometriosis. Bowel resection was deferred because of inadequate bowel preparation, and laparoscopic sigmoid resection with staple anastomosis was performed 8 weeks after the initial procedure.

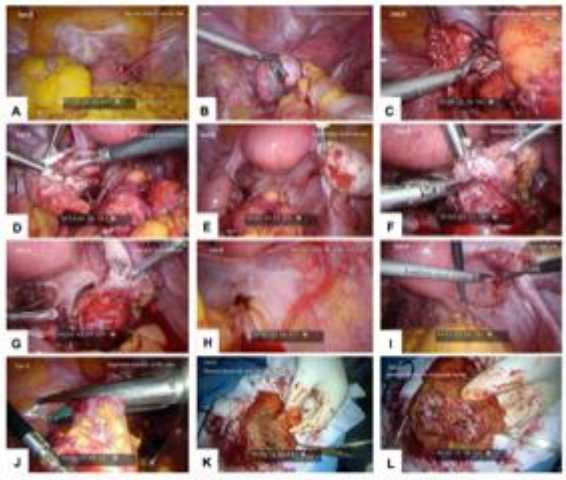

Figure 2. Laparoscopic images for Twin B. (A) Myoma at anterior uterine wall; (B) left ovarian endometrioma severely attached to sigmoid colon; (C) ovarian attachment to sigmoid colon; (D) left ovarian endometriosis; (E) right ovarian endometriosis; (F) shaving DIE off rectal surface; (G) defect at rectal surface; (H) DIE pelvic floor with rectal involvement; (I) DIE peritoneal site on invagination, right side; (J) segmental resection of RS colon; (K) resected bowel with undivided DIE nodule; and (L) resected bowel with divided DIE nodule.

Discussion

Anatomical involvement of the relevant pelvic organ in this pair of identical twins was nearly identical. The twins had grown up in the same environment all their lives, both worked as secretaries at the same institution, and both were experiencing not only the symptoms of DIE, but also signs and symptoms of intestinal involvement.

The ultrasound and MRI for both patients was reported negative, which clearly indicates the importance of experienced radiologists for the detection of deep endometriosis (15), and reiterates the importance of bimanual exam by clinicians, particularly at the time of menstruation, to achieve an accurate diagnosis,.

In fact, available data clearly indicates that the accuracy of good physical examination (PE) is not much different from that of transvaginal ultrasound (TVS), rectal endoscopic sonography (RES) and MRI, all within the 80% range (16).

Twin A was managed by laparoscopic segmental sigmoid resection and staple re-anastomosis, along with global resection of DIE from the pelvic floor and endometriomas. We were intending to do the same for Twin B, but in OR the lack of adequate bowel preparation was noted; thus bowel resection was postponed and was performed 8 weeks later under adequate bowel preparation. However, after resection of endometriomas and pelvic floor deep lesions, larger DIE at the posterior cervix with extension to the rectum was managed by laparoscopic deep shaving, and the defect was closed in two layers. No intraoperative or postoperative complications were encountered in either twin.

In many instances, the culprit for DIE is endometrioma, which starts from its peritoneal site of invagination (see Figure 3). By extension medially, it involves the parametrium, rectum, recto-vaginal space, and, by deeper extension, the ureter and hypogastric nerve may also be involved. Thus, timely and proper surgical management of endometrioma and excision of its dependent peritoneal site could serve not only to manage patients' pain, but also to conceivably prevent a significant percentage of DIE. In one study focusing exclusively on patients with rectosigmoid lesions, 48% and 84% had ovarian endometriosis and retrocervical lesions, respectively (17).

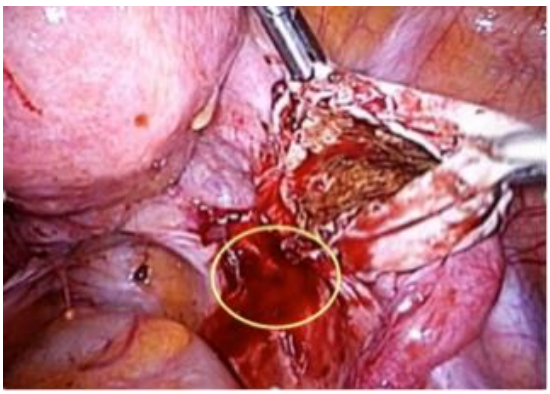

Figure 3. Endometrioma and the peritoneal site of invagination.

Both superficial peritoneal and ovarian endometriomas may be found in association with DIE in variable percentages, thus contributing to the intensity of painful stimuli, as well as to infertility (18). This raises the question of whether DIE is an independent form of the disease, or whether it represents its most severe clinical representation (19).

Nisolle and Donnez have argued that peritoneal endometriosis, ovarian endometriosis and adenomyotic nodules of the rectovaginal septum are three different entities (20). In a retrospective observational study by Kamergorodsky et al. (21) involving 176 patients and 271 biopsies, the histologic differentiation in superficial endometriosis, DIE, and ovarian endometriomas was evaluated. Results showed a predominance of the undifferentiated glandular pattern (33.5%) and mixed glandular pattern (46.9%) in deeply infiltrating endometriosis lesions, whereas the well-differentiated glandular pattern (41.8%) was most frequently seen in superficial endometriosis lesions, and in ovarian endometriomas a predominance of both the undifferentiated (40.5%) and mixed patterns (37.8%) was observed. There was no significant histologic difference between infiltrative endometriosis of the uteroscacral ligaments,

recto-vaginal endometriosis, or bowel endometriosis. There was a predominance of the well-differentiated pattern in more superficial lesions and the undifferentiated pattern in deeper lesions, suggesting a similar mechanism to explain the invasive potential of endometriosis.

Recently, Sapkota et al. (22) analysed genetic risk scores derived from two large European genome-wide association (GWA) datasets from a previous multi-ethnic GWA meta-analysis of endometriosis. They found that genetic factors contributing to minimal disease might differ from those contributing to more severe endometriosis, and that more severe endometriosis cases exhibit greater genetic load than minimal or mild disease. The genetic burden generally increased from less severe (minimal) to more severe disease, consistent with disease progression.

Considering the fact that endometriotic lesions undergo repeated cyclic bleeding (injury) and repair like other organs leading to fibrosis (23), it has been suggested that endometriotic lesions are fundamentally wounds with repeated tissue injury resulting in smooth muscle metaplasia (SMM) and ultimately fibrosis via EMT (epithelial mesenchymal transition), MET (mesenchymal epithelial transition) mechanism securing disease progression (24). This points to the complex microenvironment and cross-reaction of endometrioc lesions with other cells, i.e. platelets and macrophages, their gradual but progressive evolution to SMM and fibrosis and, on occasion, to malignancy and the new traits/phenotypes they may acquire while losing old ones. This dynamic feature of endometriotic lesions may explain the reason for some conflicting results in this area of research, including the fact that development of biomarkers for the diagnosis or prognosis of endometriosis has posed a challenge, at least until now.

Endometriosis appears to be a progressive disease and its progression is mediated by EMT,MET mechanism and stemness ability of mesenchymal cells capable of creating local invasion and progression, intravasation and extravasation providing new sites or distant metastasis, as evidenced by longitudinal laparoscopies in female baboons (25). In regard to human evidence there are at least eight studies reported (with a total of 162 patients) on repeat laparoscopy in women assigned to placebo treatment. The results indicate nearly equal distribution among women whose disease stage deteriorated (31%), was unchanged (32%), or improved (38%). In fact, all but one study noted that 23% of placebo patients had complete regression of the disease over intervals of 4 to 39 months, meaning that in over two thirds of women the disease will either persist or progress (26-33).

The clinical presentation of the disease followed a similar trend and evolution in both of our cases. Each twin had undergone a long history of slowly progressive pelvic pain from the point of intensity, developing bowel symptoms and dyschesia as time passed, with dyspareunia and change in stool diameter to the point of bowel stenosis and impending obstruction. All of this is consistent with the progressive nature of the disease reported in the existing literature.

Deep infiltrating endometriosis, contrary to being an estrogen-dependent disease, usually does not respond well to hormonal suppressive therapy. Adequate surgical excision of the lesions provides the best long-term results and symptomatic relief (31, 34) but surgical treatment of colorectal endometriosis has been

quite controversial, and there is no consensus on whether the treatment should be conservative (shaving or discoid resection) or radical (segmental bowel resection and anastomosis) (35). Lately, there has been a great deal of effort to advocate shaving as the treatment of choice, mainly because of higher early and late complications in segmental colorectal resection. (36)

The efficacy of segmental colorectal resection is very debatable; data in this regard is controversial and, to a great deal, dependent upon the surgeon's experience (37, 38).

Findings from histopathologic analysis of specimens from segmental bowel resection indicates that in 50% of cases there is a satellite lesion independent of the primary deep nodule. The deepest layer of the bowel wall containing endometriotic foci at the primary lesion is in the submucosal layer in 70% of cases and the internal circular muscle layer in 30%. Furthermore, persistent lesions are present in 50% of patients treated with discoid or shaving resection (39).

The latest systematic review of different surgical approaches to bowel and rectovaginal endometriosis showed that the complication rate is variable for conservative and radical treatment. Recurrence rate for shaving was reported at 22.2%, for discoid resection 5.17%, and in segmental resection 2.19%; these rates were significantly different (37) but comparatively complete resection of bladder DIE had no recurrence reported by different authors. Positive bowel resection margins as well as age <31 years and body mass index ≥23 kg/m2 appear to be important independent predictors of disease recurrence (40).

Our patients were managed according to the extent of their pathology, intensity of pelvic pain, and future fertility, thus minimising the chance of recurrence. Most importantly, patients' wishes and decisions regarding the type of clinical management to be pursued were incorporated, following proper counselling and explanation.

It is crucial to keep in mind that there is currently no scientifically proven best treatment technique for bowel endometriosis, and comparison of different currently practised techniques is not possible. According to our current knowledge regarding the nature of disease pathology and its progression, recurrence should be as much a source of concern as complications for any given surgical technique. Resection of DIE should be global, not local, and limited to the bowel. Operations should be performed by an experienced surgeon and a well-organised team, in a multidisciplinary fashion.

In summary, we present a case of advanced DIE in monozygotic twins; one of the first such cases to our knowledge. Our findings reiterate the importance of genetic factors in the aetiology of endometriosis. These cases also reinforce the importance of the following considerations for surgeons considering conservative treatment for colorectal endometriosis:

- The extent of lesions cannot be assessed macroscopically with certainty.
- Assessment of the depth of infiltration of endometriosis foci in the bowel requires an expert radiologist.
- In the majority of rectal endometriosis nodules, the fibrosis which is the landmark for shaving does not surround, but follows behind the glandular epithelium and stroma.
- More than 2 cm of bowel tissue must be removed from the main lesion to obtain clear margins in two thirds of patients.

- Low rectal lesions (<5–8 cm from the anal verge) are associated with a higher risk of complications.
- A great majority of patients have been through previous surgeries and have a good chance of recovering and becoming pain-free.

Acknowledgements: None.

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CASEREPORT

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Hysteroscopic removal of Retained Intra-uterine Fetal Bone which causes secondary infertility: A case report

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Abstract:

Intrauterine retention of fetal bones is a rare complication of unsafe abortion that can cause secondary infertility. We are presenting the case of a young woman who underwent an illegal abortion 4 years ago. Transvaginal ultrasonography suggested the presence of bone-like structures in the uterine cavity. Retained bony structure from a previous abortion should be considered as a cause of secondary infertility. Removal of the bony products and polypectomy was done by hysteroscopy.

Keywords: abortion, infertility, fetal bones, hysteroscopy, osseous metaplasia

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Introduction

Bone within the uterine cavity is an unusual finding in women with secondary infertility, which is usually associated with a past history of termination of pregnancy. The etiology of bone like calcification is unknown, but the most common ones include retained fetal bone and osseous metaplasia of endometrial tissue. Calcification appear as hyperechoic area on ultrasound but the gynecologist should also be aware of other differential diagnosis of endometrial ossification which includes mixed Mullerian mesenchymal tumor (MMMT), IUCD, Osseous metaplasia or endometrial tuberculosis. Hysteroscopy is gold standard as diagnostic and therapeutic method.

Retained fetal bone as an entity is not adequately reported in the literature. Prolonged retention of intrauterine bone is recognized as a cause of secondary infertility. One study reported that retained fetal bones may act as an intrauterine synechia or intrauterine device, increasing endometrial prostaglandin F2-alpha and preventing implantation, resulting in infertility (1). The reported incidence is 0.15% at diagnostic hysteroscopy (2). These patients may present with pelvic pain, dysmenorrhea, abnormal uterine bleeding and infertility (3). Apart from retained intrauterine, endometrial calcified lesions and the presence of ectopic bones can also occur by metaplasia in association with chronic inflammation and tissue destruction, mostly presented after repeated spontaneous or therapeutic abortion (4-6)

A Procedure of termination of a pregnancy can at times result in unwanted immediate complications like hemorrhage, uterine perforation, cervical injury and at times in late complications like infections, bleeding, menstrual abnormalities and uterine synechia. As 50% of patients undergoing medical termination are young adults, there is a possibility of secondary infertility due to an abortion that has become complicated (7).

Occasionally if the abortion is done in the second trimester or if the fetus was removed by destructive means, some parts of the fetus may inadvertently be left in the uterine cavity, these could cause pelvic pain, abnormal uterine bleeding, infection, dyspareunia or even vaginally expulsion of fetal bones (8). Few reports have been published describing secondary infertility after abortion. It is expected that retained fetal bones would induce a uterine reaction in the form of vaginal discharge and pain in the majority of such patients while also leading to secondary infertility.

We are presenting the case of a young female who had secondary infertility without any other associated complaints. She had a previous second-trimester abortion four years ago. shad failed to conceive again after previous curettage. Retained fetal bone was confirmed by histopathological analysis of bone fragment collected by hysteroscopy.

Case presentation

A 34 years old lady G4 P3 Ab1, presented in our office with secondary infertility. She had cesarean section delivery 3 times, according to the patient she had a history of an illegal termination of pregnancy at about 4th months of gestation by dilatation and curettage four years before. Following the abortion, she experienced abnormal uterine bleeding for several months and thereafter she could not conceive again. Even she had experienced vaginal expulsion of fetal bones several days after the curettage.

Her vital signs were within the normal range. Pelvic examination was done vaginally and abdominally, both of these were unremarkable.

She had hysteron-sonography revealed a 20 mm linear shadowing, T-shape echogenic lesion like an Intra Uterine Contraceptive Device (IUCD) in the endometrial cavity. Also, there was a 13 × 10 mm echogenic polyp in the posterior fundal wall.

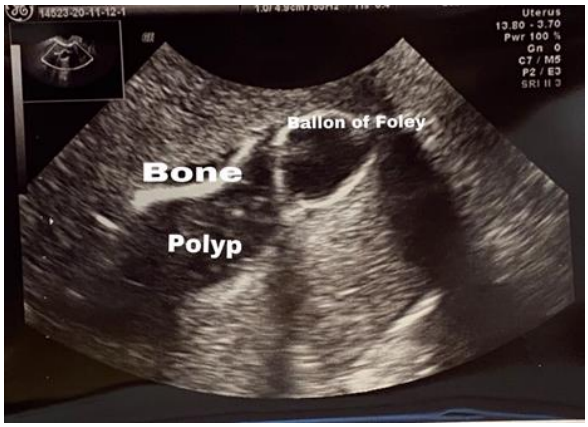

Fig.1-Hysterosonography revealed calcified intrauterine foreign body, which is suggestive of a retained fetal bone in this case

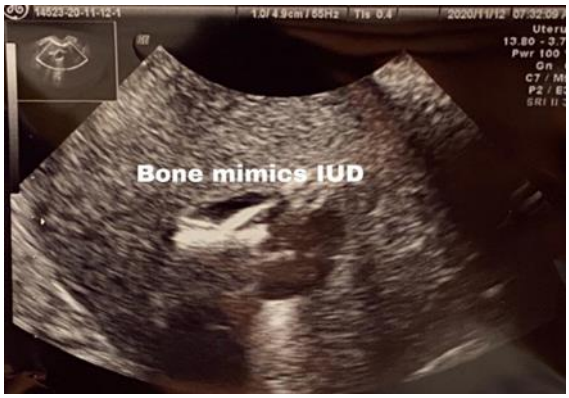

Fig.2-Hysterosonography: Retained fetal bone which mimics IUD

One of the differential diagnosis in the imaging evaluation is osseous metaplasia which can deeply embedded in the endometrium. But considering hysteroscopy findings and pathology report confirmed retained fetal bone. Under general anesthesia, a diagnostic hysteroscopy was performed that showed the presence of a bone (Figure 3) in the uterine cavity. Removal of the bony products and polypectomy was performed by hysteroscopy. On hysteroscopy evaluation there was bony structure which was lying freely in the uterine cavity and separated from underlying tissue and suggestive of bone-like structures in the uterus.

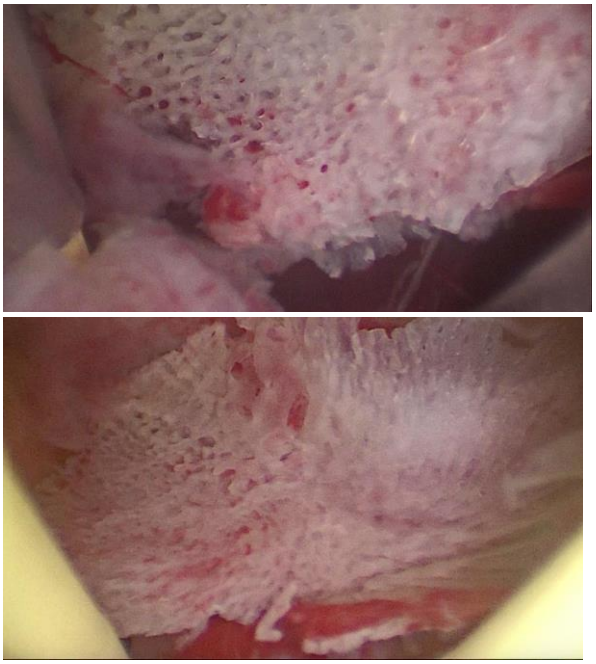

Fig.3-Hysteroscopy view of retained fetal bone

The extracted material was sent for histopathological examination. The pathology report was as follow: specimen received in formalin and consists of several soft irregular yellowish tissues with a little hard area aggregating to 3×1.8×0.8 cm/GH submitted in toto in 1 block. The histopathologic findings are consistent with endometrial polyp, few fragments of endometrium with proliferative pattern and degenerated fragments of bone trabeculae infiltrated by neutrophils. Necrosis and dead bone were observed in pathology,

retained fetal bone confirmed.

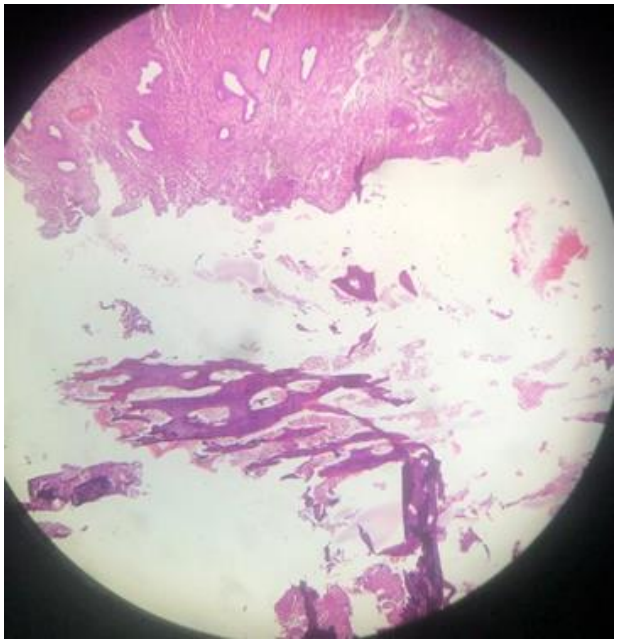

Fig.4 100x magnification of H&E histopathology photomicrograph of uterine content showing sponge bone fragments with bone marrow and secretory phase endometrium

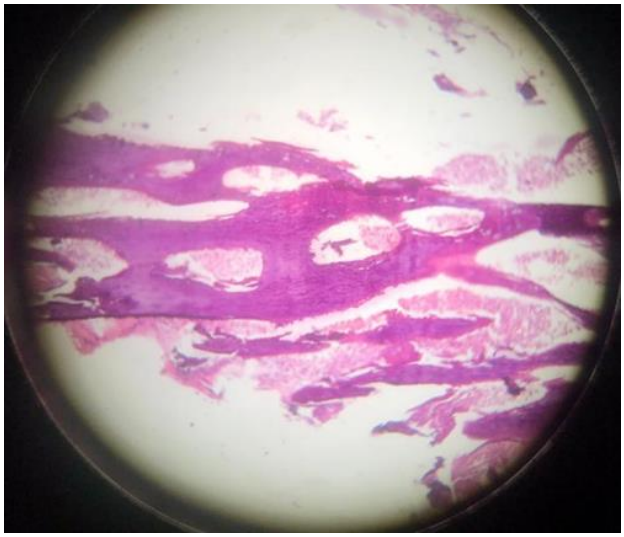

Fig.5-400x magnification of H&E histopathology photomicrograph of uterine content showing sponge bone fragments with bone marrow

The Post-operative recovery was unremarkable and subsequent menstrual function was normal.

Discussion

While termination of pregnancy (abortion) is usually a safe procedure, sometimes it has its own complications. Incomplete evacuation of the uterus can result in multiple complications including the rare complication of secondary infertility as presented by us in this case.

Many women with retained fetal bone will have symptoms of menometrorrhagia, dysmenorrhea, vaginal discharge, pelvic pain and spontaneous elimination of bony fragments during the menses in addition to their infertility (9).

Previously published case reports are suggestive that retained fetal bones may be responsible for secondary infertility (6,7,10).

The risk of infertility depends on the location of the retained pieces of bone, whether it is embedded in the myometrium or in the endometrial cavity. There is some evidence that the presence of an intramural bony fragment per se does not seem to compromise fertility if it is completely embedded (9) However commonly, it is speculated Retained fetal bone has been thought to act as a 'uterine synechia' or IUCD in the uterine cavity and prevent implantation (1, 10, 11).

Several theories for formation osseous metaplasia cited that previously, endometrial stroma is capable of cartilaginous metaplasia (18). Another hypothesis is that heteroplasia may occur in the multipotential stroma cells present in the uterus thereby forming osseous tissue (12).

In terms of observing hyperechoic area on ultrasound, the gynecologist should be aware of other differential diagnosis of endometrial ossification which includes IUD, osseous metaplasia and fetal retained bone.

It is also possible that the presence of bones near the fundal region (where blastocyst implantation mostly takes place) can lead to the elevation of endometrial prostaglandins (e.g., F2alpha) and thus prevent implantation (1).

In case of a history of previous abortion, the clinician should keep in mind the remote possibility of retained fetal bones as a cause of infertility. Transvaginal ultrasonography must be performed in all cases and if any suspicious echoes are seen a hysteroscopy should be planned.

Hystero-sonography allows a detailed visualization of the uterine cavity in both longitudinal and transverse planes, and the endometrium can be evaluated for the presence of polyps, submucous fibroids, intrauterine synechiae and foreign bodies (13)

Hysteroscopy has both accurate diagnostic and therapeutic values. Hysteroscopic removal of the bony pieces should be regarded as the gold standard of treatment since it enables a complete removal of the bones under direct vision.

In terms of pathology, minimal adjacent endometrial reaction with endochondral ossification may help to distinguish osseous metaplasia from retained fetal osteoblastic tissue after an abortion.

Conclusion:

Considering that most reports of retention of fetal bones have been in patients with second trimester abortions, routine transvaginal ultrasound to evaluate the completeness of evacuation is suggested. Hysteroscopy guided removal of bony fragments is the gold standard and leads to complete resolution of symptoms.

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Laparoscopic management of cesarean scar pregnancy: a case report and literature review

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Abstract

Background: Cesarean section scar pregnancy is one of the rarest forms of ectopic pregnancy where the gestational sac is fully or partially implanted within the scar caused by a previous caesarean section. Its incidence is on the rise due to the increasing rate of cesarean sections (CS) and also to the increasing awareness and the better ultrasound diagnosis

Case presentation: A 28-year-old woman G3P2 with a history of 2 cesarean deliveries who was diagnosed by ultrasound scan at 8 weeks gestation with cesarean scar ectopic pregnancy that was confirmed by pelvic magnetic resonance imaging (MRI). At 4 weeks the patient had a pelvic ultrasound suggested an intrauterine pregnancy with a gestational sac visualized in the lower uterine segment suggesting a cervical stage of miscarriage. Surgical management was opted for a combination of laparoscopy and hysteroscopy. A ligation of bilateral ascending uterine artery ligation was performed.

Conclusion: Cesarean section scar pregnancy (CSP) is rare but life threating complication. It should be diagnosed and treated early because of the increased risk of massive hemorrhage or uterine rupture that engages the vital prognosis and the functional prognosis.

Keywords: Cesarean scar pregnancy, ectopic pregnancy, laparoscopy, hysteroscopy

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Introduction

Cesarean section scar pregnancy is a rare form of ectopic pregnancy that increased in recent years due to the parallel increase of CS. It's a complex iatrogenic pathology defined as the implantation of the gestational sac in the myometrium at the site of a previous CS [1]. The first case was reported by Larsen and Solomon in 1978[2]. At early pregnancy it can be confused with a cervical stage of miscarriage or cervical pregnancy. Because of the dangerous complications such as uterine rupture, uncontrollable bleeding, and hemorrhage into the abdominal cavity; an early diagnosis and therapy are necessary to prevent the development of severe complications [3]. Here we report a case of this uncommon presentation of ectopic pregnancy with laparoscopic and hysteroscopic management in our department, this is the first case in the country managed by laparoscopy.

Case report

A 28 years old woman G3P2 with a history of 2 cesarean deliveries in the past, presented to the emergency department for minimal subacute intermittent blackish abnormal uterine bleeding at eight weeks and three days of amenorrhea without pelvic pain. Her most recent pregnancy was 3 years prior to the consultation. At 4 weeks and 4 days the patient did have a pelvic ultrasound suggesting an intrauterine pregnancy with a gestational sac visualized in the lower uterine segment revealing a non-viable detached intrauterine pregnancy (fig1) with a serum concentration of human chorionic gonadotropin (HCG) at 8306 mUI/ml.

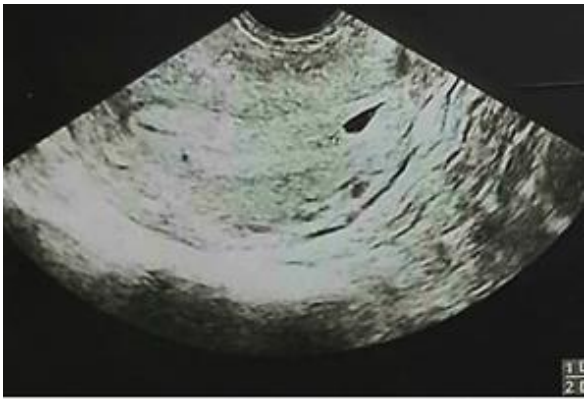


Figure 1: Sagittal Transabdominal ultrasound shows the gestational sac in the lower uterine segment

On clinical examination, the hemodynamic parameters were stable. The abdomen was supple at deep palpation. At Gynecological examination the only notable finding was a minimal blackish bleeding originating from a closed cervix at speculum. A pelvic and endo-vaginal ultrasound performed in the emergency department showed a low implanted intra-uterine sac of 52/28 mm at the level of prior cesarean scar in the lower uterine segment with a small rim of myometrium visible anterior to the gestational sac. The gestational sac was communicating with the endometrial cavity, whilst being located in the lower uterine segment of uterus. The cervical canal was closed and empty with no intraperitoneal fluid was noted (fig2, 3)

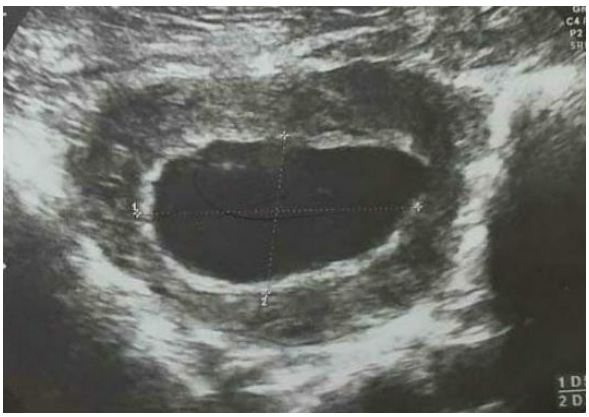


Figure 2: transversal transabdominal pelvic ultrasound imaging shows intra-uterine gestational sac of 52/28 mm

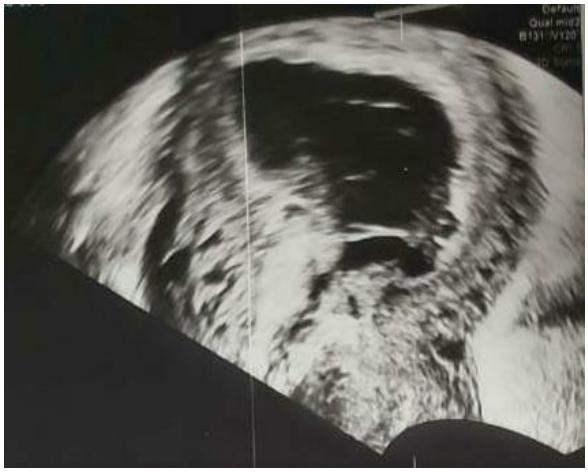


Figure 3: Sagittal endovaginal ultrasound demonstrating the characteristics of cesarean scar ectopic pregnancy : low lying gestational sac with anterior myometrial thickness , absence of cervical involvement

The serum HCG concentration was 40540 mUI/ml where the hemoglobin and hematocrit level were normal, 48 hours later serum HCG was at 53280 mUI/ml. A MRI revealed a gestational sac embedded in the hysterotomy scar, coming down to the serosa without the interposition of the myometrium, lateral on the right side (fig4). The diagnosis of caesarean section scar pregnancy was retained. A decision was made to proceed with a surgical management in the form of a laparoscopic resection of the ectopic pregnancy after hysteroscopic evaluation.

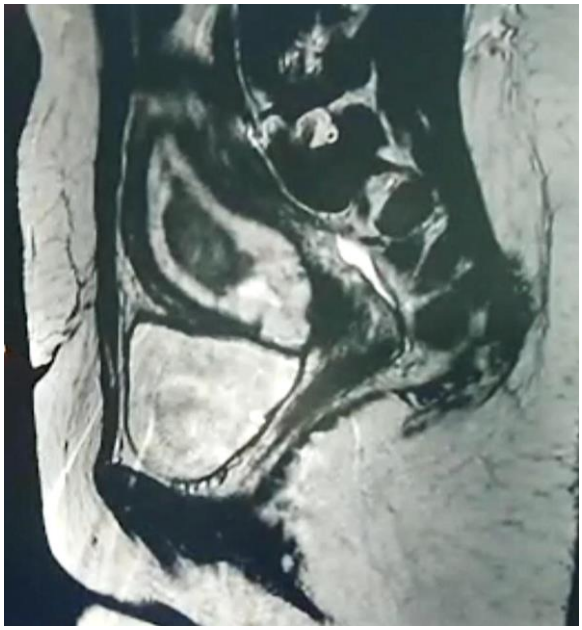


Figure 4: Magnetic resonance image shows a gestational sac implanted in the anterior wall of the uterus, and protruded into the uterine cavity.

Under general anesthesia a diagnostic hysteroscopy was performed first reveling the attachment of chorionic tissues around the isthmus (fig5). Then a laparoscopic exploration did follow. The omentum was densely adherent to the uterine surface, an adhesiolysis was done (fig6). A bilateral uterine artery ligation was performed for security reasons (fig7). The bladder was dissected down to expose vesico-vaginal space, a protrusion into the anterior wall of the uterus at the uterine isthmus was found (isthmocele) (fig8).

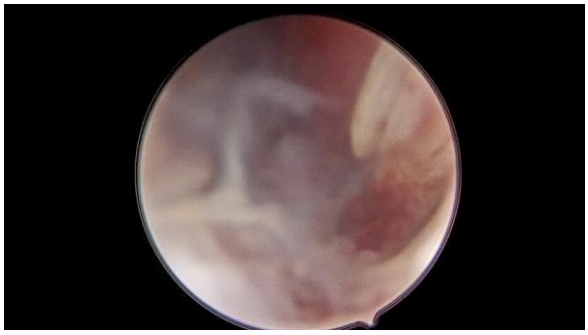
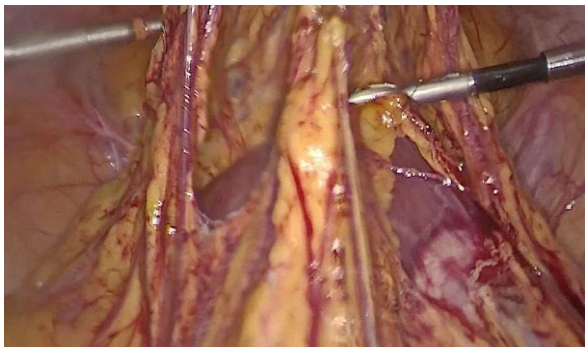


Figure 5: hysteroscopic view of trophoblastic tissue



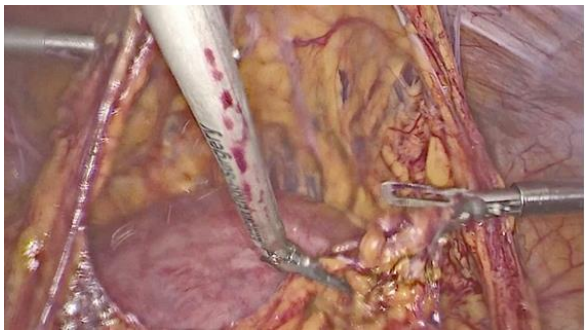


Figure 6: laparoscopic finding of the adherences of omentum to the uterine surface processes of adhesiolysis

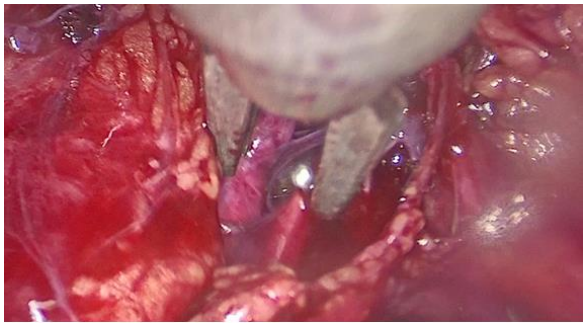


Figure 7: ligation of right and left uterine artery using clips

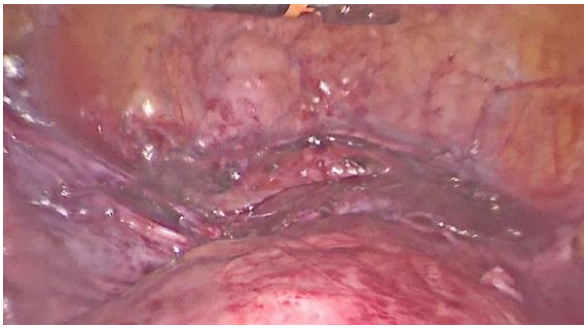


Figure 8: visualization of the isthmocele

An incision over the bulge was done then the trophoblastic tissue was removed and placed in an extractor sac (fig9). A suction curettage was performed. Interrupted Vicryl-0 sutures were used for closure of the defect (fig10). The specimen was sent for histopathological examination. The patient was discharged two days after the surgery.

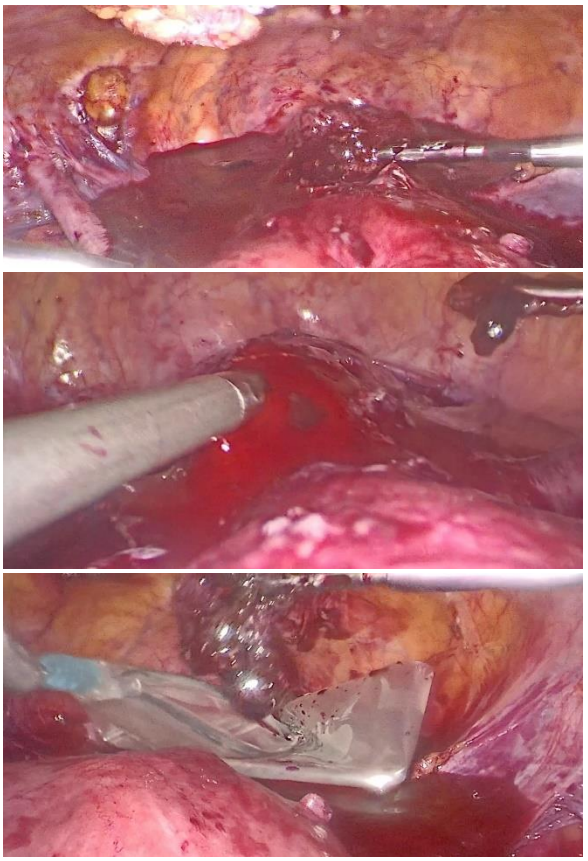


Figure 9: Incision over the bulge, excision of the trophoblastic tissue and it placement in extractor sac

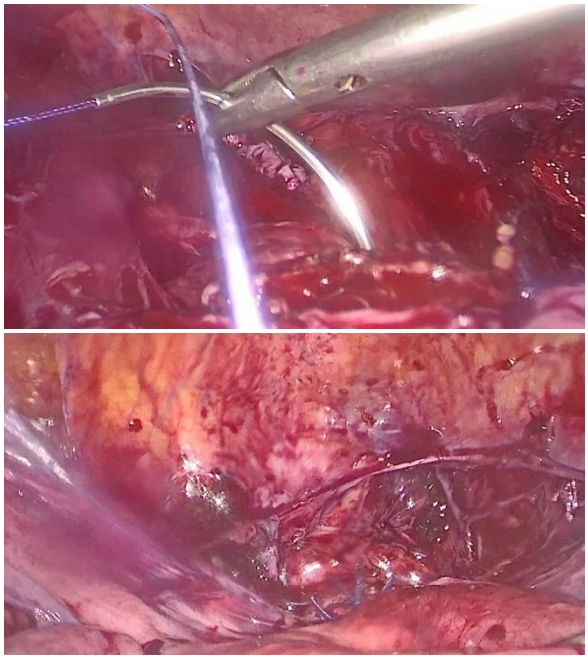


Figure 10: the closure of the defect using interrupted Vicryl-0 sutures

She was informed by the risk of recurrence of CSP, uterine rupture and placenta accreta for future pregnancies. Contraceptive advice was also given.

Two weeks later at follow-up, the patient's HCG was negative. The ultrasound examination revealed an empty uterine cavity.

Discussion

As one of the rarest forms of ectopic pregnancy, CSP is reported to occur in 1/1800 to 1/2216 of pregnancies, and it constitutes 6.1% of all ectopic pregnancies with a history of at least one cesarean delivery [4]. This kind of ectopic pregnancy is increasing in frequency, due to the increase in the number of caesareans in recent years.

Little is known about the mechanism and physiopathology of CSP. The mechanism suggested is the implanting blastocyst to invade the scar through a microscopic tract that develops from the trauma of an earlier cesarean scar [5]. Vial et al. suggested that there are two types of CSP. The first one results in the implantation of the fertilized egg in the scar and it development in the direction of the cervical isthmus and uterine cavity. This gives a chance for live birth, but the risk of massive bleeding from the implantation site is very high. The second type of CSP is represented by a deep implantation in a damaged cesarean scar and the development of a pregnancy leading to uterine rupture in the first trimester of gestation [6].

The diagnosis of pregnancy in a cesarean section scar is often made in the first trimester. In a review of 57 patients with CSP 38.6% presented with vaginal bleeding, and 24.6% presented with abdominal pain, as for a 36.8% of the patients were asymptomatic being incidentally detected by routine ultra-sonographic examination [7]. In an advanced stage, a CSP may result in uterine rupture leading to massive haemorrhage, haemoperitoneum and haemorrhagic shock.

Endo vaginal ultrasound allows an early diagnosis based on criteria established by Vial in 2000 represented in an empty uterine cavity and empty cervical canal, placenta or a gestational sac embedded in the scar of a previous caesarean section on sagittal section of the uterus [8]. There are also indirect ultrasound signs, such as decreased myometrial thickness between the gestational sac and the bladder and a circular blood flow surrounding the sac seen by color Doppler [9]

An MRI may provide additional confirmation of the ultrasound findings and specify the depth of trophoblastic invasion in the myometrium and the potential involvement of the serosa or bladder [10].

From the studies reviewed, no universal treatment guidelines have been established yet.

There are several variables to consider before recommending a treatment option as there are the haemodynamic stability, desire to preserve fertility and acceptability of prolonged follow up. Treatment modalities are either medical or surgical and are sometimes combined.

The medical approach consists of an administration of methotrexate through systemic route, or local administration (under ultrasound or laparoscopic-guidance) or a combination of the two [11, 12]. It takes about 4 to 6 weeks to normalize the HCG [9].

The surgical approach which is often conservative consist on removing the pregnancy and a repair of the uterine defect by laparotomy or laparoscopy., then the HCG normalization is reached in one to two weeks [13].

Because of the possibility of massive haemorrhage a preventive hemostasis by ligation of the uterine or hypogastric arteries may be necessary. Interventional radiology techniques like uterine artery embolization (UAE) can also be used preoperatively to decrease the risk of hemorrhage hysteroscopy [9]. UAE or vascular ligation does not appear to reflect fertility or the obstetrical prognosis following the patients [14].

The operative hysteroscopy and suction curettage have been described in various studies of CSP with a muscular layer ≥3mm [15]. It consists of the aspiration of the conceptus, haemostasis can be achieved with electro coagulation. A balloon catheter may be placed postoperatively for compression haemostasis. In a case series by Pan et al, hysteroscopy was used in conjunction with laparoscopy for patients whose muscular layer was< 3mm to avoid the risk of uterine perforation and bladder injury [15].

For future pregnancy the risk of recurrence is estimated at 5% [16]. If further pregnancies are planned a delay of 12 to 24 months between pregnancy on cesarean section scar and future pregnancy is recommended [9]. In a case series by Wei et al the reproductive outcomes for women with history of CSP were followed up, spontaneous pregnancy rate was 74.0% and the recurrence rate was 14.3% [17]. Also, Ben Nagi et Al shows that reproductive outcomes following Caesarean scar ectopic pregnancies are favorable, 83% of women were able to achieve subsequent pregnancies with a median time interval between previous scar ectopics and new conception of 5.3 months (range 1–48 months). 5% had a recurrent CSP [18].

Conclusion

CSP is an uncommon form of ectopic pregnancy that can result into life threatening complications if not diagnosed and managed early. In our case there was a high clinical suspicion for a CSP in a patient with a history of cesarean deliveries presenting with first trimester bleeding.

Diagnosis and management of CSP could sometimes be challenging and requires a multidisciplinary approach. Laparoscopic management is considered as a safe option with shorter operation - and hospitalization time, less intraoperative bleeding, faster recovery.

In order to reduce the incidence of this iatrogenic entity, the reduction in the number of primary CS performed without medical indication is necessary

Abbreviations

CSP: Cesarean scar pregnancy

CS: Cesarean Section

HCG: human chorionic gonadotropin

MRI: Magnetic Resonance Imaging

UAE : Uterine artery embolization

Acknowledgements: None

Authors' contributions

Ahmed Mimouni performed the surgery as a main surgeon. The manuscript was prepared by Hind Ennasser. Imane Skiker analyzed and interpreted the MRI. Hanane Saadi and Hafsa Taheri were the second and third surgeon.

All authors read and approved the final manuscript

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Ethics approval and consent to participate

The privacy of the patient was considered, and the manuscript does not include any identifying information.

Consent for publication: Informed consent for publication of the patient's clinical data and the accompanying/ images was obtained from the patient.

Competing interests: The authors declare that they have no competing interests.

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Antenatal ultrasound diagnosis of 'Iniencephaly Apertus'

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Abstract

Iniencephaly is a rare neural tube defect characterized by extreme retroflexion of the head with absence of the neck due to spinal deformities. On antenatal ultrasonography, features that clinch the diagnosis of iniencephaly include deficit of occipital bone with an enlarged foramen magnum, fusion of malformed cervical and thoracic vertebrae, and upward turned face with chin being continuous with the chest because of absence of neck. Differential diagnoses include anencephaly with spinal retroflexion, Klippel-Fiel syndrome, nuchal tumours (e.g. teratoma, goitre, and lymphangioma), and Jarcho-Levin syndrome. We present a case of antenatal diagnosis iniencephaly apertus with discussion of the ultrasound features and postnatal clinical and radiological correlation.

Key words: anencephaly, Iniencephaly apertus, star gazing foetus

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Introduction

Iniencephaly is a rare birth defect characterised by the triad of fixed retroflexion of head, variable degrees of cervical lordosis and dysraphism, and an occipital bone defect involving the foramen magnum.[1] Two types have been described based on the associated presence or absence of encephalocele: Iniencephaly apertus and Iniencephaly clauses, respectively.[2] Although, considered by many as invariably lethal, there are at least 8 case reports describing long-term survivals with less severe forms of disease.[3] Herein, we describe antenatal ultrasound findings in a case of iniencephaly apertus.

Case report

A 33-year-old multigravida woman visited our obstetric outpatient department for routine obstetric consultation at 15 weeks of gestation. She had neither had prior antenatal check- up nor any folic acid supplementation. She had been a hypothyroid for 4 years and was on thyroxine supplementation (50 mcg once daily). There was no h/o consanguinity. There was no history of teratogen exposure.

Patient's obstetric history was G6P1A4L1. First pregnancy had delivered full-term, low birth weight, male child weighing 1.75 kg. Second pregnancy required medical termination of pregnancy (MTP) because of intrauterine foetal demise. Third, fourth and fifth pregnancies had ended in spontaneous abortions at 6-, 6- and 7-weeks gestation, respectively. However, she had never undergone evaluation mainly because of financial issues.

Her blood group was A-positive. Laboratory tests including complete blood picture, blood sugar, urinalysis, HIV, HBsAg and VDRL were normal. She was euthyroid with high-normal serum TSH level (3.0 mIU/L). Positive serum titres of IgG against toxoplasma and rubella were found. Ultrasonography was performed (GE Voluson using broad band 2-5 MHz convex transducer and 4-8 MHz convex volume transducer). Gray scale scan revealed foetal intrauterine growth retardation. Neck was short and hyper-extended with fixed spinal retroflexion (Figure 1).

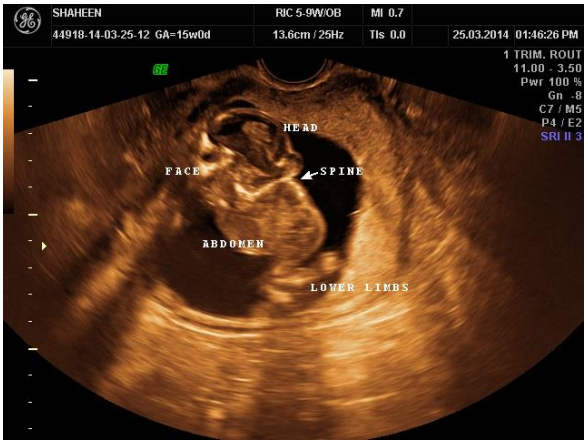

Figure 1: *On ultrasound scan, Fetus could be described as "stargazer" with upturned face, deficient short spine with hyperextended neck and fixed spinal retroflexion.*

There was fusion of upper cervical vertebrae with the occiput, along with approximately 10 mm occipital encephalocele (Figure 2). Spine was very short, and lower thoracic and lumbosacral vertebrae could not be visualized indicating complete rachischisis (Figure 3).

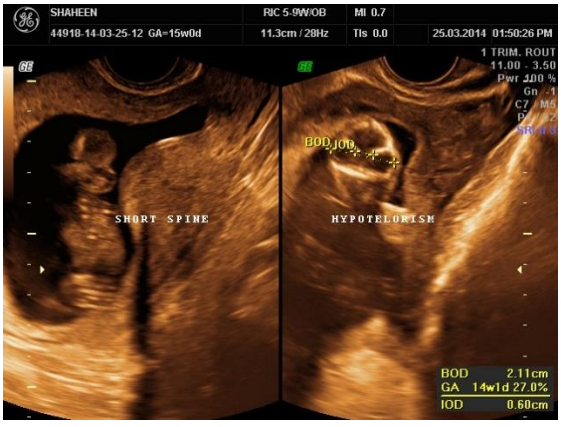

Figure 2: *There was fused occiput with upper cervical region and a 10 mm occipital encephalocele suggesting Iniencephaly Apertus*

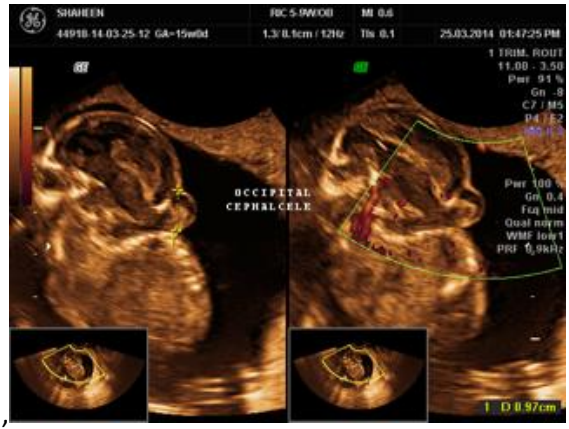

Figure 3: *The cervical and thoracolumbosacral spine was short, deficient and vertebrae could not be clearly differentiated with deficient overlying soft tissues/skin- Complete Rachischisis*

Facial profile appeared abnormal with hypotelorism. Hands and feet appeared normal. However, the foetus did not demonstrate any limb activity during the examination period of 30 min with possible foetal akinesia. An antenatal diagnosis of iniencephaly apertus was made. In view of obvious foetal anomalies incompatible with extra-uterine life, the parents were counselled for MTP. No further specific imaging or testing was done. After giving consent, patient underwent MTP.

Examination of the abortus revealed a dead female foetus weighing 32 g. Antenatal USG findings of occipital encephalocele, retroflexion of head (Figure4), and rachischisis (Figure 5) were grossly confirmed. Radiograph (fetogram) showed an abnormal short spine with hemivertebrae and open spinal defect (Figure 6). The limb bones were normal. Karyotype analysis revealed normal chromosomal pattern.

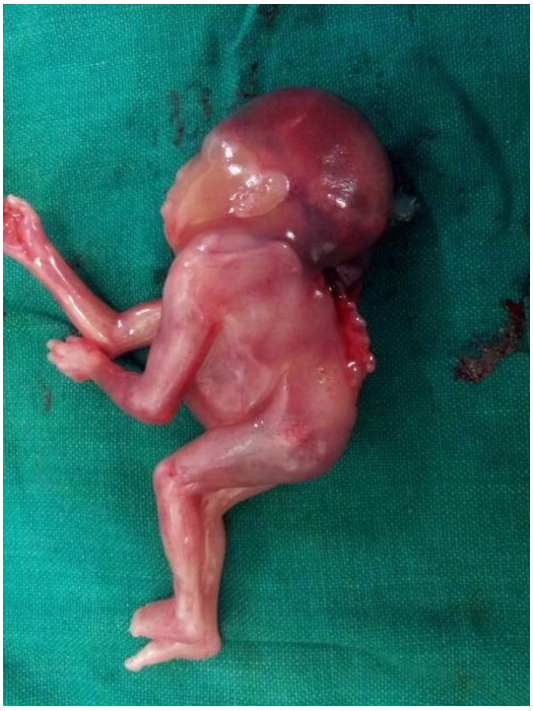

Figure: 4

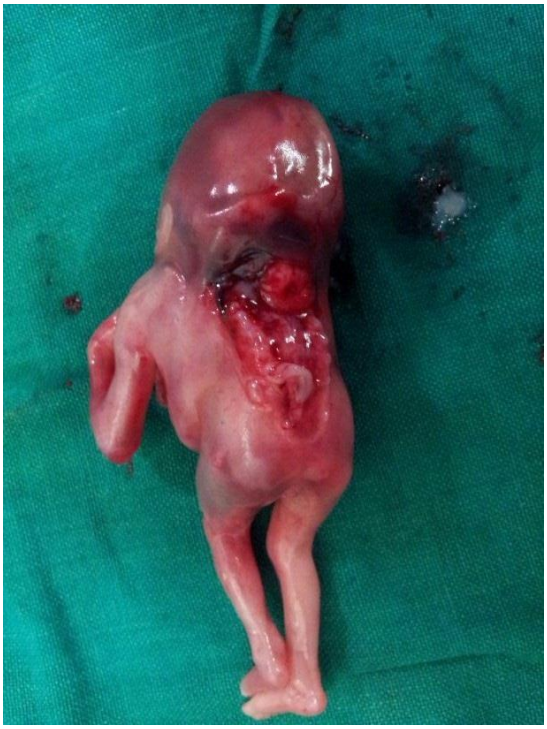

Figure: 5 The antenatal findings were seen in the gross postnatal fetus on examination in lateral position, figure 4 – in prone position 5

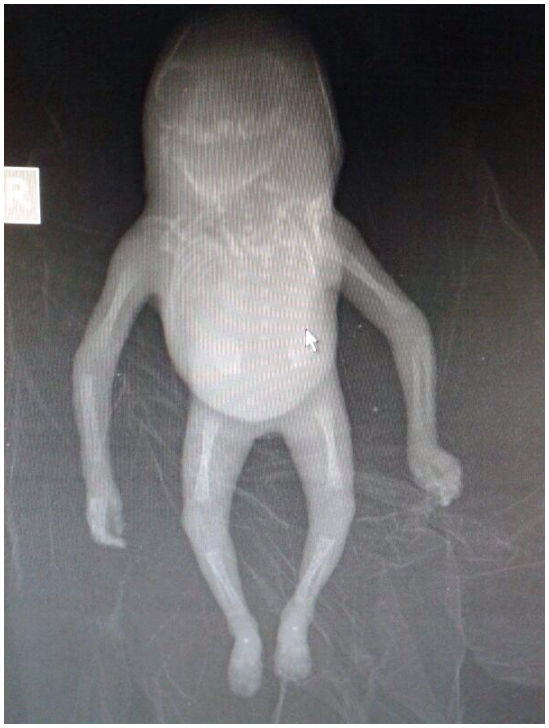

Figure: 6 *Radiograph (Fetogram) revealed abnormal undifferentiated deficient vertebrae from cervical to sacral region*.

Discussion

Iniencephaly is an uncommon neural tube defect (NTD). The word 'inien' is derived from the Greek word 'inion' which means nape of neck. The defective 'inion' (posterior most part of occipital bone) fuses with the back resulting in absence of neck and fixed retroflexion of head.4 Incidence in India is estimated to be 1 in 65,000 deliveries.[5] Overall reported incidence is higher and ranges from 0.1 to 10 in 10,000 deliveries.[6] Majority of cases are females (90%) 5 as was our case. The exact etiology and pathogenesis is unknown. Environmental factors that increase the risk of NTDs in foetus include poor socioeconomic conditions, low parity, lack of folic acid supplementation, obesity and drugs including sulphonamide, tetracycline, antihistamines and antitumor agents.[7] Iniencephaly has been reported in association with few chromosomal abnormalities including trisomy 18, trisomy 13, and monosomy X.2 Environmental factors like maternal syphilis and drugs like clomiphene citrate and sedatives have been also implicated in causation of iniencephaly.[6] However, no definite evidence exists in favour of any specific genetic and/or environmental factors being the cause (s) of iniencephaly in particular.

In our case, poor nutrition and lack of folic acid supplementation during pregnancy may be the risk factors. Further, the patient was suffering from hypothyroidism with long-term history of thyroxin intake. Also, she was tested positive for IgG against toxoplasma and rubella, which indicates prior infection. However, we could not find any established evidence of association between these factors and risk of iniencephaly.

Antenatal diagnosis of iniencephaly mainly relies on USG, and/or magnetic resonance imaging (MRI).[8] Although, in most cases a reliable diagnosis can be made on USG alone, MRI might help in cases of suspicious but atypical findings on USG. On USG, the characteristic appearance of iniencephaly has been described as a 'star-gazing foetus'. Other features that help in the diagnosis include:

1. Occipital bone deficit leading to enlarged foramen magnum without (iniencephaly clausus) or with encephalocele (iniencephaly apertus)

2. Irregular fusion of malformed vertebrae

3. Incomplete closure of vertebral arches and bodies

4. Retroflexion of the cervical spine

5. Upward turned face with chin continuous with chest because of the absence of neck

Our case had all these features, and thus, was diagnosed as iniencephaly apertus. Iniencephaly apertus should be differentiated from anencephaly with retroflexion of spine. Anencephaly shows a total or partial absence of neurocranium and retroflexed head is not covered with skin. However, in iniencephaly the retroflexed head is completely covered with skin. Cervical vertebrae are abnormal in iniencephaly, but almost normal in anencephaly. In the present case the retroflexed head was completely covered with skin and there was no encephalocele.

Iniencephaly carries bad prognosis, with the classic malformation-complex being incompatible with extra-uterine life. However, few instances of long-term survival have been reported in cases of lesser severity of iniencephaly & associated malformations.7

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casereport

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Learning for our complication

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Keywords: bleeding, via false, hysteroscopy

Introduction

At our reference centre of hysteroscopy in Constantine Algeria we perform more than a thousand hysteroscopies per a year most of these are office procedures.

The complication rate for all the procedures is about 05 per thousand failures and 15 per thousand incomplete diagnosis and operative procedures, 04 cases of vagal reactions had to be managed, 59 per thousand have had a painful hysteroscopy up to a score of 07/10 (Analogue Visual score 1 – 10 where the patients rate there pain) no premedication nor analgesics were used, the local vocal was the sole method applied (1).

All the procedures were performed by vaginoscopy (2) using 05 mm and 04mm Bettocchi hysteroscopes a Gubbini 14.5 French resectoscope and 05 French bipolar needle, also hardly ever a resectoscope of 18,5 was used when the cervix allowed an easy passage.

Case report

A very rare complication is described as a case report.

A woman aged 41, married with a story of one caesarean section four years ago and one year later she had an incomplete abortion followed by a curettage, a false route was made accidently during the curettage as described in her operatory protocol.

From that time the patient had probably developed synechia because her symptoms of hypo and oligomenorrhea. An hysterosalpingography was performed one year ago and failed because of the possible passage in the old false route. Her ultrasound showed a normal shaped uterus where the endometrial lining is seen. She was referred to the centre in order to treat the possible synechia (3),

At first, on day nine of her cycle, an office hysteroscopy was performed and the scar of the cervical false passage was clearly visible. However, a successful passage into the uterine cavity through the cervical canal averred to be possible. The endometrium was clearly seen and

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the uterine cavity did look tunnel shaped without seeing the two ostia possibly covered by the evident fibrotic tissue. The procedure was stopped when the patient started to feel some pain when a trial to cut this fibrotic tissue was started.

Four months later (because of Covid-19 pandemic) she was scheduled for hysteroscopy under spinal anaesthesia.

At the theatre a Bettocchi five mm hysteroscope was used and after getting into the cervical canal the scope did pass directly into and through the false passage to finish outside the uterus into the peritoneal cavity as witnessed by the intra pelvic fatty tissue clearly visible (4).

The procedure was stopped immediately and the patient was followed for 24 hours and she was discharged home in a very good clinical condition.

Seven days later the patient came back with an abnormal vaginal bleeding, vaginal and pelvic ultrasound did show a big hematoma in the broad ligament measuring some 15cm over 10 cm. the patient was hospitalized for a close follow up, her Haemoglobin (Hb) was at 11mg/dl at intake. She was placed under tow different antibiotics (cefalexin and metronidazole) since her first hysteroscopy.

One day later early in the morning the team was called in for an important vaginal bleeding and a low blood pressure, her Hb was 07mg/dl and the clotting tests were normal. The abdominal and vaginal ultrasound revealed no hematoma at all, so a decision to go to the OR and open the patient (the centre does not allow to perform laparoscopy when the blood pressure is down) was made. through the ancient Pfannenstiel the patient was opened and after adhesiolysis the peritoneum of the left broad ligament has just a bluish colour with the same findings behind the bladder. A bilateral uterine artery ligation was performed and an unusual cervical cerclage with Vicryl 1 in order to close the possible false route, the uterus had a normal aspect and a clear opening of the perforation couldn't be found, a drain was brought in. A transfusion by three units of blood was made.

The follow up was normal for three days, and the patient was discharged home in a satisfactory clinical condition. A vaginal ultrasound was done three and five days after the second surgery and the situation did remain normal.

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Videoarticle

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Hemorrhagic ascites and pleural effusion: an uncommon presentation of endometriosis

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Case report: We present a case of a 29-year-old woman with a history of primary infertility and laparoscopic excision for recurrent ovarian endometrioma. She presented to the emergency department for asthenia, abdominal swelling and pain. On examination she was stable with signs of massive ascites and right-sided pleural effusion.

This was confirmed on ultrasound scan and computed tomography. Paracentesis was done reveling a bloody ascetic fluid. She declined a pleurodesis for the pleural effusions. The patient was treated with a GnRH analogues.

Conclusion: This case highlights the importance of evoking endometriosis in reproductive-age women presenting with a massive ascites, with or without pleural effusions in differential diagnosis. Treating these patients can be difficult as they are usually of childbearing age and nulliparous and so wish to preserve their fertility based on conservative surgery and suppressing ovarian function with mainly GnRH agonist

Keywords: Endometriosis, hemorrhagic ascites, pleural effusion, hemothorax

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Video Article

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Laparoscopic management of a case of accessory cavitated uterine mass (ACUM)

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Keywords: laparoscopy, juvenile cystic adenomyoma, accessory cavitated uterine mass

Abstract

Study Objective: To demonstrate a technique of laparoscopic management of accessory cavitated uterine mass, along with a clinical, imaging and histopathology co relation.

Design: A step by step description of the surgery using an instructional video Setting: Patient: Twenty-seven-year-old woman married for 1 year with complaints of severe dysmenorrhoea. Ultrasound showed a hype echoic mass lesion in the uterus, likely to be a broad ligament fibroid, adenomyoma or a functional non communicating uterine horn.

Intervention: Diagnostic hysteroscopy revealed a normal uterine cavity. Diagnostic laparoscopy revealed a mass approximately 5 cm in size in the right lateral wall, below the right round ligament. There was no distortion of pelvic anatomy by endometriosis. The mass was infiltrated with diluted vasopressin solution and excised completely. Chocolate fluid in the mass was drained during removal. The myometrial defect was sutured in two layers, and covered with an adhesion barrier membrane. Histopathology revealed features specific to endometriosis. Since all aspects of the ACIEN criteria diagnostic of ACUM were satisfied, a retrospective diagnosis of ACUM was made.

Measurements and main results: Intra operative blood loss was 20 ml, and intra operative time was 90 minutes. The patient had complete relief of dysmenorrhoea from the very next menstrual cycle.

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Conclusion: ACUM (previously called Juvenile Cystic Adenomyoma, JCA) is a rare congenital uterine malformation that usually presents in young women less than 30 years of age. Common presenting symptoms are dysmenorrhoea and chronic pelvic pain. Treatment is laparoscopic excision. Diagnosis is confirmed by the presence of a normal uterus, absence of pelvic endometriosis, and the identification of endometriotic tissue on histopathology.

Erratum

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Article: Page (25-30)

Intra-ovarian direct trocar penetration and drainage for access prior to laparoscopic surgery for giant ovarian cyst

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