

Literature review of vaginal leech infestation and the use of vaginoscopy during its management

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Abstract: There are few case reports in the world literature in English of vaginal leech infestation in pre-pubertal, menstrual and post-menopausal females. The usual presentation is painless continuous vaginal bleeding. Blood transfusion was required in half the published case reports. Vaginoscopy seems to be underused, as there is only one recorded case report of its use during the management of vaginal leech infestation in a teenage girl.

Key words: Vaginal leech infestation; Painless vaginal bleeding; Pre-pubertal vaginal bleeding.

LITERATURE REVIEW

Leeches are annelids (segmented worms) in the subclass Hirudinea. They can be found in most areas in Australia in both damp land and aquatic habitats. Australian leeches have two jaws and produce a V-shaped wound. Leeches of the *Gnatobdellida* genus are sanguivorous and release hirudin, an anticoagulant protein which acts via direct thrombin inhibition¹.

There are few case reports in the world literature in English of vaginal bleeding secondary to leech infestation in pre-pubertal, menstrual and post-menopausal females. They generally describe continuous, painless passage of bright blood per vagina. In 1968, was the first recorded case of a 45 year Bulgarian woman who had haemorrhage secondary to a leech in the vaginal wall². The severity of bleeding reported varies among case reports from mild asymptomatic bleeding to significant anaemia. The most recent publication in 2013 describes two cases of profuse vaginal bleeding and hypovolaemic shock in women aged 21 years and 20 years in South India³. Both women had histories of swimming in natural bodies of water and each presented after a number of days with profuse, bright vaginal bleeding and haemodynamic compromise. Their haemoglobin levels were 60g/L and 50g/L respectively and they both required blood transfusion. Rao et al described the need to pack the vagina to stop the bleeding in 48 years old woman⁴. A 45 years old woman had an unnecessary hysterectomy due to failure to diagnose the vaginal leech as the cause of her vaginal bleeding⁵.

The duration of bleeding varies between reports from a few hours to days and even a report of four months of continuous vaginal bleeding. Patient demographics and clinical details of other reported cases, including the duration of bleeding, haemoglobin, method of diagnosis and treatment of vaginal leech are summarised below (Table 1).

The usual method of removing leeches involves application of salt to the skin or disruption of the attached jaw by sliding a sharp object or fingernail under the point of attachment. However, when the leech embeds within the vagina, this makes both diagnosis and management challenging. A number of cases report irrigation of the vagina with normal saline to dislodge a leech and one case describes a method of injection of the leech with lignocaine.

Previously, there were four cases published in world literature of vaginal leech bite in pre-pubertal girls, two of them required EUA and none used vaginoscopy during the diagnosis⁶⁻⁸. The examination of pre-pubertal children poses difficulty due to a small vaginal orifice, unestrogenised vaginal tissue susceptible to trauma and an intact hymen. Prior to this case, there were no reports of the use of vaginoscopy under anaesthesia for diagnosis of leech bite in a pre-pubertal

child and there are no published photos of the vaginal lesion appearance caused by a leech bite. In 1983, Vallerino et al reported the use of vaginoscopy in diagnosis of vaginal leech in a teenage girl⁹.

VAGINOSCOPY

Vaginoscopy seems to be underused in this clinical situation, as there is only one recorded case report of its use during the management of vaginal leech infestation in a teenage girl.

The examination of pre-pubertal children poses difficulty due to a small vaginal orifice, unestrogenised vaginal tissue susceptible to trauma and an intact hymen. There are no reported cases of the use of vaginoscopy in pre-pubertal girls.

We report the first use of vaginoscopy under anaesthesia for the diagnosis of leech bite in a pre-pubertal girl.

An 8-year-old pre-pubertal female underwent air evacuation from a remote Northern Territory community to Royal Darwin Hospital (RDH) for heavy vaginal bleeding. History revealed that she had been swimming in a local billabong and a number of had removed a leech from her vaginal introitus after experiencing a non-painful sensation. Following this event child experienced continuous bright vaginal bleeding of moderate amount. This was noticed by her aunt who sought review at the community primary

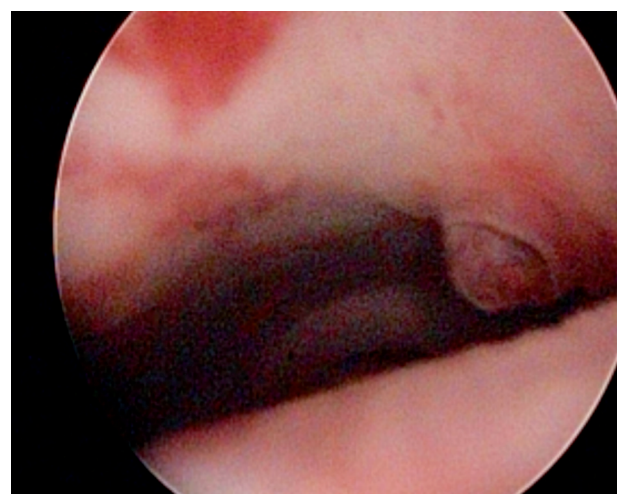


Figure 1. – Vaginoscopy revealed a 3 – 4 mm ulcer on the left side of anterior vaginal wall caused by vaginal leech (the ulcer is seen on the right side of the photo). The anterior lip of the cervix is seen (above the ulcer) at the centre of the top of vagina.

TABLE 1. – All cases of vaginal leech published in World Literatures in English up to August 2015. EUA: Examination Under Anaesthesia - Hb: Haemoglobin – BT: Blood Transfusion.

Publication	Age	Bleeding duration	Hb (g/L)	BT needed	Method of Diagnosis and Management
Senthilkumaran et al. 2013 (3)	21	2 days	60	Yes	EUA showed 3 live, engorged leeches removed with forceps. Initial treatment with oral contraceptive pill unsuccessful. EUA showed 2 live leeches removed with surgical forceps.
	20	6 days	50	Yes	
Rao et al. 2010 (4)	48	1 day		No	Per vaginal speculum examination showed a large leech extruding from the introitus that was dislodged with vaginal irrigation of NaCl. Vaginal packing of wound due to ongoing bleeding.
Shamdeen. 2007 (5)	45	4 months	70	Yes	Initially presumed to be uterine bleeding and managed unsuccessfully with oral progestagen and diagnostic curettage. Proceeded to total abdominal hysterectomy but ongoing bleeding post-operatively. EUA on day 5 post operative revealed a leech that was dislodged by injection with lignocaine and removed with sponge forceps.
Habtai et al. 2009 (6)	6	4 days	100	No	Saline vaginal irrigation and removal of leech with forceps.
Saha et al. 2005 (7)	5	2 hours	95	No	EUA and exploration of vagina with nasal speculum showed a live leech removed with surgical forceps. Vaginal packing of wound due to ongoing bleeding.
	7	14 hours	75	Yes	
Ibrahim et al. 2003 (8)	9	24 hours	73	Yes	EUA with vaginal irrigation of NaCl via small feeding tube and extrusion of a leech. Vaginal packing for ongoing bleeding. Vaginal irrigation with 20ml NaCl and extrusion of a leech. Bleeding continued for 24 hours after removal of leech and was managed conservatively.
Vallerino et al. 1983 (9)	14	8 days	110	No	Leech crawled out from vagina after admission to hospital. Bleeding ceased immediately. Vaginoscopy showed 1cm ulcerated lesion on posterior vaginal wall.

health clinic. There was no visible trauma to the external genitalia noted by the clinic staff and bleeding continued despite fifteen minutes of application of pressure with gauze pack to the introitus. In response, the clinic organised transfer of the child and her aunt to RDH for further evaluation and management.

On history taking at RDH there were no inconsistencies between the child's account and that of her aunt and no suspicion of child sexual abuse. The amount of blood loss was unable to be quantified and there was no report of dizziness, dyspnea or other symptoms of anaemia. Examination revealed the child was pre-pubertal (Tanner Stage 1) and of slight stature with weight 26kg. Her vital signs were within normal limits and her abdomen was soft and non-tender with no palpable masses. Examination of the external genitalia was unremarkable other than a large blood clot at the introitus, which was dislodged, causing a continuous trickle of bright blood per vaginum. It was therefore decided to proceed with examination under anaesthesia (EUA) and vaginoscopy. This occurred 24 hours after the child first noted the bleeding. The patient's haemoglobin was 111g/L on admission.

Findings from EUA were normal external genitalia with intact hymen and no evidence of injury. A 4mm, 30-degree Hysteroscope with irrigating sheath was used to perform vaginoscopy with Normal Saline distension of the vaginal. A large abdominal pack was held against the introitus to reduce saline leak to achieve adequate distention of the vagina. Good view was achieved, there was a 3-4 mm lesion in the form of an ulcer on left side of anterior vaginal wall at the proximal third of the vagina. The lesion was consistent with a leech bite and was not actively bleeding (Fig. 1). We provide the first ever photo showing the appearance of vaginal lesion caused by a leech bite.

Although not needed in this case but the saline jet used during vaginoscopy may be very helpful in washing out a leech that may be seen inside the vagina during the procedure.

This is the first published account of vaginal leech bite in Australia, a setting where leeches occur in all areas of the country that are non-arid.

CONCLUSION

Vaginal leech infestation is a rare event. It should be considered when encountering women especially young girls presenting with painless vaginal bleeding without history of trauma. The use of saline vaginoscopy should be considered when vaginal leech infestation is suspected especially in pre-pubertal girls.

STATEMENT

There was no conflict of interest.

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Surgical treatment of a patient with “enteroptosis” (prolapse of uterus, vagina, bladder, rectum)

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Abstract: Abstract: A 81-year-old patient with prolapse of uterus, vagina, bladder and rectum (enteroptosis) was cured anatomically and functionally by an entirely vaginal operation. During the same session all deficient connective tissue structures of the pelvic floor were repaired according to the Integral Theory. This case with excellent results demonstrate a less invasive alternative to classical surgery.

Key words: Rectal prolapse; Uterine prolapse; Enteroptosis; Integral Theory; Sacrospinous fixation; Total pelvic reconstruction.

INTRODUCTION

Understanding the anatomical basis for rectal and uterine prolapse formation is fundamental to plan surgical repair thereof. Rectocele and rectal intussusception repair has been influenced by the Integral Theory¹. The theory states, that abnormal bladder symptoms, abnormal bowel symptoms and vaginal prolapse are related and mainly caused by connective tissue defects in 3 zones of the vagina. A normal defecation is only possible if the anterior rectal wall is stretched. With a defect in the rectovaginal fascia (RVF) or uterosacral ligaments (USL), the anterior rectal wall can not be stretched sufficiently. The stool can not escape directly to the anus, because faeces reaches first the dilated anterior rectal wall to be decelerated in the bulge. The increased pressures in the wrong direction overstretched the RVF further and generates a rectocele. The evolving outward rectocele pulls the proximal rectal wall down inside. This leads to invagination of the wall, to intussusception, and lastly to internal and external rectal prolapse. Clinically this manifests itself in incomplete emptying, symptoms of abnormal anorectal closure, rectal intussusception, external hemorrhoids or – as in our case – anorectal prolapse. Because all structures, rectum, vagina, suspensory ligaments, fascia and muscle forces work synergistically, all deficient connective tissue structures need to be repaired for adequate restoration of anatomy and function.

PATIENT AND METHOD

A 81-year-old patient came with complaints of a palpable mass in the vagina and anus, obstructive defecation symptoms, rectal bleeding, stress and faecal incontinence. She had a uterine and posterior vaginal wall prolapse III^o according to the halfway classification system, an entero/rectocele III^o and a full-thickness rectal prolapse (Fig. 1). On admission she was asked to fill a questionnaire developed by P. Petros².

According to this questionnaire she suffered from

- discomfort and pain due to prolapse dragging
- nocturia (>2 per night)
- bladder emptying problems
- stool outlet obstruction
- discharge of mucus and blood from the anus
- faecal incontinence
- urine incontinence.

Therapy

1. In consideration of replacing the rectal prolapse and normalizing the anatomy of the posterior wall we performed a posterior IVS operation in combination with bilateral

sacrospinous tape fixation, a posterior bridge repair and a perineal body repair.

2. In order to restore the uterine and anterior vaginal wall prolapse we used an anterior 4-arms mesh. The anterior 2 arms we inserted transobturatorially. Then the cervix was surrounded by the posterior 2 arms, which were fixed to the sacrospinous ligament both sides.

3. Concerning the stress incontinence we inserted a sub-urethral transobturatorial sling.

Local estrogens were administered six week before surgery and a proper informed consent was obtained for the operation.

Procedure in detail

Level I repair: After local injection of bupivacain with adrenalin a transverse incision approximately 4-5 cm long was made in the posterior vaginal wall 1,5 cm below the cervix and opened antero-posteriorly. A Sims speculum was inserted to open out the incision. A rectal examination was performed to identify the limits of the rectocele and enterocele. Adherent enterocele was freed from adjoining tissues so as to avoid damage by the tunneller. The laterally displaced uterosacral ligament remnants were localized. The enterocele sac was reduced with a pursestring suture.

With a digital blunt preparation the sacrospinous ligament was freed from adhearent tissue and 2 prolene sutures 2-0 were inserted into the ligament on both sides with a sacrofix device according to Goeschen.

Bilateral incisions 0.5 cm long were made in the perianal skin at 4 and 8 o'clock, halfway between the coccyx and the



Figure 1. – Uterine and posterior vaginal wall prolapse III^o combined with a full-thickness rectal prolapse.

external anal sphincter (EAS) in a line 2 cm lateral the EAS. Starting at the left side the IVS Tunneler was placed into the ischioanal fossa for a distance of 6 cm. Under direct vision, with a finger placed into the rectum, the tip of the tunneler was gently pushed through the levator plate, inclined medially towards the vaginal vault by rotating the delta wing and brought just behind the uterosacral ligament. The tip penetrated through the recto-vaginal fascia and reached the transverse incision. 1 prolene suture from each side was stiched through the middle of the tape in a distance of 4 cm.

Then the procedure was repeated on the contralateral right side, leaving the tape as U entirely unfixated at the sacral end. The tape was secured to the vaginal vault, also to the remnants of the uterosacral ligaments and the cervix with interrupted no. 1 vicryl. The tape was then gently stretched by pulling on each perineal end.

Level II and III repair: Repair of the anterior vaginal wall: After local injection of bupivacain with adrenalin a full thickness elliptical incision, 3 cm wide, over the herniation of the cystocele was made, extending from 2 cm distal of the bladder neck to the cervix. The cut edges of the incision were grasped with Allis forceps and the space between bladder and vaginal wall was opened up with a scissors and blunt dissection up to the arcus tendineus fasciae pelvis (ATFP).

Extensive diathermy was used to destroy the superficial vaginal epithelium overlying the bridge. The anterior part of the bridge was anchored by burrowing 0.5 cm below the anterior border of the incision, the posterior part in the cervix. The cystocele was reduced with 4 mattress sutures extending from one to the other side and attached to the bridge in two layers.

Anterior transobturator mesh support: Because of the poor tissue a ribbon of polypropylene mesh (Premilene LP) 5 cm wide, 30 cm long was cut in a figure with two arms on each side. The anterior two arms of both sides were placed with a special instrument between the pubococcygeous muscle and the vaginal skin, running from the arcus tendineus of one side to the arcus of the other side and pulled out transobturatorially. It was placed without tension across the midline defect to reinforce the weakened tissue. The mesh was sutured with Ethibond 0 lateral to the pubococcygeous muscle and to the cervix in order to get the mesh flat. The posterior two arms were placed around the cervix subepithelially and then connected with the remaining sacrospinous sutures, one right, one left. The pubocer-

vical fascia was narrowed with U-sutures to cover the mesh. The skin incision was closed by a continuous longitudinal no. 1 Dexon locked running suture.

Repair of the posterior vaginal wall: After aquadissection using bupivacain with adrenalin in the posterior vaginal wall two full-thickness parallel longitudinal incisions, 2cm wide, were made along the posterior vaginal wall, extending from the transverse incision to 1 cm distal to the introitus. Extensive diathermy was used to destroy the superficial vaginal epithelium overlying the bridge. The margins of the bridge were sutured with interrupted sutures resulting in a double layer bridge. Adherent rectum was freed from the vaginal wall and perineal body (PB) over the distal 3-4 cm of vagina. The rectocele was reduced by using laterally placed horizontal mattress sutures. These sutures run through the bridge and were tied without tension so that the rectum-tube had a normal width and strength. Two transverse vaginal holding sutures (1 PDS) were placed as laterally as possible, one at the level of the uterosacral ligaments and the other at the level of the rectovaginal fascia (RVF) and left untied. These sutures run subepithelially as a horizontal mattress suture through the bridge. The bridge was anchored separately at the tape above and at the perineal body below.

The horizontal limbs of the “Y” were sutured with interrupted no. 1 vicryl sutures, followed by a continuous longitudinal one. The four sacrospinous and the PDS sutures were tied only with so much tension, that was necessary to bring all pelvic organs in normal position. Finally one vicryl unlocked running suture was placed which approximated the lateral cut edges. The bridge was buried below the lateral flaps. The coccygeal and obturatorial ends of tape were cut, and left tension-free.

Paraurethral transobturator IVS: After local injection of bupivacain with adrenalin paraurethraly two full-thickness longitudinal incisions were made in the lateral urethral sulcus, extending from just below the level of the external urethral meatus to the level of midurethra. A suburethral tunnel was created between vagina and urethra. On the left side a small skin incision was made at the level of the clitoris. A special Deschamp was inserted transobturatorially into the paraurethral cut. Then an 8 mm polypropylene tape was threaded into the eye of the instrument, and pulled out through the skin cut. By inserting an Overholt forceps into the suburethral tunnel the tape was brought into the right contralateral vaginal incision. The tape insertion was completed on the contralateral right side. On both sides the



Figure 2. – Final result after the operation.

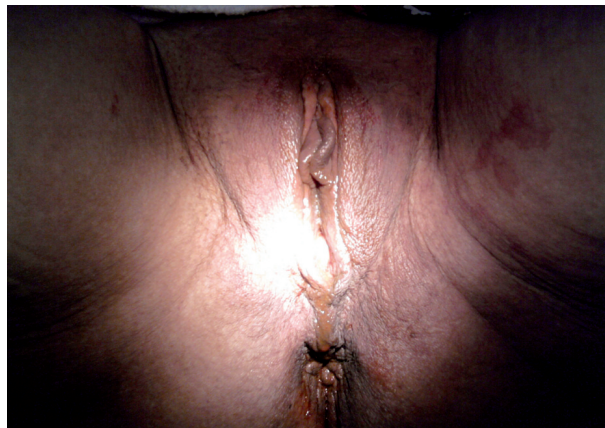


Figure 3. – Seven days postoperatively the anorectal and vaginal anatomy has already become normal.

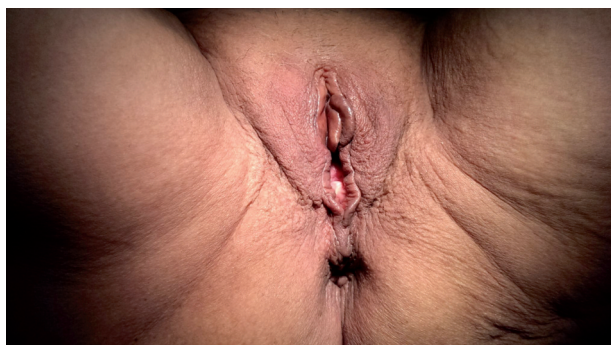


Figure 4. – Anatomical result one year after operation.

hammock was attached with vicryl No 1 to the anterior portion of pubococcygeus muscle. The incisions were closed with a continuous suture.

The bladder was filled with 300 ml fluid and the catheter removed. In order to avoid excessive tension and urethral constriction by the tape the tunneler was inserted into the urethral cavity when tightening the two limbs of the tape. While testing for continence by tapping on the bladder no urine leakage was observed.

RESULTS

Anatomy: The final anatomical result immediately after surgery is demonstrated in Fig. 2. Seven days postoperatively the ano/rectal and vaginal anatomy has already become normal (Fig. 3).

Clinical Course: The patient had no complications and only less pain during the hospital stay and later on. Bladder- and ano/rectal-function were normal without any signs of incontinence. Discharge from hospital was on the third day postoperatively.

Follow up: A follow up check was performed 3 months and one year after surgery. Both examinations showed a normal anatomy (Fig. 4.) In addition to the preoperative examination the patient was encouraged to fill the questionnaire again with the result that now bladder and anorectal function were physiological and the quality of life excellent.

DISCUSSION

Rectal prolapse is a rare entity which is formed by an intussusception of all rectum layers due to lax USL³. With lax USL levator plate cannot tension the RVF. The force of gravity and pressure from above during defaecation causes the rectal wall to prolapse inwards³ and finally throughout the anus (Fig. 5).

Despite its rarity more than 100 surgical procedures have been described and there are no good evidence based recommendations for these surgical procedures^{4,5,6}. According to Graf et al.⁷ the defecation improved only in 5%, fecal incontinence only in 16% after conventional operations. Overall good or excellent surgical outcomes were only achieved in 16% of all cases.

A recent publication from Koscinski et al.⁸ showed good anatomical and functional results after a combined abdomino/vaginal perineocolporectomy using a polypropylene mesh. We tried to cure our patient suffering from complex pelvic floor pathology entirely with an much less invasive vaginal approach.

Our patient had a combined prolapse of all pelvic organs (uterus, vagina, bladder, rectum). The better description for this situation is “evisceration or enteroptosis” as a result of weakening in the complete ligamentary holding system.

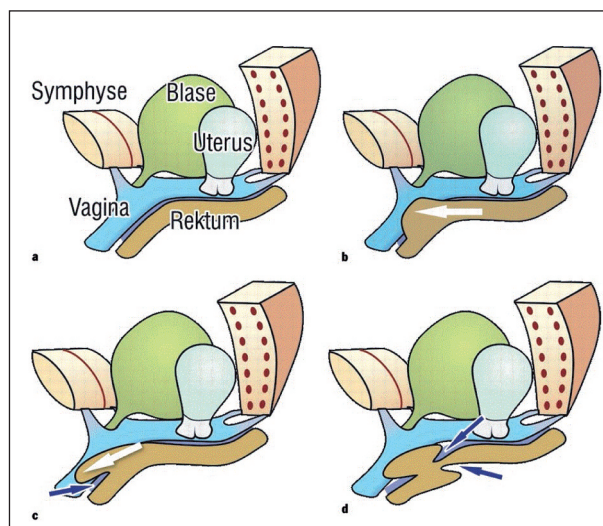


Figure 5. – Development of a rectocele, intussusception and rectal prolapse. a) With intact rectovaginal fascia and tight uterosacral ligaments the anterior rectal wall is stretched and faeces freely can pass through the rectum. b) If a defect in the rectovaginal fascia occurs the stool the anterior rectal wall to the front and slow down (white arrow). The patient must press increased, in order to empty the intestine. c) Due to increased pressure intensified the rectocele (white arrow), leading to the notch in the intact rectovaginal fascia (blue Arrow) and pulls at the proximal rectum. d) In case of defective uterosacral ligaments, the rectum can be curl inward, to narrowing of the lumen lead (blue arrows) and the increase constipation (9).

Therefore our aim was not to excise herniated tissue, but to repair all deficient connective tissue structures for adequate restoration of function according to the Integral Theory of Petros^{1,2}. This concept is consistent with the Baden and Walker (1992)¹⁰ developed tent theory. They have postulated that if a tent roof incident, the walls will follow.

The Integral Theory states that abnormal bladder and bowel symptoms as well as vaginal and rectal prolapse are related and mainly caused by connective tissue defects in the 3 zones of the vagina. Lax connective tissue structures invalidate the muscle forces involved in opening and closure of urethra and anorectum, leading to incontinence (abnormal closure) or retention (abnormal opening). Normal defecation thus is only possible if the anterior rectal wall is stretched.

The posterior vaginal wall is suspended between perineal body (PB), which underlies half its length, and USL, which also support the anterior wall of rectum^{1,2}. These structures are at least 6 times stronger than the vaginal or rectal mucosa which they support¹¹. Loose USL and laterally displaced RVF therefore leads to entero-/rectocele and anorectal prolapse in a high percentage.

Understanding these interrelationships we repaired all 3 zones of the vagina in our patient. In analogy to the tent theory we renewed the USL at level 1 and all defective structures in level 2 and 3. This stretched the RVF, eliminated the bulge of the anterior rectal wall and pulled the rectum back in normal position.

We added sacrospinous fixation of the posterior tape and the 2 cervical mesh arms for a higher apical support. Excision of excessive vaginal mucosa was avoided. This procedure is not able to cure a genital prolapse, but will only narrow and shorten the vagina. A short vagina cannot be properly angulated backwards and downwards around the perineal body and leads to bad functional results.

CONCLUSIONS

In the presence of intussusception or rectal prolapse, transanal, open or laparoscopic rectopexy, with or without resection, still is most popular. The Integral Theory states that these problems are mainly caused by connective tissue defects and can not be solved by excision of herniated tissue. We learned from our patient with enteroptosis (prolapse of uterus, vagina, bladder and rectum), that adequate restoration of all deficient connective tissue structures leads to perfect anatomical and functional results and can be reached only by a less invasive vaginal approach.

DISCLOSURE STATEMENT

We declare no conflict of interest.

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Multidisciplinary Colorectal Comment

To improve the integration among the three segments of the pelvic floor, some of the articles published in *Pelviperrineology* are commented on by **Urologists, Gynecologists, Proctologists/Colo Rectal Surgeons or other Specialists**, with their critical opinion and a teaching purpose. Differences, similarities and possible relationships between the data presented and what is known in the three fields of competence are stressed, or the absence of any analogy is indicated. The discussion is not a peer review, it concerns concepts, ideas, theories, not the methodology of the presentation.

The *tetralogy of fallout* is another interesting definition of the *total pelvic organ prolapse* or *enteroptosi* as the Author of this article names this quite incapacitating condition involving the descent of the anterior, central and posterior compartments of the pelvis in the female. Caliskan et al. describe a surgical procedure where all the three levels of pelvic support are considered and repaired. Conforming to the Integral Theory the rectal prolapse is due to a lax uterosacral ligament. Colorectal surgeons, who obviously face also in males the problem of the total rectal prolapse and of the rectal intussusception, usually perform sacrorectopexies through an abdominal approach or transanally resecting the prolapse (Altemayer procedure), or reducing it into the rectum itself (Delorme). It has been an interesting surprise for the colorectal surgeon applying the CR-

Mesh transvaginally (Farnsworth's procedure) in patients with the "tetralogy of fallout" to observe the complete and definitive cure of the rectal prolapse without any apparent surgical involvement of the rectum itself. But one has to be careful! Actually this technique works, but only in the cases where the rectal invagination starts at the level of the recto-vaginal septum, where the posterior mesh is placed. A rectopexy or any other posterior approach will be needed if the intussusception starts above it, like when the proximal rectum or even the sigmoid colon prolapses into the rectum all the way through the anus.

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