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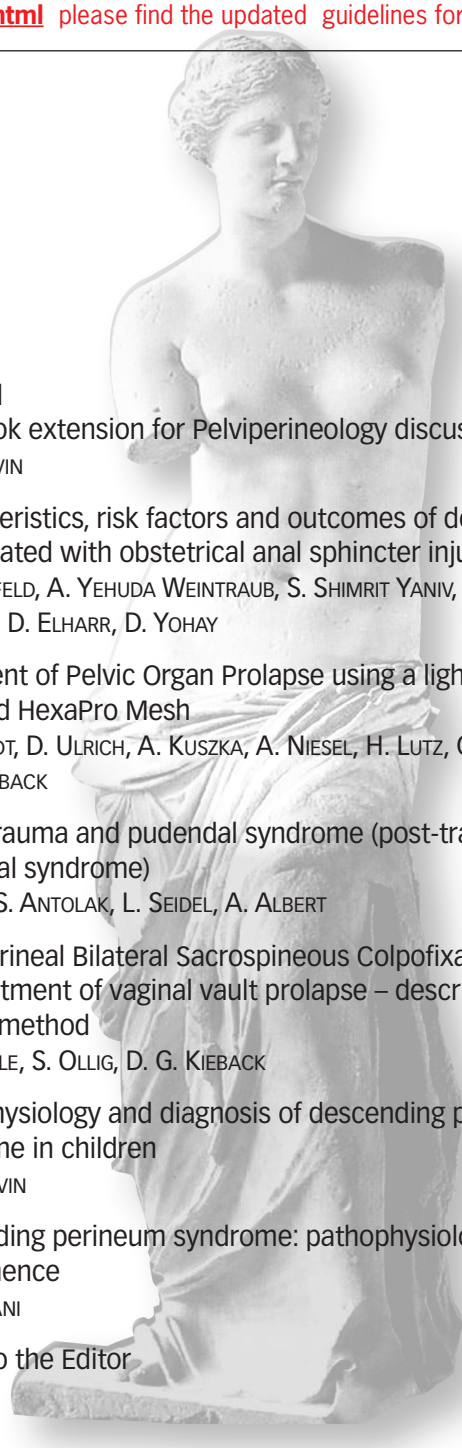

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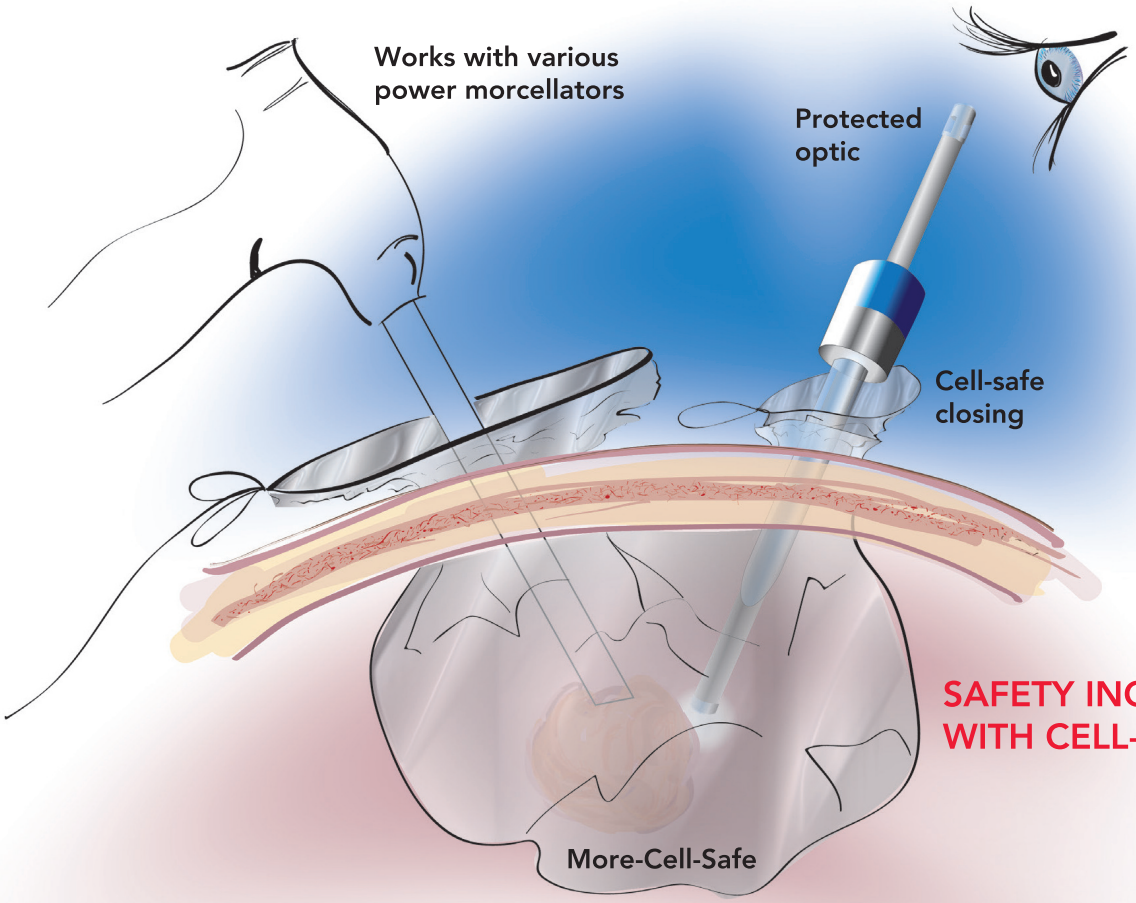

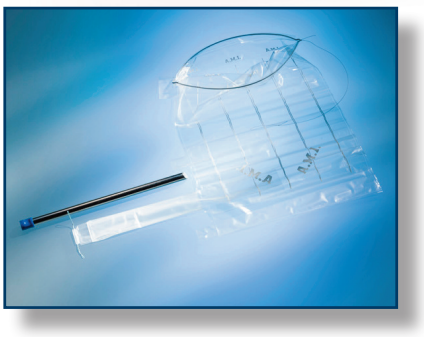

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Editorial

"Precise, almost mathematical knowledge of anatomy is a highly fertile source of surgical applications, suggesting new techniques and helping perfect and simplify existing surgical methods, making them less mutilating and more benign and, in short, raising surgery to the rank of true science".

SALVADOR GIL-VERNET (1893-1987)

Facebook extension for Pelviperineology discussion

MICHAEL D. LEVIN

State Geriatric Center, Netanya, Israel

Abstract: Invitation to participate in the discussion of published articles.

Keywords: Pelvic floor; Colon; Normal physiology; Anorectal pathology.

Dear colleagues!

As an extension to the discussion pages in Pelviperineology journal, we are creating a group in Facebook to discuss, analyse, debate, research on the pelvic floor and colon. Why? To return medical science to its basics, informed discussion, respectful, informed debate. It is the only way to counter what many of us perceive as an unhealthy retrograde step in specialist medical journals, to 'standardize' everything: endless definitions, 'consensus statements'. Opinion has replaced science. 'Definitions' have replaced scientific research. Conforming has replaced independent thought. Conformist scorn has replaced contrary opinions, the mainstay of traditional science. Journals have ceased to be a forum for discussion. They have become a parade of conformist thought. Because of this, science has become ossified. Outdated ideas continue to prevail, yet another casualty of the political correctness which is poisoning our society. Original thought, new concepts, the very engines of progress, inevitably come into conflict with the ossified concepts which these 'standardizations' bring. Needless to say, new ideas do not find their way into these 'traditional' journals.

The journal 'Pelviperineology' is an outstanding exception. Since its inception, it has welcomed new ideas and scientific criticism thereof. The group we are establishing in Facebook is an extension of this philosophy. We cordially invite all interested colleagues to join us in these open discussions.

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Please join our Anorectal pathology group on Facebook to participate in the open discussion of the article via the link:

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Original article

Characteristics, risk factors and outcomes of deliveries complicated with obstetrical anal sphincter injury

YAEL BAUMFELD[1,2], ADI YEHUDA WEINTRAUB[1], SALEM SHIMRIT YANIV[1], EFRAT SPIEGEL[1], ZEHAVA YOHAY[1], DEBBIE ELHARR[1], DAVID YOHAY[1]

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Abstract: Aim: Vaginal delivery is often complicated with perineal tears, mostly in the first and second degree, in severe cases there is damage to the anal sphincter with possible long term morbidity. Risk factors for severe perineal include nulliparity, previous severe perineal tears, macrosomia, prolonged second stage and instrumental deliveries. These risk factors are non-modifiable, the aim of our study was to explore the risk factors and seek for a modifiable risk factor. *Methods:* Our study included all deliveries that took at place in a 1000-bed tertiary teaching hospital between January 2011 and March 2016. A total of of sixty pateints sufferd a severe perineal tear at this time. *Results:* Our main findings included that the common risk factors for severe perineal tears were found in our study population, other important findings include a 39% rate of positive vaginal culture for Candida species, 15% of newborns had a first minute Apgar score<r 7 and 19% had a pH <7.1. *Conclusions:* Severe perineal tears have grave long term morbidity and with many known unmodifiable risk factors. We found an elevated rate of vaginal *candida* colonization in women with severe perineal tears and believe there may be a possible association.

Keywords: Obstetric complications; Anal incontinence.

INTRODUCTION

Vaginal delivery is often complicated with perineal tears which may be associated with short and long term morbidity. The majority of these tears are first and second degree tears-lacerations which extends the fourchette, perineal skin and vaginal mucosa to perineal muscles and fascia. However, on rare occasions the tear can be severe- with a disruption that involves the anal sphincter and even the rectal mucosa (third or fourth degree tears).

Perineal tears are associated with significant long-term morbidity and pelvic floor dysfunction which can lead to pelvic organ prolapse, urinary incontinence[8] and dyspareunia[24]. In cases of OASIS there may also be damage to the anal sphincter leading to flatulence and fecal incontinence[7,22,23]. Post-partum depression was reported in higher rates in these patients[21].

Having the considerable morbidity in young healthy women, many studies have explored the risk factors for severe perineal tears and OASIS. These include nulliparity, OASIS in previous deliveries, Asian race, increased fetal birth weight and head circumference, prolonged duration of the second stage of labor and instrumental deliveries[8-20]. Studies regarding episiotomy have contradicting results, while some have found it to be a risk factor[1,6] others did not find such an association or even found episiotomies to be protective against OASIS[2,17].

Most of these risk factors are non-modifiable. There may be some additional modifiable risk factors such as the work experience of the midwife birthing the patient that according to one study, was found to be associated with all degrees of perineal tears, but not with OASIS[25].

In order to reduce the rate of perineal lacerations an increase in cesarean deliveries was noted which too holds substantial risks to the mother[26].

Vaginal candidiasis has been associated with different pregnancy complications including preterm delivery, premature rupture of membranes and low birth weight[27]. The prevalence of candida was found to be as high as 15-30% in pregnant women using a vaginal swab which is an easy to use, low cost and painless test[27,28]. The treatment with locally administered antifungal agents is effective and safe during gestation[29].

Only few studies have explored vaginal flora's effect on perineal tears. One study found no association between bacterial vaginosis and perineal tears[5]. However, no studies to date, explored the association of perineal tears with vaginal candidiasis or other pathogens in the vaginal flora.

We evaluated the current case series in order to explore the characteristics, unmodifiable as well as some modifiable risk factors and perinatal outcomes of deliveries complicated with OASIS.

METHODS

Study population

This study is a retrospective cohort study of all women who gave birth between January 2011 and March 2016 in a 1000-bed tertiary teaching hospital. It is the only tertiary center for a population of 700,000 people. During the study period the average annual number of deliveries managed at our medical center was around 15,000.

Inclusion criteria for the study was a delivery complicated with a severe perineal tear.

Data was collected from the computerized database of all the deliveries and the infant hospitalization data. Data collected includes information on maternal baseline characteristics, co-morbidities and pregnancy complications; data regarding the delivery – including mode of delivery, length of first and second stages of birth, length of work experience of the midwife; the infant – including birth weight, Apgar scores, cord blood gas and complications; vaginal culture results and data regarding the suture of the severe perineal tear.

The study was approved by the Institutional Review Board Committee.

Clinical definitions

Perineal tear is a discontinuation in the integrity of the perineal tissues that usually occurs during childbirth. perineal tears are divided into four degrees according to severity.

First-degree tear: laceration is limited to the fourchette and superficial perineal skin or vaginal mucosa

Second-degree tear: laceration extends beyond fourchette, perineal skin and vaginal mucosa to perineal muscles and fascia, but not the anal sphincter.

Third-degree tear: fourchette, perineal skin, vaginal mucosa, muscles, and anal sphincter are torn; third-degree tears may be further subdivided into three subcategories:

3a: partial tear of the external anal sphincter involving less than 50% thickness;

3b: greater than 50% tear of the external anal sphincter;

3c: internal sphincter is torn.

Fourth-degree tear: fourchette, perineal skin, vaginal mucosa, muscles, anal sphincter, and rectal mucosa are torn. OASIS obstetric anal sphincter injuries are third and fourth perineal tears.

Data analysis

The data on continuous variables with normal distribution were presented as mean ± SD. Continuous variables not normally distributed and ordinal variables were presented as median with inter-quartile range (IQ range). Categorical data were shown in counts and percentages.

RESULTS

A total of 60 women suffered a severe perineal tear during the study period.

Baseline characteristics of the study population included a mean maternal age was 26.87±5.08. Regarding ethnicity, 54.2% (32) were Jewish and 45.8% (27) were Bedouin Arabs. The mean height was 162.79±5.89 cm and a mean weight of 77.00±14.73 kg.

The majority of subjects (39, 81.7%) were nulliparous. Mean gravidity and parity of the study population were 1.52±0.81 and 1.23±0.53, respectively.

Perineal tears characteristics

More than half of the tears (51%) were sutured by a gynecologist , while another third (30%) were treated by a general surgeon and 19.0% by a colorectal surgeon.

The suture took place in the operating room under general anesthesia in 50.9% and in the labor and delivery room under local or regional anesthesia in 49.1%.

The distribution of degrees of the perineal tears is presented in figure 1, it consists of ten cases of forth degree tears (16.9%), and the rest were third degree: 3a (47.5%), 3b (15.3%) and 3c (20.3%).

Midwives' characteristics

We investigated the midwives' years of experience. The mean length of work experience of the midwives was 10.20±9.98, with a median of six year (interquartile range of 3.25-13.75). Close to half (46.7%) of the midwives were young and had five years of experience or less. The distribution of the midwives' years of experience is presented in figure 2.

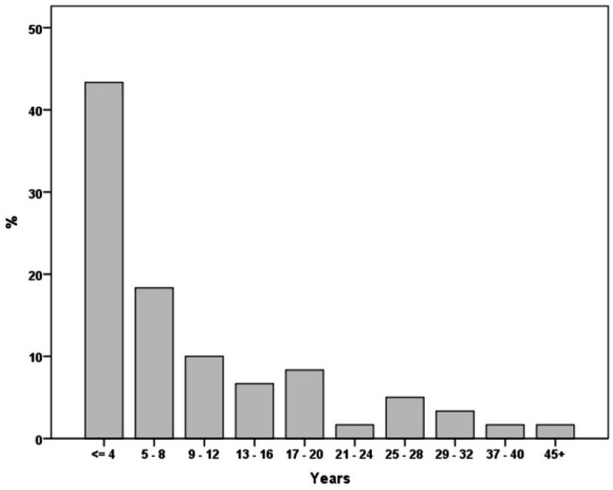

Figure 1. – Distribution of midwives years of experience.

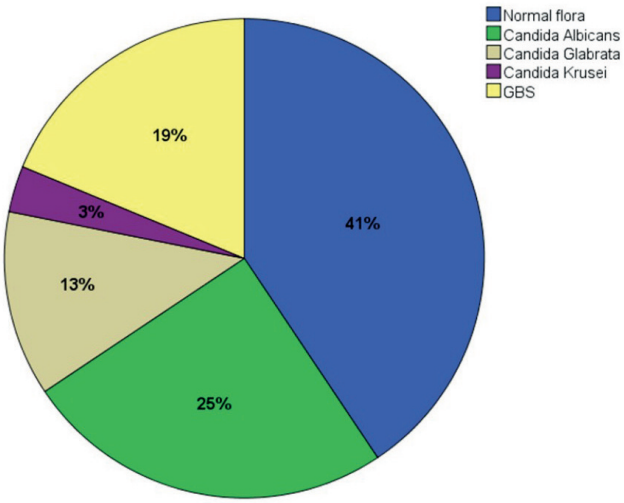

Figure 2. – Vaginal flora distribution.

Delivery characteristics

The first stage of labor was 5.93±4.67 hours (a median of 4.63, interquartile range 3.0-7.25). The second stage of labor was 1.40±1.15 hours long (a median of 1.27, interquartile range 0.33-2.17). Six patients had a prolonged second stage of labor (> 3 hours).

Most of the cases were delivered vaginally, in 27.1% of deliveries a vacuum extraction was performed and episiotomy was performed 40.7% (24) of deliveries.

Vaginal culture

Vaginal culture was taken from 31 women (51.7%). Of these, 13 (41.9%) resulted in growth of normal flora, six patients (19.4%) were positive for *group B streptococcus* (GBS) and another 12 patients (38.7%) had growth of *candida species*.

Of patients with *candida species* growth, eight had *Candida Albicans* (61.5%), 3 had *Candida Glaberta* (30.8%) and one with *Candida Krusei* (7.7%).

Neonatal characteristics

The mean birth weight was 3450.22±540.62, with twelve large for gestational age (> 4000 gr) newborns (20.3%). There were 63.2% male neonates (36) and 36.8% (21) female neonates.

The mean first minute Apgar score was 8.36±1.30 with a median of 9 (interquartile range 8-9). Nine newborns had a first minute Apgar score under 7 (15.3%). The mean five minute Apgar score was 9.90±0.44 with a median of 10 (interquartile 10-10), only in one case was the fifth minute Apgar score under 7 (1.7%).

The mean cord blood gas pH was 7.22±0.11, 11 cases (19.3%) had a low pH (<7.1).

The mean PCO2 was 54.93±14.90, base excess was recorded at -7.82±8.65, mean HCO3 level was 21.28±6.13, mean PO2 level was 21.77±24.41 and mean lactic acid level was 18.63±23.70.

DISCUSSION

Our study is a retrospective case series study which included all cases of severe perineal tears that took place at the

SUMC during the study period. The main findings of our study include a high rate of nulliparity (82%) among patients with OASIS, prolonged second stage of labor was found in 10% of the cases, vacuum extraction was used in 27% of deliveries and episiotomy was found in 41% of the cases.

We also found that the midwives' work experience was short, with 44% of the midwives treating the cases of OASIS had less than 4 years of experience.

Vaginal cultures revealed a rate of 38% of *candidal* infection and 20% of *GBS* infection. Most of the *candida* cases were infected with

candida albicans.

Additionally, a high rate of neonates were large for gestational age (20% of neonates weighed over 4 kg), a high rate of hypoxia was noted (19% of newborns had a cord blood pH level of less than 7.1 and 15% had a first minute Apgar score of less than seven).

These results are in accordance with previous studies[1-20]. Known risk factors for OASIS include prolonged second stage of labor, instrumental delivery, macrosomic fetuses and unexperienced midwifes[25]. Episiotomy has been found as a risk factor in some studies but others have not found such an association.

The higher rates of hypoxia and lower Apgar scores that we (have found in this group are known to be associated with the use of vacuum extraction and preforming an episiotomy[31]. These risk factors for severe perineal tears are unmodifiable. A rise in performing cesarean[26] sections in order to avoid severe perineal tears holds short and long term morbidity, as stated in the ACOG committee opinion .

The rate of vaginal *candidiasis* in this study population was 38% whereas the rate reported during pregnancy was reported to be between 15-30%[29]. We believe that the *candida* infection may cause changes in the vaginal tissue and make it more friable, leading to more tears of all grades as well as more severe tears. No previous studies investigating this association have been found to date. Since that detection and the treatment of vaginal *candidiasis* are simple, not expensive and have little side effects, we believe that this modifiable risk factor is worth exploring in a clinical trial setting.

The merits of this study include being a single center for the entire region of southern Israel serving a large obstetrical population accounting for a relatively large and unselected study population. However, there are shortcomings to the study including its retrospective nature leading to some missing data –such as missing vaginal cultures. This was a descriptive retrospective cohort study. Farther studies are needed in order to investigate the association we have described between vaginal *candida* colonization and severe perineal tears.

In conclusion, severe perineal tears have grave long term morbidity and with many known unmodifiable risk factors. We found an elevated rate of vaginal *candida* colonization in women with OASIS and believe there might be an association between the two.

DISCLOSURE STATEMENT

No author has any potential conflict of interest, any relationship with the companies that have financial interest in the information in this manuscript.

REFERENCES:

1. Shmueli A, Gabbay Benziv R, Hiersch L, Ashwal E, Aviram R, Yogev Y, Aviram A. Episiotomy - risk factors and outcomes. J Matern Fetal Neonatal Med. 2016 Mar 28, 1-19.
2. Verghese TS, Champaneria R, Kapoor DS, Latthe PM. Obstetric anal sphincter injuries after episiotomy: systematic review and meta-analysis. Int Urogynecol J. 2016, Feb 19.
3. Pennycuff JF, Northington GM, Loucks T, Suciu G, Karp DR. Obstetric Anal Sphincter Injury as a Quality Metric: 16-Year Experience at a Single Institution. Obstet Gynecol. 2016 Mar, 127 (3), 496-500.
4. Hamouda S, Mancini J, Marchand F et al. Severe perineal morbidity of instrumental deliveries using Thierry's spatulas and vacuum extraction: A prospective observational cohort study. J Gynecol Obstet Biol Reprod (Paris). 2015 Dec 17. pii: S0368- 2315(15)00350-6. doi: 10.1016/j.jgyn.2015.11.003.
5. Letouzey V, Bastide S, Ulrich D et al. Impact of Bacterial Vaginosis on Perineal Tears during Delivery: A Prospective Cohort Study. PLoS One. 2015 Nov 6, 10 (11): e0139334. doi: 10.1371/journal.pone.0139334. eCollection 2015.
6. Blondel B, Alexander S, Bjarnadóttir RI et al. Variations in rates of severe perineal tears and episiotomies in 20 European countries: a study based on routine national data in Euro-Peristat Project. Acta Obstet Gynecol Scand. 2016 Mar 9. doi: 10.1111/aogs.12894.
7. Halle TK, Salvesen KÅ, Volløyhaug I. Obstetric anal sphincter injury and incontinence 15-23 years after vaginal delivery. Acta Obstet Gynecol Scand. 2016 Mar 17. doi: 10.1111/aogs.12898.
8. Yohay D, Weintraub AY, Mauer-Perry N et al. Prevalence and trends of pelvic floor disorders in late pregnancy and after delivery in a cohort of Israeli women using the PFDI-20. Eur J Obstet Gynecol Reprod Biol. 2016 Feb 28, 200, 35-39. doi: 10.1016/j.ejogrb.2016.02.037.
9. Meister MR, Cahill AG, Conner SN, Woolfolk CL, Lowder JL. Predicting obstetric anal sphincter injuries in a modern obstetric population. Am J Obstet Gynecol. 2016 Feb 20. pii: S0002-9378(16)00341-0. doi: 10.1016/j.ajog.2016.02.041.
10. Groutz A, Cohen A, Gold R et al. Risk factors for severe perineal injury during childbirth: a case-control study of 60 consecutive cases. Colorectal Dis. 2011 Aug; 13 (8): e216-9. doi: 10.1111/j.1463-1318.2011.02620.x.
11. Landy HJ, Laughon SK, Bailit JL et al. Characteristics associated with severe perineal and cervical lacerations during vaginal delivery. Obstet Gynecol. 2011 Mar; 117 (3): 627-35.
12. Yeaton-Massey A, Wong L, Sparks TN et al. Racial/ethnic variations in perineal length and association with perineal lacerations: a prospective cohort study. J Matern Fetal Neonatal Med. 2015 Feb; 28 (3): 320-3. doi: 10.3109/14767058.2014.916675. Epub 2014 May 22.
13. Grobman WA, Bailit JL, Rice MM et al. Racial and ethnic disparities in maternal morbidity and obstetric care. Obstet Gynecol. 2015 Jun; 125 (6): 1460-7.
14. Hopkins LM, Caughey AB, Glidden DV, Laros RK Jr. Racial/ethnic differences in perineal, vaginal and cervical lacerations. Am J Obstet Gynecol. 2005 Aug; 193 (2): 455-9.
15. Vale de Castro Monteiro M, Pereira GM, Aguiar RA, Azevedo RL, Correia-Junior MD, Reis ZS. Risk factors for severe obstetric perineal lacerations. Int Urogynecol J. 2016 Jan; 27 (1): 61-7.
16. Hsieh WC, Liang CC, Wu D, Chang SD, Chueh HY, Chao AS. Prevalence and contributing factors of severe perineal damage following episiotomy-assisted vaginal delivery. Taiwan J Obstet Gynecol. 2014 Dec; 53 (4): 481-5.
17. Hauck YL, Lewis L, Nathan EA, White C, Doherty Risk factors for severe perineal trauma during vaginal childbirth: a Western Australian retrospective cohort study. Women Birth. 2015 Mar; 28 (1): 16-20. doi: 10.1016/j.wombi.2014.10.007. Epub 2014 Dec 1.
18. Schmitz T, Alberti C, Andriss B, Moutafoff C, Oury JF, Sibony O. Identification of women at high risk for severe perineal lacerations. Eur J Obstet Gynecol Reprod Biol. 2014 Nov;182:11-5. doi: 10.1016/j.ejogrb.2014.08.031. Epub 2014 Aug 29.
19. Edozien LC, Gurol-Urganci I, Cromwell DA et al. Impact of third- and fourth-degree perineal tears at first birth on subsequent pregnancy outcomes: a cohort study. BJOG. 2014 Dec; 121 (13): 1695-703. doi: 10.1111/1471-0528.12886. Epub 2014 Jul 9.
20. Hirsch E, Elue R, Wagner A Jr et al. Severe perineal laceration during operative vaginal delivery: the impact of occiput posterior position. J Perinatol. 2014 Dec; 34 (12): 898-900. doi: 10.1038/jp.2014.103. Epub 2014 May 29.

21. Dunn AB, Paul S, Ware LZ, Corwin EJ. Perineal Injury During Childbirth Increases Risk of Postpartum Depressive Symptoms and Inflammatory Markers. J Midwifery Womens Health. 2015 Jul-Aug; 60 (4): 428-36. doi: 10.1111/jmwh.12294.
22. McNicol FJ, Bruce CA, Chaudhri S et al. Outcome of repair of obstetric anal sphincter injuries after three years. Int J Gynaecol Obstet. 2014 Oct; 127 (1): 47-50. doi: 10.1016/j.ijgo.2014.04.013. Epub 2014 Jun 12.
23. Reid AJ, Beggs AD, Sultan AH, Roos AM, Thakar R. Management of obstetric anal sphincter injuries – a role for the colorectal surgeon.Colorectal Dis. 2010 Sep; 12 (9): 927-30. doi: 10.1111/j.1463-1318.2009.01897.x. Epub 2009 Apr 13.
24. Sundquist JC. Long-term outcome after obstetric injury: a retrospective study. Acta Obstet Gynecol Scand. 2012 Jun; 91 (6): 715-8. doi: 10.1111/j.1600-0412.2012.01398.x. Epub 2012 Apr 30.
25. Ott J, Gritsch E, Pils S et al. A retrospective study on perineal lacerations in vaginal delivery and the individual performance of experienced mifwives. BMC Pregnancy Childbirth. 2015 Oct 22;15:270. doi: 10.1186/s12884-015-0703-0.
26. ACOG Committee Opinion No. 647: Limitations of Perineal Lacerations as an Obstetric Quality Measure. Committee on Obstetric Practice. Obstet Gynecol. 2015 Nov; 126 (5): e108-11.
27. Farr A, Kiss H, Holzer I, Husslein P, Hagmann M, Petricevic L. Effect of asymptomatic vaginal colonization with Candida albicans on pregnancy outcome. Acta Obstet Gynecol Scand. 2015 Sep; 94 (9): 989-96. doi: 10.1111/aogs.12697. Epub 2015 Jul 14.
28. Aguin TJ, Sobel JD. Vulvovaginal candidiasis in pregnancy. Curr Infect Dis Rep. 2015 Jun; 17 (6): 462. doi: 10.1007/s11908-015- 0462-0.
29. Soong D, Einarson A. Vaginal yeast infections during pregnancy. Can Fam Physician. 2009 Mar; 55 (3): 255-6.
30. Smith LA, Price N, Simonite V, Burns EE. Incidence of and risk factors for perineal trauma: a prospective observational study. BMC Pregnancy Childbirth. 2013 Mar 7; 13: 59. doi: 10.1186/1471-2393-13-59.
31. Corrêa Junior MD, Passini Júnior R. Selective Episiotomy: Indications, Techinique, and Association with Severe Perineal Lacerations. Rev Bras Ginecol Obstet. 2016 Jun; 38 (6): 301-7. doi: 10.1055/s-0036-1584942. Epub 2016 Jul 11.

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Original article

Treatment of Pelvic Organ Prolapse using a lightweight modified HexaPro Mesh

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Abstract: The surgical procedure of vaginal mesh placement for the treatment of Pelvic Organ Prolapse is described. A modified ultralightweight macroporous monofilament polypropylene mesh with insertion aids at the six fixation points is placed vaginally through a single incision.

Keywords: Minimally invasive; Single incision; Six point mesh; Pelvic organ prolapse; Macroporous polypropylene; Vaginal surgery.

INTRODUCTION

Recurrence rates of Pelvic Organ Prolapse (POP) of up to 30% have been described after classical surgical procedures without prosthetic implants[1-3]. While controversy exists regarding the role of larger implants of surgical meshes, recent developments of new materials and insights regarding tissue tolerance and fibroblast proliferation warrant reevaluation of previously held concerns[4]. Advisories i.e. of the Food and Drug Administration (FDA) in 2008[5] and Health Canada in 2010 were largely based on experiences with comparably heavy meshes with microporous structure frequently using only two or four points of fixation in the pelvis with the apical sacrospinous attachment being absent[6]. Some of the early meshes that frequently caused poor tissue tolerance and erosion were much higher than the newer meshes. The surface area of only laterally fixated four-point meshes was prone to contract further compromising the therapeutic effect and necessitating secondary surgical interventions to treat complications and/or anatomical failure. With ultralight macroporous monofilament polypropylene meshes available on the market erosions have only been reported to be up to 7%[7]. In preparation for a multicenter prospective trial evaluating POP treatment by primary mesh placement, a standardized surgical method was developed for the placement of a newly modified single-incision vaginal mesh with 6 point fixation[8].

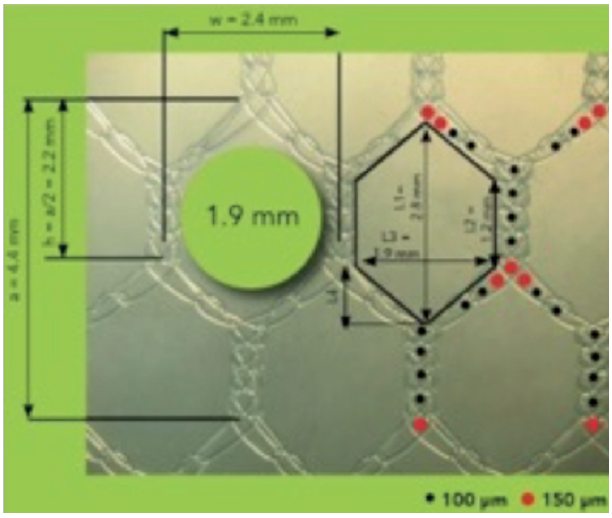

Figure 1. – HexaPro Mesh Structure and Pore Size.

MATERIALS AND METHODS

Material. An ultralightweigt monofilament polypropylene mesh (21g/m^2) was redesigned on the basis of an earlier configuration inaugurated by Mistrangelo et al.[8]. The new mesh (INGYNious®)is manufactured by A.M.I. Inc, Feldkirch, Austria and CE certification has been obtained. This mesh is characterized by large micropores of 100-150 μm and macropores of 1,9 to 2.8 mm (Fig. 1). In comparison with the earlier design the implant was markedly elongated in the direction of the sacrum while excising an arch for the passage of the rectum. The fixation points were predefined and passage aids were placed through the mesh in six positions facilitating suturing and at the same time further standardizing the surgical procedure. Two sizes were configured, a larger one for anterior mesh placement in the vesico-vaginal space and a smaller one for positioning in the recto-vaginal compartment (Fig. 2).

In order to fix the mesh in the pelvis in a safe and reproducible manner minimizing preparatory effort and tissue trauma the i-Stitch® instrument was used (A.M.I.). This instrument consists of a stainless steel hook with a long hollow

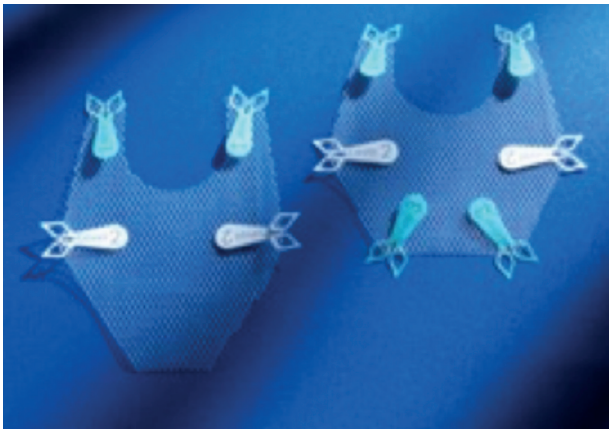

Figure 2. – InGYNious Anterior and Posterior Six-Point Meshes.

Figure 3. – i-Stitch Instrumentation for Intrapelvic Suture Placement.

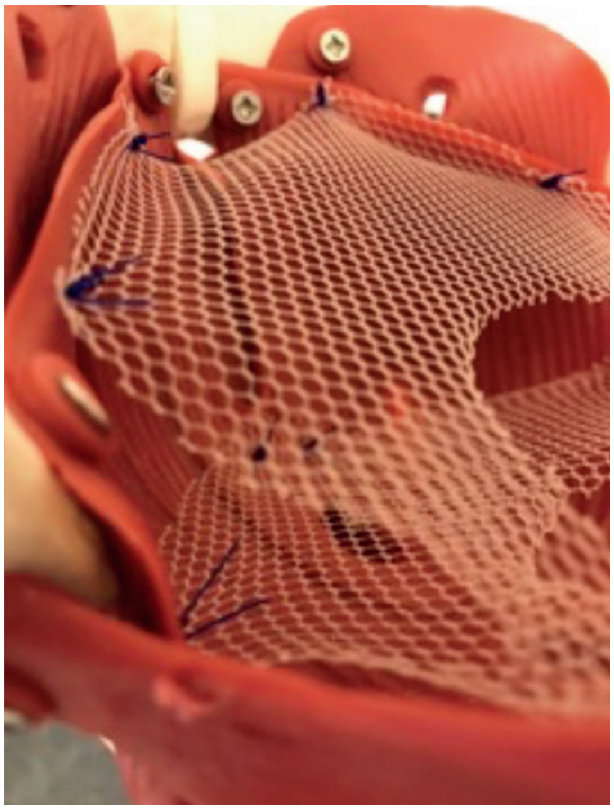

Figure 4. – Anterior and Posterior InGYNious Mesh in an Anatomical Model. View from parasacrally towards the Perineum.

handle admitting a disposable guide that advances a non-resorbable or resorbable suture towards the inner surface of the blunt hook where a pre-formed patented knot at the end of the suture is pushed into a groove first compressing it on entry and thereafter capturing it through it's re-expansion (Fig. 3). Two i-Stitch instruments are available. One with the hook pointing away from the palm of the surgeon's hand holding the handle and one with the hook pointing towards it. It is the surgeon's preference to choose the one lending itself to most comfortably placing the suture.

Methods. After single-shot antibiotic prophylaxis a single longitudinal full-thickness incision is made in the anterior or posterior wall of the vagina. The paravesical and/or pararectal spaces are opened. Three sutures of 2-0 polyester are placed on each side of the pelvis in a symmetrical fashion for a total of six sutures (Fig. 4).

The anterior vaginal mesh is fixed in the apical third of the sacrospinous ligament on boths sides. (Suture 1 is placed us-

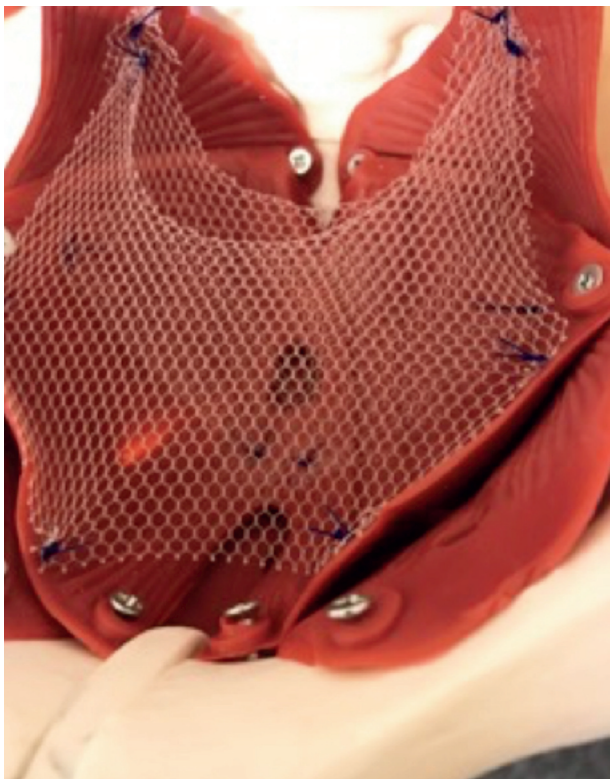

Figure 5. – Anterior and Posterior InGYNious Mesh with Six-Point Fixation in an Anatomical Model. View from the Perineum towards the Presacral Region.

ing by i-Stitch-up). The second point of fixation is the tendinous arch at the ischiadic spine on both sides. The sutures are placed through the ligament using the I-Stitch down (Suture 2). The third point of fixation by I-Stitch down is the tendinous arch of the pubic bone (Suture 3).

For posterior mesh placement in the rectovaginal space the fixation points are similar: fixation point number one is the medial part of the sacrospinous ligament on both sides, the second fixation point is the ileococcygeal muscle at the ischiadic spine on both sides. The third point of fixation is the perineal body bilaterally to keep the mesh in place.

The sutures are threaded through the mesh via the passage aids and tied starting with positions 1 followed by 2 and 3. The colpotomy is closed with a resorbable braided 2-0 suture. A vaginal pack is used and left in place for at least 24 hours.

DISCUSSION

In this paper we describe the use of a newly designed mesh with 6 point fixation and user friendly suture placement. Standardization is one of the most important features to assure high quality in surgery. This manuscript has been written in the preparation of a large multicenter study based on the surgical techniques described.

Negative experiences with first generation alloplastic meshes have lead to widely disseminated concerns regarding the use of meshes and their role in the treatment of POP leading to uncertainties in therapeutic recommendations on the part of the treating physicians and to negative preconceived notions on the part of the patients.

With the development of improved materials and standardized minimally invasive placement techniques these concerns need to be re-addressed. Clinical impression suggests that modern meshes may be well tolerated and anatomically efficient providing a good quality of life, es-

pecially also regarding sexual function. Prospective multicenter data are therefore urgently needed to test the merits and caveats of such newly designed meshes. The procedure of using the modified InGYNious mesh placement as described above provides a standardized basis for such an evaluation utilizing an advanced prosthetic material.

DISCLOSURES

All authors have received honoraria from A.M.I. for presentations at internal or scientific meetings related to pelvic floor surgery.

REFERENCES

1. Nygaard I, Barber MD, Burgio KL et al. Prevalence of symptomatic pelvic floor disorders in US women. JAMA 2008; 300 (11): 1311-6.
2. Maher C, Feiner B, Baessler K et al. Surgical management of pelvic organ prolapse in women. Cochrane Database of Systematic Reviews 2013 (4).
3. Walker GJA, Gunasekera P. Pelvic organ prolapse and incontinence in developing countries: review of prevalence and risk factors. Int Urogynecol J 2011; 22 (2): 127-35.
4. Jia XL, Glazener C, Mowatt G et al. Systematic review of the efficacy and safety of using mesh in surgery for uterine or vaginal vault prolapse. Int Urogynecol J 2010; 21 (11): 1413-31.
5. FDA Public Health Notification: Serious Complications Associated with Transvaginal Placement of Surgical Mesh in Repair of Pelvic Organ Prolapse and Stress Urinary Incontinence 2008 [cited 2010 9 November 2010].
6. Barski D, Otto T, Gerullis H. Systematic review and classification of complications after anterior, posterior, apical, and total vaginal mesh implantation for prolapse repair. Surg Technol Int 2014; 24: 217-24.
7. Mowat A, Maher D, Baessler K et al. Surgery for women with posterior compartment prolapse. The Cochrane database of systematic reviews 2018; 3: Cd012975.
8. Mistrangelo E. Pelvic floor reconstruction using CR-mesh (A.M.I.): past, present, future. Pelviperineology 2011; 30 (4).

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Commentary

I am glad to read this paper written by German authors on the characteristics of "InGYNious" of which I am an inventor. It is not just a new type of mesh but a new philosophy of the prosthetic technique of repairing pelvic floor defects. Unfortunately, in the recent past the use of old meshes with transcutaneous passages have caused serious damage to the patients. Today ultralight meshes with single incision transvaginal access have completely changed the story with excellent results, lasting effectiveness over time, rapidity of intervention, spinal anesthesia, very low blood loss, reproducibility, short hospitalization.

Before providing some of the results of my 7 years experience, I must unfortunately recall that in the literature there are still studies that demonize the use of meshes by vaginal route. After the bad English study Prospect[1], recently the Prospere study appeared on European Urology[2], signed by authoritative French colleagues. It supports the superiority of the laparoscopic approach compared to the vaginal one. I have just written a letter to the editor to challenge the scientific method that leads to these conclusions. Furthermore, beyond the methodology, the authors' superficialiy is highlighted by many points of the vaginal operative technique. The heterogeneity in mesh composition (polypropylene alone / combined with absorbable components), kits and techniques for VMR (variable mesh size, with / without sacrospinous-fixation), as well as the considerable number of LS requiring a conversion to the vaginal route may affect the comparison between groups in terms of severe complications. The same thing happened for the study Prospect, published in Lancet, in which it was said that mesh should be placed "possibly" below the vaginal fascia, when it is now established that this is the key point to avoid vaginal erosion.

There are changes after years of experience in the first form of InGYNious with a 3rd level for the correction of cystocele with lateral attachment to the tissues lateral to bladder neck and then a narrower anterior part and for the correction of rectocele with a central attachment to the perineal body and laterally to the deep transversus muscle.

My experience concerns 296 patients operated in the last 3 years. I had 3 recurrence of cystocele, 1 of rectocele, 2 of hysterocele. The latter were treated with vaginal hysterectomy and with the vaginal vault attachement to the upper part of the normal-positioned mesh at the 1st level. As far as complications are concerned, there were no infections, no hematomas to be treated surgically, no extrusions, no schrinking, 4 vaginal erosions treated with the removal of part of the mesh and of eroded vagina. 10% of patients have an IUS "de novo", which is the same as that of the fascial surgery, of which only 3% has been corrected with TOT. 1% of patients had pelvic pain that lasted a month, but among active sex life only 0.5% of patients had modest dyspareunia.

Unfortunately, in Italy, due to legal sue, it is almost impossible to do randomized studies and therefore we will have only observational studies. The Italian Association of UroGinecology (AIUG), to which I belong, has been providing for two years a data collection software (SRD) on the surgical correction of pelvic floor defects that now contains more than 1000 patients. The SRD will allow an observational analysis of an appropriate number of patients operated with different techniques by 2018.

I conclude with the words of the last Cochrane 2018 on the back compartment inviting to give a precise meaning to the written words "Evidence does not support the utilization of any mesh or graft materials at the time of posterior vaginal repair", which means that, with the support of a favorable personal experience, meshes can be used.

REFERENCES

1. Glazener CMA, Breeman S, Elders A et al (for the PROSPECT study group)* Mesh, graft, or standard repair for women having primary transvaginal anterior or posterior compartment prolapse surgery: two parallel-group, multicentre, randomised, controlled trials (PROSPECT) www.thelancet.com Published online December 20, 2016 http://dx.doi.org/10.1016/S0140-6736(16)31596-3.
2. Lucot JP, Cosson M, Bader G et al. Safety of Vaginal Mesh Surgery Versus Laparoscopic Mesh Sacropexy for Cystocele Repair: Results of the Prosthetic Pelvic Floor Repair Randomized Controlled Trial. EURURO https://doi.org/10.1016/j.eururo.2018.01.044

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Original article

Pelvic trauma and pudendal syndrome (post-traumatic pudendal syndrome)

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Abstract: The pudendal syndrome can result from pelvic trauma but the link is difficult to prove. The study was intended to demonstrate the existence of post-traumatic pudendal syndromes. Two case reports were used to illustrate the point. The study was based on 394 female patients of whom 216 (54.8%) had a history of pelvic trauma. Patients were further classified into mutually exclusive groups according to presence/absence of perineodynia and latency of pain appearance in those with pelvic trauma (pain before trauma, early onset, or late onset). Urge urinary incontinence, cystalgia, anal incontinence and proctalgia fugax were found statistically more frequent in the pelvic trauma group. Perineodynia visual analog score, NHI-CPSI score and Wexner's score were also significantly greater in traumatic patients. The three pudendal syndrome clinical signs were significantly more present in pelvic trauma patients than in non-trauma subjects. In the three post-traumatic perineodynia groups with different latency, only minor significant symptom frequency differences were observed but importantly urge incontinence, cystalgia, anal incontinence and proctalgia fugax remained more frequent than in the non-trauma perineodynia group for similar pain scores. Post-traumatic pudendal syndrome is a reality. Perineodynia, urge incontinence, anal incontinence, proctalgia fugax and cystalgia are the most frequently symptoms encountered. These findings recommend performing a detailed history search for any symptom of the pudendal syndrome and a comprehensive clinical examination including its three clinical signs after any significant pelvic trauma.

Keywords: Bladder pain; Incontinence; Medico-legal; Pelvic trauma; Pudendal neuralgia.

INTRODUCTION

Pudendal nerves are involved in the three basic functions of the perineum: defecation, voiding and sexual activity. Because they contain sensory, motor and autonomic fibers, any lesion of these nerves can induce a great variety of symptoms depending on which of the three branches (rectal, perineal, penile/clitoral) and which fibers are compressed or stretched[1-9]. The symptoms vary widely among subjects but in the same patient they are often related to activities affecting the nerves and may be viewed as part of a genuine "pudendal syndrome".

The pudendal syndrome can result from a pelvic trauma but as it is rarely diagnosed patients facing the syndrome after a work or a motor vehicle accident (MVA) have great difficulties proving the link between the trauma and the sequelae that may persist sometimes for the rest of their life.

In the experience of the first author (JB), a history of pelvic trauma (e.g. fall on the tailbone, traffic accident) with or without fracture was found in 60% of the patients who needed a pudendal nerve decompression surgery regardless of the time elapsed between the accident and the appearance of the pudendal syndrome. Amazingly, in the same population, only 5 % of the patients described pelvic trauma as the cause of their symptoms. In a recent study establishing normative values for skin temperature and thermal sensory thresholds in the pudendal nerve territory in a population of presumably healthy women, a history of pelvic trauma was evidenced in only 24.4% of them[10] .

The primary goal of this study was to assess the potential link between pelvic trauma and appearance of a pudendal syndrome. Secondary objectives were to evaluate more carefully perineodynia (perineal pain), one of the salient symptoms of the pudendal syndrome, and its relationship with trauma. Two case reports were used to illustrate the point.

MATERIAL AND METHODS

A retrospective study was carried out on female patients of the perineology [11-14] consultation database of the first author (JB) based on the question: "In your life did you experience any significant trauma that could have had an impact on your pelvic bones (e.g. fall on the buttock-tailbone, professional and/or traffic accident, leg fracture)". A complete history including urinary, ano-rectal and sexual functions was recorded from each patient. Four scores were used to evaluate the main symptoms : ICIQ-SF for urinary incontinence [15], St Mark's score for anal incontinence[16], Wexner's score for constipation [17] and NHI-CPSI adapted for women by Antolak for pain, lower urinary tract symptoms and quality of life[7, 18]. Perineodynia (perineal pain) intensity was measured using a visual analog scale (VAS) from 0 (no pain) to 10 (extreme pain). Three clinical signs characteristic of the pudendal syndrome were checked (asymmetric pinprick sensibility of the perineum, painful skin rolling test and painful pudendal nerve during vaginal or rectal examination)[8]. Perineal descent (and perineal position at rest and during Valsalva's maneuver) was measured with a perineocaliper® [19]. Lack of any score value, clinical sign evaluation or information about urinary incontinence, urinary frequency, nocturia, dysuria, cystalgia, dyspareunia, sexual arousal syndrome, anal incontinence (gas, liquid and solid), dyschesia, proctalgia fugax and perineodynia was defined as an exclusion criterion.

Pelvic trauma and pudendal syndrome

The study material consisted of 394 female patients; 216 (54.8%) had a history of pelvic trauma (17 from MVA) and 178 (45.2%) had not. When comparing the two groups of patients with respect to age, weight, height and parity, no significant difference was observed.

Pelvic trauma and perineodynia

The link between perineodynia, one of the main symptoms of the pudendal syndrome, and pelvic trauma was scrutinized in detail. Proctalgia fugax, a short lasting pain in the anus, was not considered as perineodynia and was therefore studied separately.

The study patients were further classified in 6 mutually exclusive groups according to presence or absence of peri-

neodynia, the occurrence or not of a pelvic trauma, and the latency between pain appearance and pelvic trauma. Seven subjects could not be allocated because latency was unknown, leaving only 387 patients for group analyses. Specifically, the 6 groups were defined as follows.

Patients with a VAS pain score equal to 0 ("no pain", n=103) were distributed in 2 groups:

– Group 1: "No pain- No trauma" (n=65)
– Group 2: "No pain-Trauma" (n=38)

Patients with a VAS pain score > 0 ("pain", n= 284) were split into 4 groups:

– Group 3: "Pain-No trauma" (n=113): perineodynia without any history of pelvic trauma.
– Group 4: "Pain early onset-Trauma" (n=35): early onset of post-traumatic perineodynia. Appearance of pain just after trauma (latency trauma-beginning of pain = 0)
– Group 5: "Pain late onset-Trauma" (n=119): late onset of post-traumatic perineodynia. Appearance of pain several months after trauma (latency trauma-beginning of pain > 0)
– Group 6: "Pain before- Trauma" (n=17): pelvic trauma after the appearance of perineodynia (latency trauma-beginning of pain < 0).

Other early onset post-traumatic symptoms were retrieved from the database.

Case Reports

Two examples of pelvic trauma resulting in pudendal syndrome were selected manually from the personal patient database of the second author (SA).

Statistics

Data were summarized as mean and standard deviation (SD) or as median and interquartile range (IQR) for quantitative variables and as numbers and percentages for categorical findings. Mean values were compared by one-way analysis of variance (ANOVA) or by the Kruskal-Wallis non parametric test for skewed data; the comparison of proportions was done by the chi-squared test for contingency tables. Results were considered significant at the 5% critical level (P<0.05). All calculations were done with SAS version 9.4 (SAS Institute, Cary, NC, USA) statistical package and R version 3.2.2.

RESULTS

Pelvic trauma and pudendal syndrome

Patients and pelvic trauma

When comparing patients with pelvic trauma and patients without pelvic trauma, the only significant difference observed was a reduced frequency of grade 2 or 3 cystocele in the first group (Table I). This could be explained by a selection bias inherent to the perineology consultation database which mainly consists of patients with two types of problems: perineodynia and/or prolapse and urinary incontinence. Four common factors inducing the pudendal syndrome, namely perineal descent, parity (number of deliveries), transobturator mesh surgeries (TVT-O®, Prolift®) and cycling, were equally prevalent in the two groups. Pelvic trauma was associated with fracture or luxation in only 11% of the cases.

Pelvic trauma and symptoms of pudendal syndrome

After adjusting for the difference in the distribution of cystocele grade 2 or 3 reported above, urge urinary incontinence, cystalgia (bladder pain), anal incontinence and proctalgia fugax were found statistically more frequent in the pelvic trauma group (Table II). Perineodynia VAS score, NHI-CPSI score and Wexner's score were also significantly greater in patients with trauma. The proportion of patients with perineodynia (VAS score > 0) was significantly higher in the pelvic trauma group than in the other group (82.4% vs.63.5%; P<0.0001).

Table I. Characteristics of patients with and without pelvic trauma

Parameter	Without trauma (n=178)	With trauma (n=216)	P-value
Age (years)	50.8 ± 14.2	51.3 ± 13.2	0.72
Parity	1.7 ± 1.3	1.8 ± 1.2	0.75
Weight (kg)	62.8 ± 11.5	62.7 ± 11.6	0.92
Height (cm)	163.3 ± 6.1	163.7 ± 7.3	0.62
BMI (kg/m²)	23.5 ± 4.2	23.6 ± 4.4	0.88
Biking (%)	19.8	20.9	0.84
Trans-obturator mesh surgeries (%)	4.5	6.5	0.39
Cystocele (%)[a]	21.1	11.8	0.014
Rectocele (%)[a]	12.9	11.3	0.64
Perineum at rest (cm)	0.23 ± 1.0	0.38 ± 1.0	0.18
Perineum during valsalva (cm)	-0.35 ± 1.2	-0.15 ± 1.3	0.14
Perineal descent (cm)	0.58 ± 0.7	0.53 ± 0.6	0.49

[a] grade 2 and 3

Table II. Symptoms of the pudendal syndrome in patients with and without pelvic trauma

Parameter	Without trauma (n=178)	With trauma (n=216)	P-value
Stress urinary incontinence (%)[a]	38.8	39.4	0.96
Urge urinary incontinence (%)[a]	21.9	32.9	0.020
Urinary frequency (min)[a]	135 ± 63.8	126 ± 63.7	0.12
Nocturia[a]	0.98 ± 1.3	1.0 ± 1.3	0.68
Dysuria (%)[a]	33.1	40.7	0.063
Cystalgia (%)[a]	22.5	32.4	0.031
ICIQ-SF (/21)[a]	4.4 ± 5.1	5.4 ± 6.0	0.26
Perineodynia (/10)	3.7 ± 3.3	4.8 ± 2.9	0.0002
NHI-CPSI (/44)	24.6 ± 9.3	27.8 ± 8.1	0.017
Dyspareunia (%)	39.9	46.8	0.17
Sexual arousal syndrome (%)	20.2	25.9	0.18
Anal incontinence (%)	31.5	41.9	0.034
St Marks score (/24)	3.5 ± 4.7	4.4 ± 5.1	0.22
Prolapse disturbance (%)	20.9	13.6	0.058
Proctalgia fugax (%)	20.2	34.7	0.0015
Dyschesia (%)	75.6	74.5	0.85
Wexner score (/30)	7.4 ± 4.8	9.4 ± 5.6	0.013

[a] P-value adjusted for cystocele

Clinical signs of the pudendal syndrome

The three pudendal syndrome clinical signs were significantly more present in pelvic trauma patients than in non-trauma subjects (Table III)

Pelvic trauma and perineodynia

Pain characteristics

The duration of pain was significantly different in the 4 "Pain" groups (P=0.0002). The median (IQR) pain duration was respectively 22 (10-48) months for "Pain-No Trauma",

TABLE III. Clinical signs of the pudendal syndrome in patients with and without pelvic trauma

Parameter	Without trauma (n=178)	With trauma (n=216)	P-value
Asymmetric pinprick sensibility (%)	40.4	64.4	<0.0001
Painful pudendal nerve (%)	61.2	81.9	<0.0001
Painful skin rolling test (%)	51.7	66.2	0.0035
3 clinical signs positive (%)	28.1	49.1	<0.0001
3 clinical signs negative (%)	27.5	10.6	<0.0001
Number of clinical signs positive	1.5 ± 1.2	2.1 ± 1.0	<0.0001

30 (12-84) months for "Pain early onset-Trauma", 15 (7-48) months for "Pain late onset-Trauma", and 60 (48-96) months for "Pain before-Trauma". The median (IQR) latency between trauma and pain was 0 (0-0) month in the "Pain early onset-Trauma" group and 156 (58-309) months in the "Pain late onset-Trauma" group, while it amounted -36 (-48 to -18) months in the "Pain before-Trauma" group.

Trigger factors

Pelvic trauma was the trigger factor that precipitated symptoms in all 35 patients of the "Pain early onset-Trauma" group. This represents 12.5 % of the patients with perineodynia (20.7% of the patients with perineodynia and a history of pelvic trauma). For the other groups ("Pain-No trauma", "Pain late onset- Trauma" and Pain before-Trauma"), no difference was observed with respect to the trigger factors (Table IV).

TABLE IV. Trigger factors in the "Pain" groups ("Pain early onset-Trauma" excluded)

Trigger factor	Pain-No trauma (N=113) Number (%)	Pain late onset-Trauma (N=119) Number (%)	Pain before-Trauma (N=17) Number (%)	All pain groups (N=249) Number (%)
None	51 (47.7)	55 (50.0)	5 (31.3)	111 (47.6)
Invasive medical procedures or surgeries	16 (14.9)	18 (16.4)	5 (31.3)	39 (16.7)
Long time sitting (work, journey...)	8 (7.5)	8 (7.3)	0 (0.0)	16 (6.9)
Psychological trauma	5 (4.7)	7 (6.4)	2 (12.5)	14 (6)
Wrong movement	7 (6.5)	5 (4.6)	2 (12.5)	14 (6)
Childbirth	7 (6.5)	5 (4.6)	1 (6.3)	13 (5.6)
Cystitis	4 (3.7)	4 (3.6)	0 (0.0)	8 (3.4)
Biking	5 (4.7)	3 (2.7)	0 (0.0)	8 (3.4)
Sexual intercourse	3 (2.8)	2 (1.8)	1 (6.3)	6 (2.6)
Severe dyschesia	1 (0.9)	3 (2.7)	0 (0.0)	4 (1.7)

TABLE V. Symptoms and clinical signs of patients classified in 6 distinct groups as defined in text (N=387)

Parameter	Group 1 No pain - No trauma (n=65)	Group 2 No pain - Trauma (n=38)	Group 3 Pain - No trauma (n=113)	Group 4 early onset-trauma (n=35)	Group 5 pain late onset-trauma (n=119)	Group 6 Pain before-trauma (n=17)	P-value** Group 1 vs Group 2	P-value** Group 3 vs Groups 4-6
General characteristics								
Parity	2.0 ± 1.2	2.3 ± 1.0	1.5 ± 1.3	1.3 ± 1.1	1.7 ± 1.3	2.0 ± 1.4	0.076	0.19
Cystocele grade 2 et 3 (%)	40.3	23.7	10.1	2.9	13.0	0.0	0.040	0.59
Rectocele grade 2 et 3 (%)	19.4	21.1	9.2	11.4	9.5	6.2	0.95	0.91
Symptoms								
Stress urinary incontinence (%)*	24.6	39.5	5.3	8.6	10.1	5.9	0.72	0.22
Urge incontinence (%)	36.9	65.8	13.3	34.3	20.2	47.1	0.020	0.012
Urinary frequency (min)	128.1 ± 63.2	99.1 ± 51.2	139.5 ± 64.0	122.6 ± 61.4	134.4 ± 67.5	144.7 ± 52.2	0.028	0.46
Nocturia	1.1 ± 1.2	1.2 ± 1.8	0.9 ± 1.3	0.6 ± 0.8	1.0 ± 1.2	1.1 ± 1.2	0.90	0.71
Dysuria (%)	40.0	31.6	29.2	31.4	47.1	47.1	0.49	0.0071
Cystalgia (%)	18.5	15.8	24.8	34.3	34.4	41.2	0.62	0.049
ICIQ-SF (/21)	6.2 ± 6.3	11.9 ± 4.3	3.7 ± 4.4	3.3 ± 5.5	4.6 ± 5.8	4.9 ± 4.9	0.070	0.72
Perineodynia (/10)	0.0 ± 0.0	0.0 ± 0.0	5.8 ± 2.2	6.1 ± 1.9	5.6 ± 2.0	7.2 ± 1.8	NA	0.56
NHI-CPSI (/44)	18.9 ± 9.9	20.1 ± 8.0	27.0 ± 7.9	26.7 ± 7.8	29.3 ± 7.1	32.8 ± 6.0	0.41	0.062
Dyspareunia (%)	29.2	28.9	46.0	57.1	47.9	52.9	0.64	0.68
Sexual arousal syndrome (%)	10.8	18.4	25.7	28.6	26.9	23.5	0.22	0.47
Anal incontinence (%)	35.4	34.2	29.2	42.9	43.2	41.2	0.81	0.036
St Marks score (/24)	3.5 ± 5.2	3.8 ± 5.6	3.5 ± 4.6	2.9 ± 4.1	5.0 ± 5.3	3.8 ± 3.1	0.28	0.39
Prolapse disturbance (%)	37.5	29.7	11.1	9.4	9.6	17.6	0.91	0.95
Proctalgia fugax (%)	10.8	18.4	25.7	42.9	36.1	41.2	0.41	0.014
Dyschesia (%)	88.9	38.1	69.8	86.4	76.6	100	0.0020	0.15
Wexner score (/30)	6.0 ± 4.1	5.2 ± 3.2	8.0 ± 4.9	9.7 ± 5.3	10.1 ± 5.5	10.8 ± 7.6	0.16	0.035
Clinical signs								
Asymmetric Pinprick sensibility (%)	29.2	60.5	46.9	62.9	66.4	58.8	0.0008	0.0030
Painful Alcock's canal (%)	35.4	57.9	76.1	97.1	85.7	82.4	0.014	0.027
Painful skin rolling test (%)	36.9	39.5	60.2	85.7	65.6	88.2	0.70	0.094
3 signs negative (%)	47.7	26.3	15.9	2.9	7.6	11.8	0.023	0.030
3 signs positive (%)	15.4	31.6	35.4	57.1	50.4	58.8	0.027	0.0069
Number of clinical signs	1.0 ± 1.1	1.6 ± 1.2	1.8 ± 1.1	2.5 ± 0.7	2.2 ± 1.0	2.3 ± 1.0	0.0093	0.0027

* moderate or severe

** P-values adjusted for grade 2-3 cystocele, rectocele and parity

Relation between trauma, pain occurrence and onset (6 study groups)

Parity, cystocele and rectocele grade 2 or 3 were more frequent in the two "No pain" groups (Table V). The other characteristics were comparable in the 6 groups. Results for symptoms and clinical signs are presented in Table V. P-values were adjusted for parity and grade 2 and 3 cystoceles and rectoceles. Comparisons were made between groups with P-values being adjusted for parity, grade 2 and 3 cystoceles and rectoceles.

Impact of trauma (with or without pain)

« No pain » groups (group 2 versus group 1): In the "Trauma" group (No pain-Trauma, group 2), dyschesia was less frequent (38.1 versus 88.9; P=0.0020) than in the "No pain-No trauma" group (group 1). Further, the rate of urge incontinence (65.8 versus 36.9%, P=0.020) was higher and so were the urinary frequency (99.1 ± 51.2 versus 128 ± 63.2 min, P=0.028) and the number of clinical signs (1.6 ± 1.2 versus 1.0 ± 1.1, P=0.0093). The only clinical sign whose prevalence was comparable in the two groups (with or without trauma) was "painful skin rolling test" (39.5 versus 36.9%, P=0.70).

« Pain » groups (groups 4-5-6 versus group 3): In the "Trauma" groups (groups 4-5-6), urge incontinence (25.7 versus 13.3%, P= 0.012), dysuria (43.9 versus 29.2%, P=0.0071), cystalgia (35.1 versus 24.8%, P=0.049), anal incontinence (42.9 versus 29.2%, P=0.036), proctalgia fugax (38 versus 25.7%, P=0.014) were more frequent than in the "Pain-No trauma" group (group 3). Likewise, Wexner's score values (10.1 ± 5.7 versus 8.0 ± 4.9, P=0.035) and the number of clinical signs were higher (2.3 ± 0.9 versus 1.8 ± 1.1, P =0.0027). The only clinical sign equally prevalent in the two groups (with or without trauma) was "painful skin rolling test" (71.9 versus 60.2%, P=0.094).

Differences between post-traumatic perineodynia groups

"Pain early onset-Trauma "versus "Pain late onset-Trauma": The only significant differences were a lower number of night time voids (nocturia, 0.60 ± 0.81 versus 1.05 ± 1.20, P=0.039) and a greater frequency of "painful skin rolling test" in the "Pain early onset-Trauma" group (85.7% versus 65.5%, P=0.022).

"Pain early onset-Trauma" versus "Pain before-Trauma": The NHI-CPSI score was greater in the "Pain before-Trauma" group (32.8 ± 6.0 versus 26.7 ± 7.8, P=0.037), while perineodynia VAS score tended to be higher (7.2 ± 1.8 versus 6.1 ± 1.9, P=0.055).

"Pain late onset-Trauma" versus "Pain before-Trauma": Urge incontinence (47.1 versus 20.2%, P=0.014) was more frequent and the perineodynia VAS score was greater (7.24 ± 1.79 versus 5.56 ± 1.99, P=0.0014) in the *"Pain before-Trauma"* than in the late onset group but for dyschesia (100% versus 76.6 %, P=0.051) only a tendency was observed.

Other early onset post-traumatic symptoms

Because post-traumatic symptom latencies were not systematically recorded, they could not be analyzed. For urinary symptoms, latencies between trauma and appearance of the problem were available but not specifically for each symptom. The group called lower urinary tract disorders (LUTD) included urinary incontinences (stress and urge), urinary frequency, urgency and dysuria. Of the 95 latency cases available, LUTD appeared just after pelvic trauma in 9 cases, 8 of which together with perineodynia ("Pain early onset-Trauma").

Latencies were also available for sexual troubles as a group (dyspareunia and sexual arousal syndrome). In 3 cases out of 50 available, sexual disorders appeared immediately after trauma (one of these cases was a sexual arousal syndrome associated with dyspareunia). The 3 cases arose together with perineodynia and LUTD.

Latencies for anal incontinence were available for 75 cases. This symptom appeared directly after trauma in 8 cases. In 2 of them, it was the only early onset symptom. Further, it was either associated with perineodynia, sexual troubles and LUTD (n=2), appeared together with perineodynia and LUTD (n=2) or it arose with perineodynia only (n=2).

Case reports

Patient 1 - A 53 year old female was driving through an intersection when her auto was struck on the driver's side by another auto traveling at a high rate of speed. Her automobile was lifted an estimated six feet in the air, rotated and landed upright. She was wearing a seat belt and air bags deployed. She suffered immediate pains in the left leg, left foot and left buttock. Pelvic fractures were not identified on CT scanner evaluation. Within 10 to 14 days, she noted perineal pain and a sensation of "sitting on one of her sitting bones". She developed urinary frequency and double voiding that had not been present in the past. There was gradual onset of persistent genital arousal. Underclothing began to cause the left labial pain. For several months, treatments focused on her left leg and foot and included a neurotomy of a distal portion of the sural nerve. Progressive increase in pelvic pains, originally ignored by caregivers, resulted in referral for evaluation. There was no history of previous pelvic pain. Since the MVA, she was unable to work as an accountant because sitting was too painful. The persistent pain impaired cognitive function. Pain distracted her thinking, reduced her mental acuity and affected her efficiency.

Sensory examination in the pudendal territory revealed hypoalgesia at the left side of the clitoris and hyperalgesia at the right side of the clitoris and left anal site. Pinprick was normal at the labia and right anal site. Pelvic floor descent was < 1 cm. There was mild erythema of the labia majora. The skin over the coccygeal area and natal cleft was smooth and pale suggesting trophic changes.

In the 16 months prior to diagnosis of pudendal neuropathy unsuccessful treatments had included sacral iliac joint block, caudal epidural steroid injection, sympathetic nerve block. She had slight, transient pain reduction from pelvic floor physical therapy and also when using a transcutaneous nerve stimulator.

Additional neuropathic pain generators were identified by using palpation and skin *rolling* [or "pinch roll"] test (thoracolumbar junction syndrome, ilioinguinal and iliohypogastric nerves bilaterally) [20] .

She had limited relief with a series of three pudendal blocks and was advised to undergo decompression surgery of the pudendal nerve. She is recovering from that operation with early improvement in her pudendal syndrome.

Patient 2 - A 56 year old woman was stopped at a traffic light when her heavy sports utility vehicle was struck from the rear by an auto traveling at approximately 56 km/h. Seat belt was being worn. She had immediate low back pain, a transient "electric shock" pain in the left lower quadrant and leg and a marked sensation of nausea. Neck and shoulder pain developed over a few days. Emergency evaluation included magnetic resonance imaging of the lumbar spine. No acute abnormalities were identified. She

had physical therapy, acupuncture and multiple medications for the low back and leg pains.

Five months after the MVA she began to notice rectal pain while driving her auto. Dyspareunia developed. Over several weeks the rectal pain became severe. She was unable to sit in her job. There were occasional episodes of fecal and urine incontinence. A foreign body sensation developed in the perineum. Pains became progressively worse and required use of oral morphine for control.

Eighteen months after the MVA the diagnosis of pudendal neuropathy was made by second author (SA). Pinprick caused hyperalgesia at the clitoris and labia (right > left). Skin changes of cutis anserina and dilated pores were observed over the coccygeal area. There was no prolapse and perineal descent with Valsalva maneuver was < 1.5cm. No additional pelvic pain generators were found at examination. The pudendal nerve terminal motor latency was prolonged bilaterally, showed temporal dispersion and caused hyperpathia (after sensation). The warm temperature threshold detection test was abnormal[10, 21].

A series of three pudendal nerve blocks was performed. Each relieved her pain but only for seven to ten days. Decompression surgery was recommended but was postponed because of insurance problems. After additional pudendal nerve blocks for pain control the surgery was completed five years after the MVA. Early, complete pain relief lasted only a few weeks. Rectal pains returned. She has not required post-operative pudendal nerve blocks. Central sensitization requires continued medications.

DISCUSSION

Pelvic trauma and pudendal syndrome

Based on case reports and clinical experience, pelvic trauma is a well-known trigger factor for pudendal neuralgia[7, 22]. This study is the first one comparing all symptoms of the pudendal syndrome in female patients with or without trauma of comparable age, parity and biometry.

History of pelvic trauma significantly increased frequency of urge incontinence, perineodynia, cystalgia, anal incontinence and proctalgia fugax. Wexner's score and NHI-CPSI score were also greater in case of trauma. Other symptoms of the pudendal syndrome (urinary frequency, dysuria, sexual arousal syndrome, dyspareunia) were more frequent after trauma but not significantly so because of the characteristics of the studied population (high prevalence of pudendal syndrome). Around 10% of LUTD, sexual troubles and anal incontinences began just after pelvic trauma.

Because the pudendal syndrome can be the consequence of a pelvic trauma and because it is rarely diagnosed, patients facing this problem after a professional or traffic accident have great difficulties to prove the link between trauma and sequelae that might persist for the rest of their life. In an attempt to explain the possible causal effect of pelvic trauma on pudendal syndrome, the expert could be guided by the seven Simonin's criteria which can be adapted for post-traumatic pudendal syndrome (these criteria are similar to those used in the English and American legal and medical systems to develop evidence of causation for both industrial problems and medical issues) [23-27]. Specifically, these criteria are:

C1. *The nature and intensity of the trauma*: pelvic trauma must be clearly documented and enough important to explain the lesion.

C2. *The quality and quantity of symptomatology*: the link between pelvic trauma and pudendal syndrome must be clinically possible and logical.

C3. *The coherence of impact-anatomical site-and type of injury su*stained: directly or closely related.

C4. *The natural history of the condition*: the symptoms have to follow a logic evolution since the trauma.

C5. *The elapsed time between event and complaint*: latency between trauma and pudendal syndrome must agree with the clinical experience.

C6. *The pre-morbid state of the claimant*: the pudendal syndrome didn't exist before the trauma.

C7. *Exclusion of another etiology of pudendal syndrome*: like a delivery, a trans-obturator surgery, biking or any other.

Regarding the importance of the trauma which could induce a pudendal syndrome (Criterion 1: Nature and intensity of the trauma), it is important to note that 89% of the trauma included in this study were not associated with a luxation or a fracture. It is therefore not necessary to have an organic – radiological lesion to have severe perineal sequelae.

Functional and/or organic damages to the pudendal nerves (and the other nerves involved in perineal innervation) after a pelvic trauma is easily understandable (Criterion 2) if one compares the pelvic nerves with electric cables in a car chassis (pelvic bones, fascia and muscles). Pudendal nerves have a tortuous and narrow path going between the anterior part of the piriformis muscles and the posterior border of the sacro-spinal ligament. Then passing through the interligamentary space between the sacrospinous (and its muscular part, the coccygeal muscle) and sacro-tuberous ligaments, through the fascia lunata and, at last enclosed in the Alcock's canal lying on the obturator muscle. Furthermore, some branches of the pudendal nerve may pass directly through the ligaments. Each trauma can create a distortion of the fibrous part (ligaments and fascia) of the pudendal nerve path. Because the chassis has moved, the electric cables are no longer in the same position and electrical (neuritic) short circuit can occur. Reflex muscle contractures, trigger points (piriformis, obturator, coccygeus, levator ani) directly induced by the trauma or secondary to neuralgias (the muscles tried to avoid any nerve movement), abnormal posture or psychological distress could be responsible for more nerve compression or stretch. In such a case, the nerve begins to swell and cannot move any more freely in its path giving rise to more pain. The vicious cycle is ongoing. Furthermore, pelvic hematoma or callus formation around a fracture may cause perineural scarring and compression. Results presented in Table II and Table III clearly show that the link between pelvic trauma and pudendal syndrome is clinically possible and logical (Criterion 2).

If the patient experienced a significant trauma that could have an impact on her pelvic bones (fall on the buttock-tailbone, traffic accident, leg fracture…), there is a coherence of impact-anatomical site-and type of injury sustained (Criterion 3).

For Criterion 4, the symptoms have to follow a logic evolution since the occurrence of the trauma (natural history of the condition). Of course, there is no problem in case of early onset post-traumatic symptoms. But, to fulfill this criterion for late onset post-traumatic symptoms, a prospective study should be considered to describe precisely the natural history of the condition (e.g. latency between pelvic trauma and each symptom occurrence, spontaneous evolution of the syndrome with time…).

Criterion 5 corresponds to the elapsed time between pelvic trauma and appearance of the pudendal syndrome. It is probably the most important one. This aspect has been studied carefully for perineodynia, one of the key symp-

toms of pudendal syndrome, because latencies were available for almost all the patients.

Pelvic trauma and perineodynia

Normally, pain is triggered by the stimulation of pain receptors (nociceptive pain) but in neuralgia (neuropathic pain) it occurs without excitation of these receptors and is caused by an abnormal change in the structure or function of the nerves. Innervation of the perineal skin is mainly provided by the pudendal nerves but other nerves are slightly involved (ilio-inguinal, genito-femoral, perineal branch of the posterior femoral cutaneous nerve, inferior cluneal).

Pudendal neuralgia is usually defined according to the Nantes criteria. These criteria are quite weak and they were never validated clinically. In fact, three of the five essential criteria are unreliable.

The first key criterion is an increase of pain while sitting. According to the Nantes team itself the same arises in case of inferior cluneal neuralgia but on harder seats [28] indicating that this sign is not specific.

The second one - Pain with no objective sensory impairment – does not reflect clinical reality. In fact, in Shafik's studies[5, 6] and in our personal experience[7, 8], pinprick hypoesthesia and hyperesthesia is often found in case of perineodynia. This is logical because nerve damage can be responsible for numbness or allodynia like in other parts of the body. Increase of warm detection threshold (reduced sensitivity to warmth) and qualitative abnormalities during this test (habituation, allodynia, dysesthesia, radiation or aftersensation) are also frequently encountered in case of pudendal (and other) nerve damage [21, 29].

A positive pudendal block (clear reduction of pain during anesthesia time) is a third weak criterion. According to the Nantes team itself: "a positive pudendal block is not specific as it simply indicates that the pain is situated in the territory of the pudendal nerve; pain related to any perineal disease (e.g., anal) would also be relieved by pudendal nerve block. A negative block does not formally exclude the diagnosis when it is not performed with sufficient precision"[30]. Therefore according to the Nantes team, this test is neither specific nor sensitive. It is even less reliable if perineal sensation is not checked 2 hours after the block [31].

The term perineodynia[8], a clear symptom, should be recommended rather than pudendal neuralgia, only one of the possible etiologies, because its definition criteria are weak and other nerves can be involved in perineal pain.

What about the cause and effect relationship in the 4 groups with a history of trauma?

The easiest situation is the early onset perineodynia ("Pain early onset-Trauma" group) in which pain arises just after pelvic trauma. In such a case, the cause – effect relationship could be evident (Criteria 1 to 5 positives) if there is no pre-existing pudendal syndrome in the medical history of the patient (Criterion 6) and if there is no other trigger factor (Criterion 7). Painful symptoms are frequently associated with perineodynia (cystalgia, dyspareunia and proctalgia fugax) together with urge incontinence, anal incontinence and dyschesia.

The medico-legal evaluation is the same in case of early onset post-traumatic LUTD, sexual troubles or anal incontinence without pain. This situation corresponds to patients in the "No pain-Trauma" group. More difficult are the "Pain late onset-Trauma" and "Pain before-Trauma" groups. In the "Pain before-Trauma" group, perineodynia means a problem already exists on the pudendal nerves (or other nerves involved in perineal sensibility) before pelvic trauma. Trauma just aggravates the pre-existing pudendal syndrome. The medico-legal discussion will be about a possible worsening of the syndrome after trauma. Our study suggest that it should be the case because the "Pain before-Trauma" group is notably different from the "Pain-No trauma" group. To conclude at a clear cause-effect relationship, the patient should ideally have an evaluation of the pre-existing pudendal syndrome to search for a clinical worsening.

In case of "Pain late onset-Trauma", it seems that the pudendal syndrome with trauma is worse than without trauma (with the same triggering factors). It is like if the trauma created a borderline state waiting for a trigger factor to develop more severe symptoms. Probably other symptoms of the pudendal syndrome already existed before pain appeared like in the "No Pain-Trauma" group which is possibly a step before "Pain late onset-trauma". In a medico-legal situation it is mandatory to ask about symptoms pre-existing before pain (especially all types of lower urinary tract dysfunctions, sexual arousal syndrome, anal incontinence and proctalgia fugax).

The 3 clinical signs of pudendal neuropathy-pudendal syndrome seem to be correlated with the severity of the syndrome. The skin rolling test is more frequently painful in case of perineodynia probably because it is explained by an allodynia to skin pinching. Pinprick sensibility is more frequently asymmetric in case of pelvic trauma maybe because it reflects a nerve compression or stretch.

Case reports

Case 1 - This woman did not notice immediate perineal pain. Its onset was minimally delayed (10-14 days) and should be considered as a "Pain early onset-Trauma" case. LUTS slowly developed. Persistent genital arousal appeared later. The time elapsed before diagnosis and treatment of the pudendal syndrome was 16 months which is quite typical. A correct diagnosis just after the MVA would have allowed avoiding useless treatments and unnecessary psychological and physical suffering. Furthermore, because the MVA was induced by another driver, the pudendal syndrome was part of the personal injury. This was completely ignored until treatment of the pudendal neuropathy was going on. Medico-legal actions were pursued however the long latency before a correct diagnosis caused significant medical expenses for the leg injury and loss of income.

Case 2 - Onset of perineal (rectal) pain, incontinence, and dyspareunia was delayed for several months ("Pain late onset-Trauma"). The diagnosis of pudendal neuropathy was not made for almost three years. In the meantime, treatment focused initially on back and leg pains. Legal action had started before the diagnosis of the pudendal syndrome. Delayed diagnosis of pudendal neuropathy caused loss of employment, personal stress and the expense of many medical interventions.

Study limitations and future prospects

This retrospective study has some limitations. The study population was characterized by a high prevalence of pudendal syndromes, prolapses and incontinences. Because of the retrospective aspect of the study, data concerning the type or even the existence of trauma and the timing of the symptoms was highly dependent on the patient's memory and data collection during the past consultations. Further, only the latency between trauma and perineodynia was available for analysis. Too many values of latency were lacking for lower urinary tracts, sexual and ano-rectal symptoms to make a comprehensive analysis and to draw firm conclusions.

A prospective study would be ideal. In such a study, after each pelvic trauma, clinical signs and symptoms (and questionnaires) of pudendal syndrome should be sought carefully and the follow-up duration should be at least 1 year. The patient should be warned to contact immediately the physician in case of any new perineal symptom. Latency between each symptom and the trauma should be listed to precisely characterize the classical evolution (probably different types) of the post-traumatic pudendal syndrome (Criterion 4).

Post-traumatic management of male patients should follow the same path. Particular attention is required to the appearance of impotence (partial or complete), ejaculatory impairment (loss of pleasure, pain), "prostatitis-like" pain and sexual arousal syndrome[3, 7, 32].

Faced with a medico-legal situation it is mandatory to be even more precise and in case of doubt to utilize a warm and cold detection test, a pudendal nerve terminal motor latency test and a complete electromyographic (EMG) exploration. Of course, characteristics of the trauma must be defined as precisely as possible (Criterion 1). The patient must be followed up carefully during at least 1 year and advised to inform her doctor in case of any new post-traumatic symptom.

CONCLUSION

According to this study, post-traumatic pudendal syndrome is a reality. Perineodynia, urge incontinence, anal incontinence, proctalgia fugax and cystalgia are the most frequently symptoms encountered. A complete history searching for any symptom of the pudendal syndrome and a clinical examination including its three clinical signs should be done after any significant pelvic trauma. In medico-legal situations it is suggested to add a warm and cold detection test and an EMG evaluation with latencies as soon as possible and one 1 year later in addition to the clinical control.

DISCLOSURES

This study has been approved by the Ethic Committee of Liège University. Jacques Beco, Stanley Antolak, Laurence Seidel and Adelin Albert have no conflicts of interest or financial ties to disclose.

REFERENCES

1. Shafik A, El-Sibai O, Shafik A et al. Role of pudendal canal syndrome in the pathogenesis of interstitial cystitis and its treatment by pudendal canal decompression. Curr Urol 2008, 2, 24-29.
2. Shafik A. Pudendal canal syndrome as a cause of vulvodynia and its treatment by pudendal nerve decompression. Eur J Obstet Gynecol Reprod Biol. 1998, 80, 215-20.
3. Shafik A. Pudendal canal decompression in the treatment of erectile dysfunction. Arch Androl .1994, 32, 141-9.
4. Shafik A. Pudendal canal decompression in the treatment of urinary stress incontinence. Int Urogynecol J Pelvic Floor Dysfunct. 1994, 5, 215-220.
5. Shafik A. Pudendal canal decompression in the treatment of fecal incontinence. Dig Surg, 1992, 9, 265-271.
6. Shafik A. Pudendal canal syndrome. Description of a new syndrome and its treatment. Report of 7 cases. Coloproctology. 1991, 13, 102-110.
7. Antolak S. Pudendal neuralgia: pudendal nerve entrapment, Alcock's canal syndrome, and pudendal canal syndrome. In: Potts JM, ed. Genitourinary pain and inflammation. Cleveland, USA: Humana Press, 2008.
8. Beco J, Climov D, Bex M. Pudendal nerve decompression in perineology : a case series. BMC Surg. 2004, 4, 15.
9. Robert R, Labat JJ, Bensignor M, et al. Decompression and transposition of the pudendal nerve in pudendal neuralgia: a randomized controlled trial and long-term evaluation. Eur Urol. 2005, 47, 403-8.
10. Beco J, Seidel L, Albert A. Normative values of skin temperature and thermal sensory thresholds in the pudendal nerve territory. Neurourol Urodyn. 2015, 34, 571-7.
11. Beco J, Mouchel J. Perineology or pelviperineology: the same goal but different approaches. Pelviperineology. 2007, 26, 139.
12. Mouchel J, Beco J. Perineology, the story behing the concept. Pelviperineology. 2007, 26, 185-186.
13. Beco J, de Bisschop G, Dijkstra R et al. [Perineology...reaching equilibrium and preserving it?]. J Gynecol Obstet Biol Reprod (Paris), 1999, 28, 855-6.
14. Beco J, Mouchel J, Nelissen G. La Périnéologie... comprendre un équilibre et le préserver. Verviers (Belgium): Odyssée 1372, 1998.
15. Hajebrahimi S, Corcos J, Lemieux MC. International consultation on incontinence questionnaire short form: comparison of physician versus patient completion and immediate and delayed self-administration. Urology. 2004, 63, 1076-8.
16. Vaizey CJ, Carapeti E, Cahill JA, et al. Prospective comparison of faecal incontinence grading systems. Gut. 1999, 44, 77-80.
17. Agachan F, Chen T, Pfeifer J et al. A constipation scoring system to simplify evaluation and management of constipated patients. Dis Colon Rectum. 1996, 39, 681-5.
18. Litwin MS, McNaughton-Collins M, Fowler FJ, Jr. et al. The National Institutes of Health chronic prostatitis symptom index: development and validation of a new outcome measure. Chronic Prostatitis Collaborative Research Network. J Urol. 1999, 162, 369-75.
19. Beco J. Interest of retro-anal levator plate myorrhaphy in selected cases of descending perineum syndrome with positive anti-sagging test. BMC Surg. 2008, 8, 13.
20. Antolak S. Interventions in Managing Male Pelvic Pain. In: Manchikanti L, Singh V, eds. Interventional Techniques in Chronic Non-Spinal Pain. Paducah KY: ASIPP Publishing, 2009, 303-318.
21. Antolak SJ, Antolak CM. Failed Sacral Neuromodulation: Simple Tests Demonstrate Pudendal Neuropathy. A Case Series. J Pelvic Med Surg. 2006, 12, 35-40.
22. Hibner M, Desai N, Robertson LJ, et al. Pudendal neuralgia. J Minim Invasive Gynecol. 2010, 17, 148-53.
23. Owen D. Products Liability Law. St Paul, Minnesota Thompson-West, 2008.
24. Hill A. The environment and disease: association or causation? Proc Roy Soc Med. 1965, 58, 295-300.
25. Sullivan JD. The medico-legal expertise: solid medicine, sufficient legal and a measure of common sense. Mcgill J Med. 2006, 9, 147-51.
26. Feron P. Vade-mecum de l'évaluation médico-légale: L'invalidité, l'incapacité, le handicap et le dommage corporel en droit belge. Bruxelles: De Boeck Université, 2000.
27. Rothman K, Greenland S. Modern Epidemiology. Philadelphia: Lippincott-Raven, 1998.
28. Darnis B, Robert R, Labat JJ et al. Perineal pain and inferior cluneal nerves: anatomy and surgery. Surg Radiol Anat. 2008, 30, 177-83.
29. Lindblom U. Analysis of abnormal touch, pain and temperature sensations in patients. In: Boivie J, Hansson P, Lindblom U, eds. Touch, Temperature, and Pain in Health and Disease: Mechanisms and Assessments Seattle, USA IASP press, 1994, 63-84.
30. Labat JJ, Riant T, Robert R et al. Diagnostic criteria for pudendal neuralgia by pudendal nerve entrapment (Nantes criteria). Neurourol Urodyn. 2008, 27, 306-10.
31. Antolak S, Jr., Antolak C, Lendway L. Measuring the Quality of Pudendal Nerve Perineural Injections. Pain Physician. 2016, 19, 299-306.
32. Waldinger MD, Venema PL, van Gils AP et al. Stronger evidence for small fiber sensory neuropathy in restless genital syndrome: two case reports in males. J Sex Med. 2011, 8, 325-30.

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Original article

Transperineal Bilateral Sacrospineous Colpofixation (TPBCF) for the treatment of vaginal vault prolapse – description of a refined method

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Abstract: Vaginal vault prolapse is observed with increasing frequency in the era of large aging populations. Various surgical techniques have been established, varying in performance, difficulty and outcome, specifically complications. In order to optimize both aspects, we have developed a refined transperineal bilateral sacrospineous colpofixation technique (TPBCF) and give a detailed, step-by-step description of the technique, with focus on the key differences to the "old" method of Intravaginal Sling (IVS). Importantly, we rely not on a transverse but instead on a longitudinal incision and blunt finger dissection to gain access to the sacrospinous ligament. Introducers for transischiorectal sling placement are guided from bilateral stab incisions lateral and dorsal from the anus (to avoid the rectal arteries and risk of arterial injuries) with the inserted finger, thereby ensuring that no undesired structures, mainly bowel, are injured by the advancing introducers during their passage through the pelvis. Preferably, horizontally oriented sutures are used to attach the sling to the underside of the vaginal apex or the posterior aspect of the cervix. As a minimally invasive approach with the potential for conservation of the uterus, our technique should be applicable to all age groups and the increasingly frequent elderly patient with significant co-morbidities.

Keyword: Transperineal bilateral sacrospinous colpofixation; Operative therapy; Transischiorectal vaginal sling placement; Vaginal vault prolapse.

INTRODUCTION

Vaginal vault prolapse is a known clinical entity observed increasingly frequently in the era of large aging populations[1]. Historically, treatment options included abdominal surgical interventions such as sacrocolpopexy or fascial slings[2,3] and operations via the vaginal approach such as the unilateral Amreich-Richter operation with the vaginal apex sutured to the sacral bone after hysterectomy.4 More recently, extensive reconstructions using prosthetic mesh for the induction of neo-ligaments and neo-fasciae have been advocated[5], sometimes also in the context of primary surgical interventions in the unpretreated patient[6].

Intravaginal slings (IVS) placed transischiorectally have been proposed by Petros and Farnsworth and shown to be promising in a small series of cases[7,8]. However, rectal injury and erosions were identified as major problems of this technique which led to the abandonment of IVS[9]. A multi-center series in Austria yielded better results but still described severe complications[10].

It appears that total lack of a formalized anatomically based procedure was a major contributing factor to these unfavorable outcomes, as well as a deficit of education in potential surgeons, potentially even amplified by encouragements and assurances of "simplicity" by the manufacturers. Several parts of the technical description itself already harbored the potential for major complications, for example the initial para-anal entry point at the three and nine o'clock positions where the rectal arteries is found.

In the development of our refined transperineal bilateral sacrospinous colpofixation (TPBCF) technique, we have strived to optimize the surgical procedure of transischiorectal vaginal sling placement regarding the anatomical and clinical outcome and the potential for complications. Here, we present the resulting surgical procedure and explain it step by step, with special emphasis on the aspects setting our TPBCF technique apart from the ill-fated posterior IVS.

SURGICAL TECHNIQUE

Step1: Pre-operative treatment. Each patient is treated with vaginal or systemic estriol application for four weeks before surgery. Single dose antibiotic prophylaxis with a combination of a cephalosporin and metronidazole is administered i.v. half an hour before starting the procedure. The vagina is thoroughly disinfected with copious amounts of antiseptic solution during the initial phase of the operation. The anus is thereafter covered with an adhesive sterile impermeable membrane and thereby sealed off from the operative field.

Step 2: Incision in the posterior vaginal wall. Two important changes have been made to the initial IVS approach when developing the TPBCF: A longitudinal (not transverse) incision is made in the midline of the posterior vaginal wall 3 cm distal to (not at) the vaginal apex. The injection of vasoconstringent medication under the vagina before incision may be considered, is, however, by no means necessary as significant bleeding is the exception when choosing this approach.

Step 3: Access to the sacrospinous ligament. A canal designated to admit the index finger of the surgeon is formed by advancing Metzenbaum scissors immediately under the vaginal wall horizontally in the direction of the pelvic side wall. By inserting the finger, a direct access to the sacrospinous ligament can thereafter be developed by blunt dissection. No extensive mobilization of tissue planes or retractor placement, nor visualization of the target structure is required at this point.

Step 4: Dissection of a horizontal space under the cranial vaginal tissue. From the upper end of the longitudinal vaginal incision, the tissues of the rectovaginal septum are dissected off the posterior aspect of the vaginal wall. This will facilitate the subsequent attachment of the prosthetic tape under the intact vagina, thereby removing it from the incision and thus from potential contamination during

wound healing and physiological inflammatory reactions, which both would predispose the tape to erosion. If the uterus remains in situ, the space is developed with the goal to expose the posterior surface of the cervix for later tape attachment.

Step 5: Choosing the entry points for the introducer on the buttocks of the patient.

The original description of transischiorectal sling placement involves a stab incision at the 3-and 9-o'clock positions 5 cm lateral to the anus. However, these are the precise locations of the rectal arteries. Therefore, the transischiorectal TPBCF placement uses bilateral stab incisions 3-5 cm lateral and 5 cm dorsal from the anus instead so that bilateral incisions 0.5 cm long are made in the perianal skin at 4 and 8 o'clock, halfway between the coccyx and external anal sphincter (EAS) in a line 2 cm lateral to the EAS.

Step 6: Introducer placement. It is a matter of personal preference, on which side the introducer is placed first (we used disposable DST Series EEA Introducer devices from Covidien, New Haven, CT). We have mostly placed it first on the left side of the patient and then on the right. This means that the tape is guided through from the surgeon's right to the left. Some surgeons may prefer the alternative sequence, especially during the short learning phase, as the sacrospinous ligament as the first anatomical entry point for the introducer is more removed from the bowel on the patient's right side than on the left.

From the small incision, the introducer is first guided horizontally above the fascia of Scarpa for a distance of approximately 5 cm and then oriented strictly cranially. This moves the introducer away from the rectal canal, and provides the benefit of later increasing the friction of the 1-cm-wide polypropylene monofilament sling (e.g. SERASIS from Serag-Wiessner, Naila, Germany) in the tissue. This mechanical resistance counteracts potential "pull-through" as the tape will not be sutured to the pelvic wall. The introducer is then advanced in the ischiorectal fossa peripheral to the levator plate until the desired exit point at the sacrospinous ligament is reached. The inserted finger marks the optimal entry point through the muscular structures and meets the tip of the introducer upon penetration of the muscle layer. The contact between introducer tip and finger tip should only be broken once the introducer has been advanced through the vaginal incision and its apex can be clearly seen. This "closed circle technique" ensures that no undesired structures, mainly bowel, are injured by the advancing introducer during its passage through the pelvis.

The blue stylus (from the introducer) or the tip of any alternative instrument is left in place and should be easily visible protruding into the vagina.

Step 7: Placement of the contralateral introducer. The insertion of the introducer on the contralateral side follows the same rules as above, except that after placement, the blue stylus is removed from the introducer and reinserted with its former tail end first, thereby exposing the opening at the end, which allows for threading the tape through the stylus. On this second side, the metal introducer remains in place. In this case, the tape will be guided outside-in-inside-out, while the use of alternative introducers would result in an inside-out-inside-out approach.

Step 8: Placement of the tape. At the time of placement of the second stylus, the tape is immersed in antibiotic solution. While realizing, that scientific data for this measure are lacking, we have still chosen a cephalosporin in combination with metronidazole to cover the expected potential germ spectrum in an effort to avoid clinically meaningful contamination of the tape by viable germs during placement. Gloves are changed by all team members before handling the tape.

The moist tape is now threaded through the opening of the first stylus which is situated outside the buttock. The stylus with the tape attached is then pulled through to the vagina with manual outside guidance to assure flat and undistorted orientation of the mesh. It is then detached from the stylus and threaded into the opening of the contralateral stylus which is still situated inside the introducer. An atraumatic forceps is used to stabilize the flat tape's orientation during this maneuver and the subsequent pulling through the contralateral pelvic side and out through the corresponding incision. Traction on the forceps additionally assures adequate mobility of the vaginal arch of the tape for suture fixation to the underside of the vaginal apex in the designated area of intact vaginal wall structure or the posterior aspect of the cervix, respectively. Two small instruments, e.g. Crile or short Kocher clamps, are used to mark the ends of the tape outside the patient.

Step 9: Fixation of the tape to the underside of the vaginal apex or the posterior aspect of the cervix. It is probably a more philosophical question, whether one should use resorbable or permanent suture for fixing the tape to the underside of the vagina. In an effort to assure suture stability during fibroblast invasion of the graft, while at the same time avoiding permanent multi-knotted strings under the vaginal skin, we have adopted the use of polydioxanone threads (USP 2-0) for this purpose. Three sutures are placed, the first in the midline, the second and third 2-3 cm lateral on either side of the midline. The suture technique involves threading the needle first through the graft, then through the tissue, and then out through the graft resulting in a U-shape with the branches both traversing the mesh.

A horizontal orientation of the sutures is preferred. An identical approach is used when fixing the mesh to the posterior cervix. Finally, the vaginal incision is closed with a running suture of resorbable braided material (USP 2-0 or 3-0).

Step 10: Considerations before definitive tape adjustment. At this point, if indicated, colporrhaphy can be performed, with the advantage that the uncorrected prolapse makes access to the vaginal walls easier.

Step11: Definitive tape placement. Immediately, or after additional vaginal surgeries have been completed (see Step 10), a sponge stick is inserted for positioning the vagina in the desired anatomical position. With the sponge still in place, the tape is pulled gently outward by symmetric bilateral traction on the marking clamps until the vagina is stabilized in its physiological position. The tape is then pulled out slightly and cut above skin level. The cutaneous wound margins are elevated with small surgical forceps to prevent mesh from attaching directly to the wound and eroding through the skin surface. Skin closure can be achieved by fibrous glue, one single interrupted suture, steristrips, or by simply mechanically adapting the skin margins with small Kocher clamps until the patient has been transferred back to her bed. All of the above options have been tried by our team, at the end we mostly reverted back to the traditional "one single interrupted".

Step 12: Preparation for postoperative care. At the end of the procedure, a vaginal gauze pack liberally coated

with estriol ointment is inserted into the vagina for 24 hours together with a Foley catheter for bladder drainage. If outpatient treatment is desired, which is definitely an option due to the excellent tolerability of the intervention, this step can probably be safely omitted. In any case, weekly vaginal estriol applications are prescribed, as known from other clinical management guidelines after vaginal mesh placement.

DISCUSSION

Fascia lata slings and suspension procedures using the round ligaments have been abandoned as have resorbable meshes due to the fact, that the body does not maintain neo-ligaments without continuing stimulation of fibroblasts on site. Sacrocolpopexy with or without prosthetic mesh interposition should be combined with a Burch procedure for optimal results as shown by the studies of the NIH Pelvic Floor Disease Network[11-13]. In sum, this amounts to a significant surgical intervention with laparoscopic techniques adding their own spectrum of possible complications due to their transabdominal nature.

Amreich-Richter results are known for their surgery-induced dyspareunia, deep pelvic pain and secondary urinary continence problems[14] making them unattractive especially for, but not limited to, the younger patient. While having been in clinical use for a long time, systematic studies of this entity are few. Modifications using unilateral or bilateral non-resorbable sutures that serve as fixing strings suspending the vaginal apex at a distance from the sacrum have never been formally evaluated and remain experimental with anecdotal results.

Large prosthetic implants as a primary treatment approach for female genital prolapse are meeting with increased scepticism due to their potential for complications. The FDA has recently issued a statement to the effect, that large meshes are contraindicated as primary treatment in such situations[15].

The TPBCF approach outlined here offers the potential for the generation of an anatomy-analogous support of the vaginal vault or the uterus mimicking the sacrospinous ligaments or creating sacro-vaginal ligaments in it's place. The challenge for the surgeon adopting the procedure will be to overcome a possible initial hesitancy when faced with the insertion of the introducer into the ischiorectal fossa, but in our experience the procedure becomes routine quickly. The indication for TPBCF is vaginal vault or uterine prolapse, it is not designed to correct anterior, posterior or lateral pelvic floor defects. As a minimally invasive approach with the potential for conservation of the uterus TPBCF would potentially be applicable to all age groups and also the increasingly frequent elderly patient with significant co-morbidities.

ACKNOWLEDGEMENTS AND DISCLOSURES

The authors declare no conflict of interest.

REFERENCES

1. Wilkins MF, Wu JM. Lifetime risk of surgery for stress urinary incontinence or pelvic organ prolapse. Minerva Ginecol 2017; 69: 171-7.
2. Takacs EB, Kreder KJ. Sacrocolpopexy, Surgical Technique, Outcomes, and Complications. Curr Urol Rep 2016; 17: 90.
3. Ijland MM, Fischer D-C, Kieback DG, McGrath G, Farnsworth B. Midline intravaginal slingplasty for treatment of urinary stress incontinence, Results of an independent audit up to 2 years after surgery. Int Urogynecol J Pelvic Floor Dysfunct 240 2005; 16: 447-54.
4. Lantzsch T, Goepel C, Wolters M, Koelbl H, Methfessel HD. Sacrospinous ligament fixation for vaginal vault prolapse. Arch Gynecol Obstet 2001; 265: 21-5.
5. de Ridder D. Should we use meshes in the management of vaginal prolapse? Curr 244 Opin Urol 2008; 18: 377-82.
6. Dias FGF, Dias PHGF, Prudente A, Riccetto C. New strategies to improve results of mesh surgeries for vaginal prolapses repair – an update. Int Braz J Urol 2015; 41: 623-34.
7. Farnsworth BN. Posterior intravaginal slingplasty (infracoccygeal sacropexy) for severe posthysterectomy vaginal vault prolapse – a preliminary report on efficacy and safety. Int Urogynecol J Pelvic Floor Dysfunct 2002; 13: 4-8.
8. Petros PP. Medium-term follow-up of the intravaginal slingplasty operation indicates minimal deterioration of urinary continence with time. Aust N Z J Obstet 253 Gynaecol 1999; 39: 354-6.
9. Feiner B, Jelovsek JE, Maher C. Efficacy and safety of transvaginal mesh kits in the treatment of prolapse of the vaginal apex, A systematic review. BJOG 2009; 116: 15-24.
10. Bjelic-Radisic V, Hartmann G, Abendstein B, Tamussino K, Riss PA. The posterior intravaginal slingplasty operation, Results of the Austrian registry. Eur J Obstet Gynecol Reprod Biol 2009; 144: 88-91.
11. Brubaker L, Cundiff GW, Fine P et al. Abdominal sacrocolpopexy with Burch colposuspension to reduce urinary stress incontinence. N Engl J Med, 2006; 354, 1557–66.
12. Barber MD, Brubaker L, Burgio KL et al. Comparison of 2 transvaginal surgical approaches and perioperative behavioral therapy for apical vaginal prolapse, The OPTIMAL randomized trial. JAMA 2014; 311: 1023-34.
13. Barber MD, Brubaker L, Nygaard I et al. Defining success after surgery for pelvic organ prolapse. Obstet Gynecol 2009; 114: 600-9.
14. Kraus P, Krofta L, Krčmář M et al. Řešení sestupu tří kompartmentů pomocí syntetického implantátu a sakrospinózní fixace, Kohortová prospektivní studie s délkou follow-up pěti let [The results of five years follow-up prospective study of vaginal prolapse repaired by prolift total mesh surgery or sacrospinous fixation]. Ceska Gynekol 2017; 82: 277-86.
15. FDA. Urogynecologic Surgical Mesh: Update on the Safety and Effectiveness of Transvaginal Placement for Pelvic Organ Prolapse, 2011. (https://www.fda.gov/downloads/Medical Devices/Safety/AlertsandNotices/UCM2

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Original article

Pathophysiology and diagnosis of descending perineum syndrome in children

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Abstract: *The purpose* of this study is to offer a safe, simple and accurate method for diagnosing descended perineum syndrome (DPS). *Methods:* 194 patients aged from five days to 15 years were surveyed in the Belarusian Center of Pediatric Surgery. They were divided into two groups. In the 1st group were 65 patients without abnormalities of the anorectal area. In the 2nd group were 129 patients, including 66 children with functional constipation, 55 children with anorectal malformations (ARM) with visible fistulas that were examined before surgery, and 8 patients with ARM investigated after surgery. All patients underwent barium enema, which differed from the standard study by the presence of a radiopaque marker placed near the anus. *Results and Conclusion:* DPS is caused by damage to the muscles and ligaments of the pelvic floor. A method of assessing DPS is proposed on the basis of changes in the distance between the anorectal angle and the marker near the anus. Administering a barium enema with a minimum of radiographs dramatically reduces the dose of ionizing radiation compared with defecography and contributes to a more accurate assessment of DPS not only for adults, but also for children with chronic constipation, fecal incontinence and ARM, both before and after surgery, allowing assessment of the causes of complications.

Keywords: Barium enema; Chronic constipation; Descending perineum syndrome; Defecography; Fecal incontinence; Puborectalis muscle.

INTRODUCTION

The term "descending perineum" or "pelvic floor descent" is used in two very different conditions of the anorectal region. It was first applied by Parks et al, describing normal defecation during defecography. When defecation takes place there is an increase in the anorectal angle and the "descent" of barium from the rectum to the anus. Parks et al also described a "syndrome of the descending perineum", which is defined as a pathological condition detectable during defecography. In these cases the anorectal angle increases and descends caudal to the pubococcigeal line[1]. The etiology of descending perineum syndrome (DPS) is unclear[2]. Defecography is considered the gold standard for identifying DPS, but in recent years dynamic magnetic resonance imaging has increasingly been used for this purpose[3].

Parks et al believed that the cause of increased perineal descent is excessive straining upon defecation, causing the anterior wall to protrude towards the anal canal, in turn inducing incomplete defecation and weakness of the pelvic floor muscles[4]. Other authors have noted that abnormal descent of the perineum not only causes constipation, but also frequently causes fecal incontinence, anal pain, and other symptoms[3]. Defecography has two major disadvantages. First, it is associated with a high dose of ionizing radiation, which is unacceptable for the examination of children and patients of childbearing age. Secondly, the measurement of the anorectal angle or perineal descent with defecography has poor reproducibility, and differences exist between examiners[2].

The purpose of this study is to offer a safe, simple and accurate method of diagnosing DPS.

METHOD

Patients aged from five days to 15 years were divided into two groups. In the 1st group were 65 patients with complaints of abdominal pain, or where there was a suspicion of space-occupying lesions in the abdomen. Pathology of the colon and anorectal area was excluded by barium enema and proctoscopy. The results of the survey in this group of patients were accepted as the norm. The second group consisted of 129 patients, including 66 patients aged from 11 months to 15 years with functional constipation. Hirschsprung's disease was excluded on the basis of manometric and histochemical studies. This group also included 55 patients aged from 10 days to 12 years who were examined before surgery and had anorectal malformations (ARM) with visible fistulas. Among them were seven boys. Perineal fistulas were found in six girls, and 42 patients had fistulas opening into the vestibula. 47 patients had chronic constipation, and fecal incontinence was noted in four cases. This group also included eight patients with APM, who were examined after surgery to determine the cause of chronic constipation and/or fecal incontinence.

BARIUM ENEMA

X-ray study. During barium enema a contrast medium was used to fill the colon at least up to the splenic flexure. A radiopaque marker of 1.6 cm diameter with a hole in the center was strung onto the tip of the enema. During the study, patients in the control group had the marker located in contact with the anus, and in patients of the second group with ARM it was located near the fistula. In addition, in children of the second group another radiopaque marker was glued to the place of EAS activity. In patients of the first group, on a lateral radiograph of the anorectal zone we measured the roentgen negative distance (RND) between the barium in the rectum (or anorectal angle) and the marker on the posterior contour of the tip of the enema, and the width of the rectum at the widest part of its vertical branch (Figure 1).

To determine the true anatomical size, all distances measured on radiographs were multiplied by the coefficient of the projection increase. The latter is the ratio of the true diameter of the marker (1.6 cm) to its size on the radiograph image.

The manometric study was carried out with the help of an endotracheal tube, without the use of a rectal balloon. This allowed detection of an inhibitory anorectal reflex in patients with ARM with visible fistulas and the performing of anorectal manometry during the barium enema[4].

Statistical analysis was performed by the method of the Student's t-test. Statistical significance was defined as $P < 0.05$.

RESULTS

First group. On lateral radiographs of the anorectal area the distance between the barium in the rectum and the marker near the anus (RND) increased from 1.7 cm in neonates to 3.9 cm in adolescents, and the width of the rectum increased from 1.3 to 4.6 cm (Table 1).

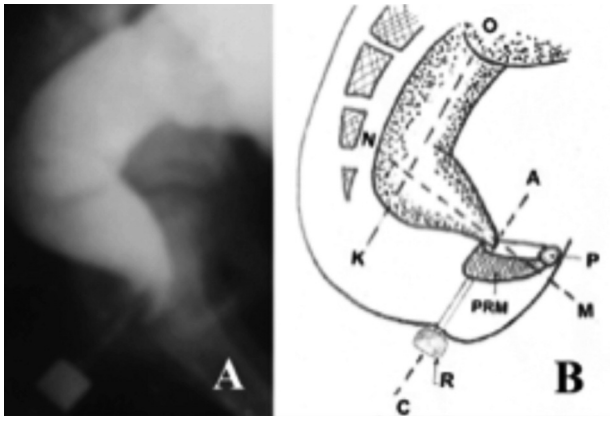

Figure 1. – Lateral x-ray image of the anorectal zone of a healthy child **(A)** and a schematic representation **(B)**. The axis of the anal canal (AC) is displaced anteriorly from the axis of the vertical branch of the rectum (OK). NM – axis of the horizontal branch of the rectum; PRM – puborectal muscle, P – pubis, R – 1.6 cm diameter radiopaque marker in proximity of the anus. The distance between the rectum and the marker lying along the posterior contour of the tip is the length of the anal canal.

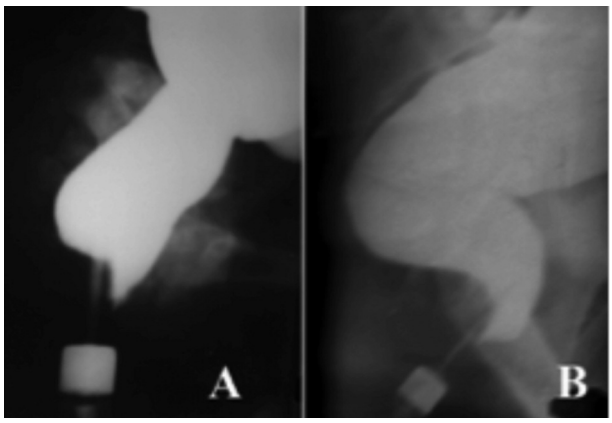

Figure 2. – Radiological representation of the rectoanal inhibitory reflex. Penetration of barium from the rectum into the upper part of the anal canal in front of the enema tip as a result of relaxation of the internal anal sphincter. The posterior wall of the anal canal at this level is pressed against the tip of the enema by the contracted Puborectalis muscle. **A.** four month old child. **B.** A 15 year old teenager.

The RND is a zone of contraction of the muscles of the perineum, which prevents leakage of barium from the rectum. Its length is equal to the length of the anal canal according to the results of manometric studies[5,6]. This proves that the RND is due to anal canal contraction.

Starting from the second half of the year, the caudal end of the rectum is bent forward, resulting in the formation of a horizontal branch of the rectum and an acute anorectal angle (ARA) between the rectum and the anal canal. Its proximal point, i.e. rectoanal compound is near the pubococcygial line. A caudal point, i.e. anus, is determined on the roentgenogram by the location of the marker contacting it. Thus, the RND between the rectum and the marker is an abridged anal canal, whose length represents the length of the anal canal.

The injection of air into the rectum through the endotracheal tube during manometric study caused a momentary decrease in anal pressure of 10-15 mm Hg, which was restored to baseline level within 7-15 seconds (a positive rectoanal inhibitory reflex). In 10 cases in which contrast agent was injected into the rectum during barium enema through the endotracheal tube of the manometric device, a transient penetration of contrast agent into the upper part of the anal canal in front of the enema tip was observed, while the posterior wall tightly pressed to the tip (Figure 2).

TABLE 1. The normal width of the rectum and anal canal length in children of differing ages add caption.

Age	Width of the rectum (cm)	Length of the anal canal (RND) (cm)
	n - 12	7
5 days - 11 months	1.3 – 3.0	1.7 – 2.5
	2.24±0.09	2.21±0.15
	9	7
1-3 years	3.0 – 3.7	2.3 – 2.8
	3.21±0.11	2.55±0.10
	9	8
4-7 years	3.0 – 3.9	2.5– 3.6
	3.43±0.14	3.17±0.14
	9	8
8-10 years	3.2 – 4.1	2.6 – 3.7
	3.72±0.05	3.11±0.10
	19	18
11-15 years	3.6 – 4.6	3.1 – 3.9
	3.95±0.07	3.43±0.10

This area occupies 40 to 50% of the entire length of the RND. Within a few seconds, the barium which penetrated into the anal canal returned to the rectum. In the process of the colon filling periodically (one to three times) penetration of contrast medium into the anal canal in front of the enema tip was observed. This lasted a few seconds each time before the barium disappeared from the anal canal. Appearance of barium in the anal canal is always accompanied by a decrease in anal pressure of 10-15 mmHg. When the barium disappeared from the anal canal, the pressure increased back to the basal level.

SECOND GROUP

In two of 66 patients with functional constipation who were surveyed in the first year of life, the width of the rectum was within normal limits. In four patients older than one year, the X-ray pattern did not differ from the norm. These were children with disease durations of less than six months, regularly receiving treatment (enemas and/or laxatives). In other cases a significant expansion of the rectum with respect to age norms was determined. The horizontal branch of the rectum on the lateral radiograph is not differentiated, due to the expansion anteriorly

TABLE 2. Size of the rectum and anal canal in 66 children with functional constipation. P – the reliability of the results compared to the standards in Table 1.

Age	Width of the rectum (cm)	Length of the anal canal (RND) (cm)
Up to 11 months	n - 2	1
	2.9	2.5
	11	8
1-3 years	2.9 – 4.8	2.2 – 3.7
	3.70+/- 0.17	3.09+/-0.15
P	< 0.02	< 0.05
	18	8
4-7 years	2.5 – 6.5	2.2 – 3.7
	4.68+/-0.20	3.17+/-0.14
P	< 0.001	> 0.2
	25	22
8-10 years	3.9 – 7.5	2.3 – 4.6
	5.14+/-0.18	3.21+/-0.12
P	< 0.001	> 0.2
	9	9
11-15 years	4.5 – 8.6	3.0 – 4.2
	5.90+/-0.38	3.44+/-0.15
P	< 0.001	> 0.2

of its vertical branch. Width of the rectum and RND length in different age groups are presented in Table 2.

X-ray study of the anorectal area in each case allows a fairly accurate determination of the function of the anal canal. Three stages of pathological change can be clearly distinguished. These follow one another, each with an increase in the degree of megarectum (Figure 3).

Only in 10 (20%) of 51 studies was the length of the RND within normal limits. In 14 (27%) patients it was longer than the maximum normal limit (Fig. 3, A). The lengthening of the RND can be explained by hypertrophy of the puborectalis muscle (PRM), whose increase in volume causes not only pressure on the posterior wall of the anal canal, but also the distal part of the rectum which pressed to the tip of the enema. Only in the initial stage of the disease, up to three years, is a significant ($P < 0.05$) elongation of the RND defined. In patients older than three years, the average length of the RND was in the normal range ($P > 0.2$), but individual values varied widely. In fifteen (29%) of 51 patients barium penetrated into the anal canal behind the enema tip (Figure 3, B), indicating a weakened PRM was unable to press the posterior wall of the anal canal to the enema tip. In 12 (24%) of the patients a significant shortening of the RND was determined compared to a normal length of the anal canal (Figure 3 C and D). This shortening was sometimes combined with an increase of ARA (Figure 3, B). In other cases, ARA was normal (Figure 3, D).

For babies up to nine months old, in ARM with fistulas on the perineum or vestibule the anorectal area before surgery differed from the norm by the presence of anterior displacement of the anus. The length of the RND between the ARA and a marker near the fistula, or a marker located near activity of the subcutaneous portion of the external anal sphincter, was equal to the length of the age norm of the anal canal ($P > 0.2$). In cases where it was possible to insert and keep the manometric device's endotracheal tube within the rectum, a rectoanal inhibitory reflex was detected. Radiological and manometric data confirmed histological and embryological studies of other authors that this pathology is anal ectopy. In children older than one year, expansion of the rectum occurs due to stenosis of the ectopic anus. This is accompanied by shortening of the RND between the ARA and the marker near the fistula. This process increases with age (Figure 4).

After endorectal pull-through (Figure 5, A) or posterior sagittal anorectoplasty (Figure 5, B) a significant (twofold or more) shortening of the RND was observed compared with the normal length of the anal canal. This was combined with expansion of the rectum beyond the maximum normal limit. In cases where the anal canal was used during surgery to reconstruct the ARM, the length of the anal canal was within normal limits (Figure 5, C).

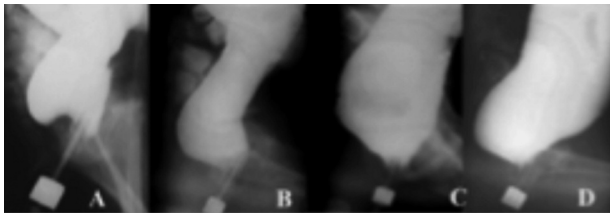

Figure 3. – Radiographs of the anorectal area of children with functional constipation. The true diameter of the marker is 1.6 cm. On all pictures the horizontal branch of the rectum is absent, as its vertical branch is greatly expanded compared to the age norm. **A.** The length of the RND is 3.5 cm (normal maximum length of the anal canal at the age of three years is 2.8 cm); **B.** The length of the anal canal is within the age limit, but barium penetrates into the anal canal behind the tip of the enema as a result of weakness of the PRM; C and **D**. Significant expansion of the rectum is combined with a sharp shortening of the anal canal and insufficiency of the PRM.

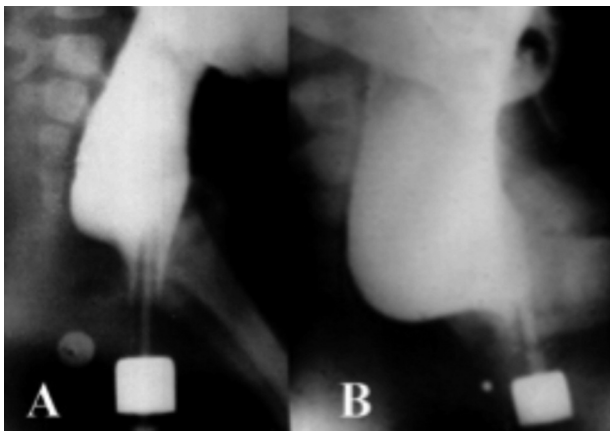

Figure 4. – Lateral radiographs of the anorectal area of the same patient with vestibular fistula. **A**. At the age of six months. Penetration of barium behind the tip of the enema is observed, testifying to weakness of the PRM. However, the length of the RND is within the limits of the age norm. **B**. At the age of one year and two months. Expansion of the rectum is combined with a sharp shortening of the RND between the marker near the activity of the external anal sphincter (fraction) and the ARA.

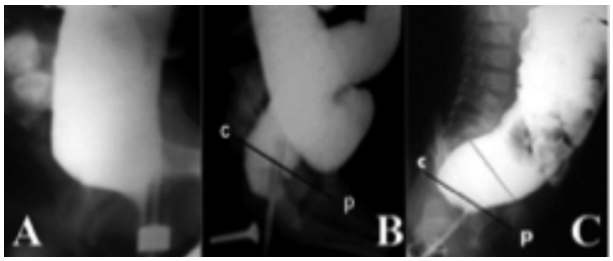

Figure 5. – Radiographs of patients with ARM after surgery. **A.** After pull-through procedure. Megarectum combined with the lack of PRM action. There is efflux of barium outward from the anal canal. **B.** After posterior sagittal anorectoplasty. Sharp shortening of the anal canal indicates failure of the PRM. **C**. After anterior sagittal anorectoplasty. Megarectum in conjunction with the normal length of the anal canal.

DISCUSSION

In healthy subjects the RND is the end zone of the intestinal tract, which in a contracted state prevents leakage of barium from the rectum. Its length ranges from 1.7 cm in newborns to 3.9 cm in adolescents, which according to manometric study corresponds to the length of the anal canal ($P < 0.01$)[5,6]. It follows that RND is a closed anal canal, and its length is equal to the length of the anal canal. On the radiograph, the highest point of the anal canal is at its junction with the rectum, i.e. at the zero point of the anorectal angle (ARA). In 1953, F. Stephens determined that the place of transition where the rectum passes into the anal canal is at the level of the pubococcygeal line, outstretched from the coccyx to the lower contour of the pubic bone[7]. Anorectal inhibitory reflex is often seen as a reflex relaxation of the internal anal sphincter (IAS) in response to pressure on the rectal wall[8]. However, it is called an inhibitory reflex because despite relaxation of the IAS, emptying of the rectum does not occur, due to contraction of the PRM and the external anal sphincter (EAS)[9].

With high definition manometry during the rectoanal inhibitory reflex, the peak pressure of the IAS is seen 1.6 cm from the anal verge, which corresponds to the peak relaxation pressure. In addition, two peaks of high pressure are determined. The lower concentric peak is located between 0.5 and 1 cm from the anal margin, caused by contraction of EAS.

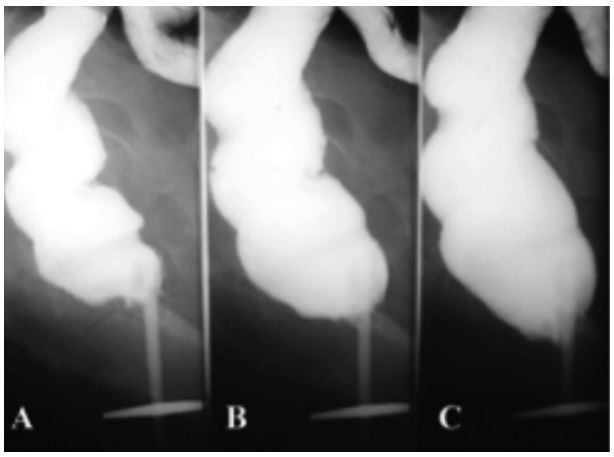

Figure 6. – Stages of rectal filling in a 68 year old patient. **A.** When filling starts the rectum is narrow and the ARA is acute. Barium enters into the anal canal behind of the tip of the enema. Length of RND is equal to 3.8 cm. **B.** Rectum widened, and RNA has become shorter. The ARA is right angled. **C.** At the end of the study the rectum has expanded to a greater extent. The RND has become two times shorter than on Figure 6, A. The ARA has become obtuse.

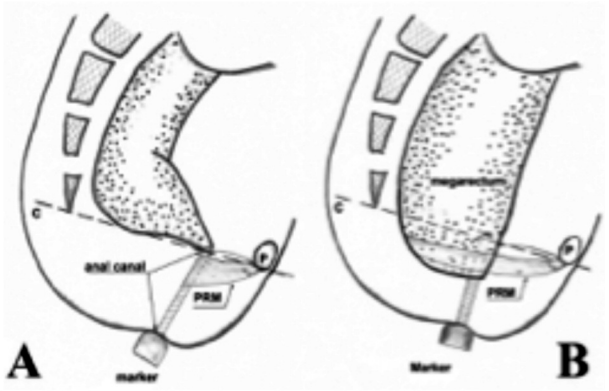

Figure 7. – Scheme of measuring descending perineum from the lowest point of the anal canal, i.e. from ARA to marker, rather than from the upper point. **A**. Healthy patient. The upper point of the anal canal is at the intersection of the axis of the anal canal with pubococcygeal line. The length of the anal canal is within the normal range. **B**. Patient with chronic constipation and "descending perineum" (see Figure 5 diagram, D). The length of the RND (functioning anal canal) is almost two times shorter than the norm.

The second peak pressure is recorded on the posterior wall between 2.4 cm and 4 cm from the anal margin. This is due to contraction of the PRM[10]. Thus, the length of the PRM's contraction zone occupies 40% of the proximal part of the anal canal on the posterior wall, which in our study corresponds to the clamping zone of the posterior wall of the anal canal to the tip of an enema.

When the rectum was filled through the manometric device's tube, we found that anal canal pressure decreased during penetration of barium into the anal canal in front of the tip of the enema. At this time, barium does not penetrate into the anal canal behind the tip, because the posterior wall of the anal canal presses to the tip of the enema as a result of contraction of the PRM. It follows that penetration of barium into the anal canal just in front of the tip of the enema is a radiological manifestation of the rectoanal inhibitory reflex.

If ARA in the tension and the more at rest descends relative to the pubococcygeal line, it is denoted by the term "descending perineum syndrome", and indicates that the PRM is weak and does not perform its function. According to Baek et al. descending perineum syndrome, combined with chronic constipation, results from intense tension during bowel movements over a period of several years. It is also observed in faecal incontinence, idiopathic anal pain syndrome, solitary rectal ulcer and rectal intussusception, including rectal prolapse. The degree of the perineal descent can be measured in centimeters on radiographs performed during defecography, as caudal displacement of the ARA relative to the pubococcygeal line. If descent of the ARA is observed at rest, it is defined as a fixed descending perineum[11].

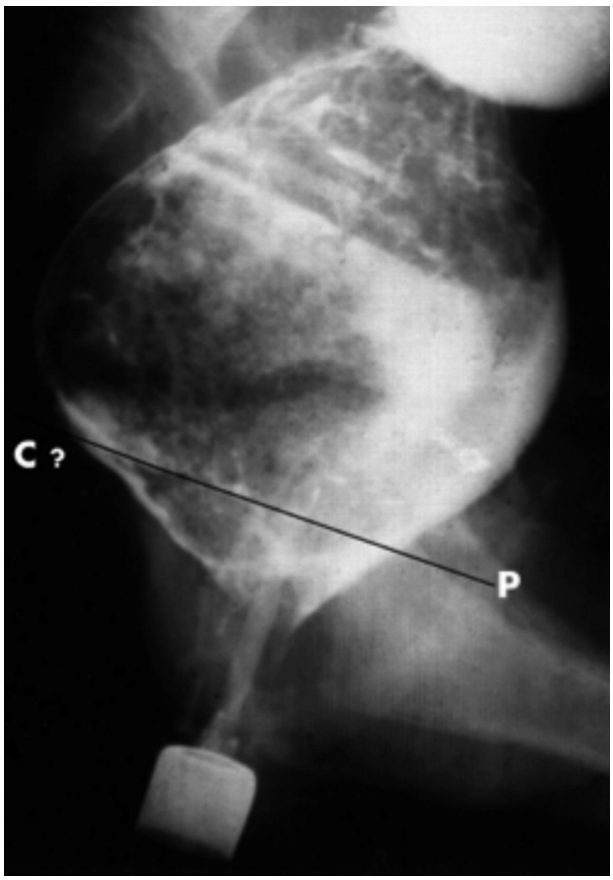

Figure 8. – 10 year old patient with functional constipation and damage to the PRM. Peristalsis of the rectum tries to expel an 8 cm fecal stone through the anal canal. Perineal descent is approximately 1.2 cm. There is shortening of RND length (2.4 cm), compared with the normal anal canal length at this age (3.2 cm).

In mild cases, when the rectum starts filling with contrast medium the length of the RND is equal to the normal length of the anal canal. However, after the introduction of an additional amount of barium the rectum expands and the pressure therein increases. If the PRM is weak, barium begins penetrating into the anal canal along the posterior contour enema tip, and the RND is shortened to two times shorter than its normal length (Figure 6).

This observation is an example of a non-fixed descending perineum. In its early stage the PRM presses the posterior wall of the anal canal to the tip. But when the pressure rises, this cannot be sustained due to PRM weakness. As a result, the upper part of the anal canal opens fully and fills with barium.

Because of the high doses of ionizing radiation involved, defecography is not normally used in child radiology. We have found only one study[12] since 2003. Another disadvantage is the low reliability of defecography. On figure 3, C and D with a sharp descending perineum shows that ARA can be obtuse and right angled. Furthermore, defining the top of a normal ARA is very difficult, and is not always accurate because the coccyx and pubis are not always visible on the X-ray.

Application of radiopaque markers near the anus and knowledge of the normal length of the anal canal allow accurate determination of the "perineal descent", not relative to the upper point of the anal canal, but with respect to its lower point (Figure 7). To obtain accurate information about the state of the PRM it is sufficient to make one lateral radiograph of the anorectal area following administration into the colon of between 200 to 500 ml (depending on age) of barium suspension.

To assess defecography on the selected image from the video, we need to find the lower contour of the pubic bone and the last vertebra. This is difficult, especially in anorectal anomalies when there is pathology of the spine. Measuring the distance from the ARA to the marker, we define the same displacement, but without the need to seek landmarks that ensure the accuracy of the measurement. Introducing a rather large amount of barium to the intestine causes high pressure in the rectum, the need to defecate and maximal tension for continence. This load on the PRM provokes its relaxation in cases of weakness.

This technique is especially useful in determining the cause of postoperative complications after surgery for anorectal anomalies. For example, in Figure 7, the anal canal is disclosed showing barium leaking because the PRM and IAS do not function. The state of the EAS is difficult to judge, since it cannot remain in a contracted state for more than one minute. Figure 7, B shows descending perineum syndrome with lack of PRM contraction following posterior sagittal anorectoplasty. Figure 5, C shows megarectum following anterior sagittal anorectoplasty. In this patient all sphincters are functioning normally (PRM, IAS and EAS). Chronic constipation is associated with damage from the act of defecation. The anal canal was divided from the levator ani muscle, which should normally open the anal canal, but being cut off from IAS it failed to disclose the anal canal during defecation. The cause of PRM damage in children with functional constipation is similar to women after childbirth[2]. In figure 8 it is easy to calculate the diameter of the fecal stone, bearing in mind that the diameter of the marker is 1.6 cm. Thus, the diameter of the fecal stone is 8 cm. Passage of such a wide fecal stone through the anal canal in a child 7-10 years old can damage the PRM to the same degree as passage through the birth canal of a newborn's head with a diameter of 10-12 cm.

Stretching and weakness of the PRM in functional constipation and after childbirth is the visible tip of the iceberg. In previous studies we found that in patients with functional constipation[13] and anorectal malformations[4], shortening of the RND in descending perineum syndrome was accompanied by violation of the opening of the anal canal during defecation. This indicates that the function of the levator ani muscle, which normally opens the anal canal during defecation[14], is disturbed too. Petros and co-authors have shown the role of stretching the pelvic floor ligaments in the pathogenesis of urinary incontinence in women[15]. Thus, all tissues of the pelvic floor are stretched and weakened in descending perineum syndrome.

CONCLUSION

Descending perineum syndrome is caused by damage to the pelvic floor muscle and ligaments. Our modification of the barium enema differs from the standard methodology by the presence of a radiopaque marker near the anus. Using this method we determined the length of the anal canal and the rectal width in children of different ages without pathology of the colon and anorectal area. A method of assessing descending perineum syndrome according to the distance between the anorectal angle and the marker near the anus is proposed. This contributes to a more accurate assessment of "descending perineum syndrome" and negates the need for defecography. Use of a barium enema with a minimum of radiographs dramatically reduces the dose of ionizing radiation. This method can be applied not only to adults, but also to children with chronic constipation, fecal incontinence and anorectal malformations both before and after surgery, to assess the causes of complications.

DISCLOSURES

I am the only author of this article. In the process of working, I did not receive any help, including financial assistance. Therefore, there can not be any conflicts of interest. This scientific work is a retrospective analysis of the results of a standard X-ray study made at the end of the last century, so there is no need for a decision of the ethics commission. This article has not been sent to other journals.

REFERENCES

1. Parks AG, Porter NH and Hardcastle J. The syndrome of the descending perineum. Proc R Soc Med. 1966 Jun; 59 (6): 477-82.
2. Chang J, Chung SS. An analysis of factors associated with increased perineal descent in women. J Korean Soc Coloproctol. 2012 Aug; 28 (4): 195-200.
3. Foti PV, Farina R, Riva G, Coronella M et al. Pelvic floor imaging: comparison between magnetic resonance imaging and conventional defecography in studying outlet obstruction syndrome. Radiol Med. 2013 Feb; 118 (1): 23-39.
4. Levin MD. Levin MD. The pathological physiology of the anorectal defects, from the new concept to the new treatment. Eksp Klin Gastroenterol. 2013; (11): 38-48.
5. Kumar S, Ramadan S, Gupta V et al. Manometric tests of anorectal function in 90 healthy children: a clinical study from Kuwait. J Pediatr Surg. 2009 Sep; 44 (9): 1786-1790.
6. Gruppo Lombardo per lo Studio della Motilita Intestinale. Anorectal manometry with water-perfused catheter in healthy adults with no functional bowel disorders. Colorectal Dis. 2010 Mar; 12 (3): 220-225.
7. Stephens FD. Imperforate rectum. A new surgical technique. Med J Australia. 1953; **1**: 202.
8. Ruttenstock EM, Zani A, Huber-Zeyringer A, Höllwarth ME. Pre- and postoperative rectal manometric assessment of patients with anorectal malformations: should we preserve the fistula? Dis Colon Rectum. 2013 Apr; 56 (4): 499-504.
9. Palit S, Lunniss PJ, Scott SM. The physiology of human defecation. Dig Dis Sci. 2012 Jun; 57 (6): 1445-64.
10. Cheeney G, Nguyen M, Valestin J, Rao SS. Topographic and manometric characterization of the recto-anal inhibitory reflex. Neurogastroenterol Motil. 2012 Mar; 24 (3): e147-54.
11. Baek HN, Hwang YH, Jung YH. Clinical Significance of Perineal Descent in Pelvic Outlet Obstruction Diagnosed by using Defecography. J Korean Soc Coloproctol. 2010 Dec; 26 (6): 395-401.
12. Zhang SC, Wang WL, Liu X. Defecography used as a screening entry for identifying evacuatory pelvic floor disorders in childhood constipation. Clin Imaging. 2014 Mar-Apr; 38 (2): 115-21.
13. Levin MD, Degtyarov IG. Perspective of the functional constipation treatment in children. Surgery News (Belarus). 2011; 2: 113-124.
14. Levin MD, Troyan VV. Anatomy and physiology of anorectal zone. Hypothesis of continence and defecation. Novosti chirurgii. 2009; 17 (2): 105-18.
15. Petros P, Swash M, Bush M, Fernandez M, at al. Defecation 1: Testing a hypothesis for pelvic striated muscle action to open the anorectum. Tech Coloproctol. 2012 Dec; 16 (6): 437-43.

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Original article

Descending perineum syndrome: pathophysiology of fecal incontinence

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Abstract: *Aim of the study:* Patients with descending perineum syndrome (DPS) may also experience fecal incontinence. This study was undertaken to understand the types and pathophysiology of fecal incontinence in patients affected by DPS. *Methods:* Two groups of DPS patients were investigated. Group 1 included 62 women who had fecal incontinence and descending perineum. Group 2 consisted of 70 female patients affected by descending perineum but without fecal incontinence. All 132 patients received a preliminary clinical evaluation and were studied using defecography, endoanal ultrasound and anorectal manometry. Their results were compared with data obtained from 20 healthy women with normal bowel habits (control group). *Results*: There was a significantly higher Fecal Incontinence Severity Index (FISI) score in Group 1 compared to Group 2 ($P < 0.001$). Urge incontinence was present in 56.4% of Group 1 patients, passive incontinence in 13 patients (20.9%), mixed incontinence in 16.1% and post-defecatory incontinence in only 4 patients (6.4%). Endoanal ultrasound revealed a significantly diffuse thinning of the external anal sphincter (EAS) in Group 1 patients ($P < 0.02$) with a linear relationship between signs of EAS atrophy and FISI score (p_s: 0.78; $P < 0.03$); EAS atrophy was also inversely correlated with anal pressure (p_s: 0.71; $P < 0.04$). Anal resting pressure (P_{max} and P_m) was significantly lower in Group 1 patients than Group 2 patients ($P < 0.04$). The pelvic floor descent values in Group 1 patients were significantly higher at rest and during evacuation ($P < 0.01$) than in Group 2 patients. *Conclusions*: Urge incontinence, related to external anal sphincter atrophy, is the predominant pattern of fecal incontinence.

Keywords: Descending perineum syndrome; Fecal incontinence; Pelvic organ prolapse; Rectoanal intussusception; Anorectal manometry.

INTRODUCTION

Descending perineum syndrome (DPS) is a complex syndrome where signs and symptoms are an expression of all the pelvic-perineal areas that are involved [1]Although it was first described by Parks *et al*. in 1966[2], as being characterized by the ballooning of the perineum several centimeters below the bony outlet of the pelvis during straining, many other attempts to define this syndrome and its symptoms have been published since that time. Descending perineum, according to Park's definition, refers only to the external perineal plane which is joined to the descent of the deep perineal plane, thus involving the whole pelvic floor and pelvic content. Straining at stool, a sensation of incomplete evacuation and sometimes of anorectal obstruction/blockage, manual maneuvers to facilitate defecation, and loss of solid or liquid stool are mixed with vaginal (sensation of a bulge, heaviness) and urological (urinary incontinence, urgency, hesitancy, feeling of incomplete emptying) symptoms[1]. The involvement of all visceral pelvic content is testified by dynamic magnetic resonance imaging which shows different grades of pelvic organ prolapse, including the rectum, combined with pelvic floor relaxation and functional evidence of impaired defecation[3]. Clinically, fecal incontinence appears late during the course of descending perineum syndrome and overlaps with obstructed defecation[1,4].

The aims of this study, conducted on patients affected by descending perineum syndrome, were 1) to describe the clinical profile of fecal incontinence and 2) to identify the main pathophysiological mechanisms of fecal incontinence.

MATERIALS AND METHODS

Between January 2010 and October 2015, 1261 patients affected by anorectal diseases were seen at the outpatient unit of the surgery clinic of the University of Florence. All patients were entered into a prospectively constructed database, which contained 2878 patients at the time of the study. This research was a retrospective, data-mining study investigating the differences between 2 groups of patients. Exclusion criteria were inflammatory bowel disease, proctitis, anal intercourse, rectal prolapse, previous pelvic and anal surgery, pelvic radiation, neurological disorders and cognitive impairment. One hundred thirty-two female patients [age range 47-78 years (median age 63.2 years)] affected by descending perineum syndrome identified according to Parks' and colleagues criteria[2] and negative colonoscopy were included in this study. All 132 patients received a preliminary clinical evaluation and were studied using defecography, endoanal ultrasound and anorectal manometry. Two groups of patients were identified and assigned to two study arms regarding the presence or absence of fecal incontinence. Group 1 was made up of 62 women [age range 55-79 years (median age 67.5 years)] who had fecal incontinence and descending perineum. Group 2 consisted of 70 female patients affected by descending perineum but without fecal incontinence [age range 43-73 years (median age 58.1 years)]. We compared their results with data obtained from 20 healthy women, age range 50-70 years (median age 60.4 years) with normal bowel habits (control group). They perceived their defecation behavior as normal and had never visited a physician for intestinal problems.

All procedures performed in this study were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study.

Clinical evaluation

Information regarding number of bowel movements/week, stool form according to Bristol scale[5], symptoms of obstructed defecation according to Bartolo[6] and pathological conditions were collected from previous outpatient charts. Obstructed defecation was classified according to the obstructed defecation syndrome (ODS) score[7]. Fecal incontinence was defined as the uncontrolled passage of fecal material recurring for > 3 months; number of fecal incontinence episodes for week was noted. Fecal incontinence was

classified according to the Fecal Incontinence Severity Index (FISI) score[8]. The pattern of incontinence was defined according to Engel's criteria[9]: fecal incontinence was classified as urge incontinence, passive incontinence, mixed incontinence, and post-defecatory incontinence. We reviewed chart data and recorded deliveries, noting obstetric tears and episiotomy, and degree of genital relaxation assessed with the Pelvic Organ Prolapse Quantification system (POP-Q) [10]. Inspection of the ano-perineal region and digital rectal examination were carried out to detect any signs of organic disease.

Defecography

All the patients and controls underwent defecography, according to the methods suggested by the Italian Working Team[11]. It was performed with the patient at rest, during squeeze, and during expulsion of the barium. All the X-rays showed latero-lateral views. The radiological measurements included the anorectal angle (measured and expressed in degrees between the longitudinal axis of the anal canal and the tangential line to the posterior rectal wall) and pelvic floor descent (defined as the vertical distance between the pubococcygeal line and the anorectal junction). The latter was expressed in millimeters. Qualitative evaluation was made by noting barium trapping, rectoanal intussusception, and the persistence of the puborectalis indentation during evacuation. Rectocele was also identified as a herniation of the anterior wall of the rectum into the vagina; the dimension was measured in millimeters and was defined as the distance between the tip of the rectocele and the longitudinal axis of the anal canal. Rectocele displacement with abnormal pelvic floor descent at evacuation was noted[12]. Rectoanal intussusception was also graded according to the Oxford Radiological grading of rectal prolapse[13].

Endoanal ultrasound

Endoanal ultrasound was performed using a three-dimensional multifrequency (9-16 MHz) probe

(Flex-Focus 1202; B-K Medical, Herlev, Denmark; endoprobe 360° rotating type 2050, proximal-to-distal length of 6 cm) with the patient in the left-lateral position. The probe was introduced into the anus to the level of the anorectal verge and slowly withdrawn. A defect of the internal (IAS) or external anal sphincter (EAS) was defined as a discontinuity of the muscle with an area of mixed echogenicity due to replacement of muscle cells by fibrous tissue. The sphincter defect was measured in degrees. Sphincter quality was described as homogeneous or heterogeneous if signs of sphincter atrophy were present. External anal sphincter atrophy was defined as diffuse thinning and/or replacement of muscle fibers by fat. Internal anal sphincter atrophy was identified as diffuse thinning of the sphincter.

Anorectal manometry

Computerized anorectal manometry was performed in all patients using previously published standard techniques[12]. Maximal anal pressure (Pmax) and mean pressure (Pm) of the anal canal expressed anal resting pressures (ARP). The computer quantified the amplitude (MVC-A) in millimeters of mercury (mmHg) and duration (MVC-T) in seconds of the maximal voluntary contraction (MVC). The rectoanal inhibitory reflex (RAIR) was elicited according to Martelli et al.[14]. The first distension volume at which internal sphincter relaxation occurred (RAIR threshold, RAIRT) and the distension volume for which an initial transient sensation occurred [conscious rectal sensitivity threshold, (CRST)] were determined in all patients. The maximal tolerated volume (MTV) was also measured in all patients and was considered an expression of rectal reservoir capacity. Compliance of the rectum (expression of the ratio mmHg/ml of inflated air) was detected by means of the pressure/volume curve. The manometric procedure was completed by measuring anal pressures when the patient was asked to attempt defecation (straining test). The straining test was considered positive if an inappropriate increase or < 20% relaxation of basal resting pressure occurred (respectively types I and III, describing dyssynergic defecation, according to Rao *et al*.)[15].

Statistical analysis

The sample size adequacy and statistical power of the study were calculated (DSS Research: statistical power calculator). The results are expressed as the mean ± standard deviation (SD). Student's t-test for paired and unpaired samples was used for statistical analyses. All correlations were evaluated using Spearman's rank correlation coefficient (rho: ρ_S), where full correlation is 1 and correlation < 0.50 is considered not significant. A $P > 0.05$ was chosen for rejection of the null hypothesis.

RESULTS

The sample size was adequate (adequacy: π=0.84) to achieve a statistical power of 100%. Patients' clinical characteristics are reported in Table 1. The number of bowel movements/week was not significantly higher in Group 1 patients than in Group 2 subjects and 45 Group 1 patients had > 3 fecal incontinence episodes/week. 17.7% of Group 1 patients had loose stool while 40% of Group 2 patients had hard stool ($P < 0.001$). There was a significantly higher FISI score in Group 1 compared to Group 2 ($P < 0.001$). Urge incontinence was present in 56.4% of Group 1 patients, while passive incontinence was predominant in 13 (20.9%), mixed incontinence in 16.1% and post-defecatory incontinence in only 4 patients (6.4%). A significantly higher ODS score was found in Group 2 patients compared to those of Group 1 ($P < 0.01$). Obstetric tears and/or episiotomy were recorded in 112 women (84.8 %) without significant differences between Group 1 and Group 2. Only 5 of all 132 study women had no uro-gynecological problems: 50 had cystoceles (37.8%) and 77 had uterine pro-

TABLE 1. Patients' clinical characteristics.

Symptom - Sign	Study cohort (n = 132)	Group 1 (n = 62)	Group 2 (n = 70)
Stool frequency (n/week)	7.7 ± 6.1	10.8 ± 2.9	6.3 ± 3.6
Fecal incontinence episodes (n/week)	2.4 ± 1.1	5.2 ± 1.6*	0
Hard stool (Bristol scale 1 or 2)	33	5	28°
Loose stool (Bristol scale 6 or 7)	13	11*	2
FISI score	11.7 ± 7.3	21.4 ± 8.7*	0
ODS score	6.9 ± 4.2	5.7 ± 2.3	11.8 ± 3.1°
Deliveries	1.3 ± 1.1	1.6 ± 0.7	1.2 ± 1.0
Obstetric tears	62/132	33/62	29/70
Episiotomy	50/132	23/62	27/70
Cystoceles	50/132	24/62	26/70
Uterine prolapse	77/132	47/62§	30/70
Urinary incontinence	42/132	19/62	23/70

*Group 1 patients vs Group 2 patients: $P < 0.001$
° Group 2 patients vs Group 1 patients: $P < 0.01$
§ Group 1 patients vs Group 2 patients: $P < 0.05$

TABLE 2. Results of endoanal sonography and anorectal manometry*Group 1 patients vs Group 2 patients and Controls: P < 0.02.

	Controls (n = 20)	**Group 1 (n = 62)**	**Group 2 (n = 70)**
IAS thickness (mm)	3.1 ± 0.5	2.4 ± 0.4	2.7 ± 0.8
EAS thickness (mm)	5.2 ± 0.7	2.5 ± 0.7*	4.3 ± 0.5
Pmax (mmHg)	88.2 ± 10.1	41.8 ± 18.0°	70.6 ± 21.9
Pm (mmHg)	47.8 ± 7.6	20.6 ± 8.2°	40.7 ± 13.4
MVC – A (mmHg)	200.0 ± 20.0	135.3 ± 25.8°°	158.2 ± 25.5
MVC – T (sec)	35.6 ± 4.2	14.1 ± 11.9°°	24.2 ± 10.2
Straining test (patients positive/ total patients)	1/20	15/62	39/70§
RAIRT (ml)	30.7 ± 10.8	25.0 ± 2.1	28.3 ± 8.1
CRST (ml)	40.7 ± 10.3	85.3 ± 24.4**	61.3 ± 27.7
MTV (ml)	205.1 ± 23.4	198.2 ± 51.1	183.3 ± 36.6

° Group 1 patients vs Group 2 patients and Controls: P < 0.04
°° Group 1 patients vs Group 2 patients and Controls: P < 0.01
§ Group 2 patients vs Group 1 patients: P < 0.01
**Group 1 patients vs Group 2 patients and Controls: P < 0.05

lapse (58.3 %). Urinary incontinence was present in 42 patients (31.8%): 29 (69.0%) had stress incontinence. Anorectal physical examinations revealed grade II hemorrhoids in 7 Group 1 and in 9 Group 2 patients. No anal fistulas or fissures were detected.

The results of endoanal ultrasound and anorectal manometry are reported in Table 2. Endoanal ultrasound revealed a significant diffuse thinning of EAS in Group 1 patients ($P < 0.02$) with a linear relationship between signs of EAS atrophy and FISI score (ρ_S: 0.78; $P < 0.03$); EAS atrophy was also inversely correlated with anal pressure (ρ_S: 0.71; $P < 0.04$). There was an internal anal sphincter disruption in 14 Group 1 patients (22.5%) and in 9 Group 2 patients (12.8%); EAS defects (width: 71.4 ± 18.5°) were detected in 22 Group 1 patients (35.4%) and in 13 Group 2 patients (18.5%). Anal resting pressure (P_{max} and P_m) was significantly lower in Group 1 patients when compared to Group 2 patients and controls ($P < 0.04$). MVC amplitude and duration in both groups were significantly different from controls ($P < 0.01$). The straining test was positive in 39 Group 2 patients (55.7%) and this was significantly different from that of Group 1 patients ($P < 0.01$). A significantly higher CRST was found in Group 1 patients ($P < 0.05$) in comparison to Group 2 patients and controls; CRST was also significantly higher than RAIRT in both groups when compared to those of controls ($P < 0.01$). There were no significant differences in maximal tolerated volume and rectal compliance in either patient group.

Defecographic data are reported in table 3. The anorectal angle was significantly greater in Group 1 when compared to group 2 and controls ($P < 0.01$). The pelvic floor descent values in Group 1 patients were significantly higher at rest and during evacuation ($P < 0.01$) than in Group 2 patients and controls. Eighty-four patients had a poor anorectal angle opening at evacuation and 72 patients (54.5%) had puborectalis indentation: Group 2 patients had a higher incidence of puborectalis indentation when compared to Group 1 ($p < 0.05$). Rectoanal intussusception, with Oxford Grade III and Grade IV, was noted in 102 (77.2%) of all patients and in 77 of these (75.4%) the rectoanal intussusception was combined with rectocele. Rectocele was present in 86 patients (65.1%) altogether and 9 patients showed a displacement rectocele alone that was > 3 cm. Six patients showed signs of enterocele, 4 had sigmoidocele.

TABLE 3. Defecographic data.

	Controls (n = 20)	**Group 1 (n = 62)**	**Group 2 (n = 70)**
Anorectal angle (degrees)			
Resting	94.5 ± 3.1	109.6 ± 1,5*	93.8 ± 3,8
Evacuation	110.1 ± 3.4	127.3 ± 6.6*	112.4 ± 5.1
Pelvic floor descent (mm)			
Resting	17.3 ± 7.2	43.3 ± 3.5*	32.1 ± 3.0
Evacuation	25.2 ± 2.5	54.3 ± 5.6*	38.7 ± 6.5
Rectocele			
Affected Patients	0	46/62	37/70
Size (mm)	0	27.0 ± 5.2	24.7 ± 9.2
Rectoanal intussusception			
Affected Patients	0	50/62	52/70
Puborectalis indentation			
Affected Patients	0	28/62	44/70°
Enterocele			
Affected Patients	0	2/62	4/70
Sigmoidocele			
Affected Patients	0	1/62	3/70

*Group 1 patients vs Group 2 patients and Controls: P < 0.01
° Group 2 patients vs Group 1 patients: P < 0.05

DISCUSSION

Descending perineum syndrome is characterized by the involvement of all visceral pelvic content. Many clues suggest that obstructed defecation slowly evolves into fecal incontinence. The initial phase of obstructed defecation sustained by the pelvic floor dyssynergia evolves, over some years, into organic changes associated with the appearance of mobile descent of the pelvic floor[1]. In our study, 54 patients (40.9%) had a positive straining test, and 84 patients (63.6%) had a poor anorectal angle opening at evacuation, both of which are diagnostic signs of dyssynergic defecation. Signs of impaired rectal sensations are also present: CRST was higher in both groups in comparison to that of controls. This report underlines the presence of obstructed defecation in DPS: rectal sensory perception is in fact blunted or absent in the majority of patients with obstructed defecation[16].

The progressive decline in normal levator ani tone, induced by defecatory overstretching, results in an open urogenital hiatus, weakening of the horizontal orientation of the levator plate, and a bowl-like configuration[17]. The consequence is impairment of manometric and defecographic signs of pelvic floor dyssynergia that no longer appear in long-term DPS forms. Such anatomical arrangements are also seen in women with pelvic organ prolapse: the ballooning of the levator hiatus and the increase in the levator plate angle are imaging signs of pelvic floor impairment in pelvic organ prolapse[18,19]. These anatomical changes help to explain the coexistence of uro-gynecological and proctologic pathologies in descending perineum syndrome, a condition also present in our patients, 96.7% of whom had urogynecological problems. The presence of episiotomy, POP, and urinary incontinence indicate the participation of the urinary and gynecological districts. Unfortunately they are not the object of our paper but it might be useful to further investigate this topic in the future.

In parallel, the natural history of descending perineum evolves thusly: the pelvic floor, upset by progressive pudendal neuropathy and pelvic-perineal muscle flabbiness, results in fecal incontinence, which at first is partial and eventually total, and is combined, in one way or another, with obstructed defecation[20]. Our incontinent patients had obstructed defecation, as testified by their ODS score, and

the close connection between impaired defecation and fecal incontinence illustrates the late evolution of the syndrome. Although our Group 1 patients were, in fact, older than Group 2 subjects (Group 1: median age 67.5 years; Group 2: median age 58.1 years), this difference was not significant, probably because the speed of DPS development was different between subjects. In our study, the FISI score in Group 1 was lower than that reported as clinically significant for impaired quality of life by Cavanaugh et al[21]. This is an indirect demonstration that even if a cut-off value is proposed the clinical reality is different: these Group 1 patients sought medical care even though their fecal incontinence was moderate. Urge incontinence, either alone (56.4%) or combined with passive incontinence (20.9%) was the predominant pattern of fecal incontinence. Thinning of the EAS, with impaired maximal voluntary contraction, was the main reason for this urge fecal incontinence[20] but several factors contemporaneously came into play: EAS defects, rectoanal intussusception, loose stool and high defecatory frequency were also all present, in various combination, in our Group 1 patients. Their pelvic floor descent was significantly higher at rest and during evacuation ($P < 0.01$) when compared to Group 2 patients, being a demonstration that DPS was evolving towards a worse stage marked by the clinical appearance of fecal incontinence. There was also a high incidence (80.6%) of rectoanal intussusception in both Group 1 and Group 2 patients. Rectoanal intussusception has a complex etiology: damaged pelvic floor muscles, mechanism of a sliding hernia, structural defects of pelvic fascia, ligaments and connective tissue, are all mixed with impaired defecation as a consequence of pelvic floor dyssynergia with vector force lines and high intra-abdominal pressures canalized into the Douglas cul-de-sac[22]. When rectoanal intussusception is combined with levator hiatal widening and levator plate descent, it can become the morphological pathology underlying DPS fecal incontinence[22]. It is difficult to provide a single pathophysiological framework for DPS fecal incontinence. Although a multifactorial etiology seems have an impact on a weak pelvic floor, it is very difficult to understand how much a single factor may destabilize the descending perineum. Surely a descending perineum possesses *per se* a pathological structure of pelviperineal muscles, perineal body and supportive elements of the endopelvic fascia that can lead to fecal incontinence. For example, lax suspensory ligaments that inactivate striated pelvic muscle forces[23], increased collagen breakdown such as a pathological etiology of urinary incontinence and pelvic organ prolapse[24], the observation that 45% of patients with joint hypermotility, stool evacuatory disorders and abnormal connective tissue also have fecal incontinence not due to sphincter dysfunction[25], are all evidence that an impaired pelvic floor may be associated with fecal incontinence.

The weakness of the study is the absence of follow-up regarding the appearance or absence of fecal incontinence but it must be considered that follow-up would have to be very long, lasting several decades, in order to show the complete evolution from obstructed defecation to fecal incontinence. Moreover, we decided not to introduce a second control group, i.e. those with fecal incontinence but without DPS, into the study because we felt that these patients would have had very diverse etiologies with pathophysiological profiles differing from case to case: this population would not be homogeneous, and therefore not useful for the purposes of comparison.

In conclusion, fecal incontinence inevitably will materialize with the passage of time in patients affected by descending perineum syndrome, characterized by hypotonia of the pelvic floor and associated pudendal neuropathy,. It is obvious that many factors may influence the evolution towards fecal incontinence and its velocity, such as childbirth[26], pelvic surgery[27], recto-anal surgery[28], anal sphincter lesions[29], radiotherapy[30] and neurological diseases[31], which all can have a negative impact on continence function, and may thus lead to the pathophysiological mechanisms of fecal incontinence. Knowledge of the physiopathology of fecal incontinence is the prerequisite for proper treatment of the patient.

REFERENCES

1. Pucciani F. Descending perineum syndrome: new perspectives. Tech Coloproctol 2015; 19: 443-448.
2. Parks AG, Porter NH, Hardcastle J. The syndrome of descending perineum. Proc R Soc Med 1966; 59: 477-482.
3. Comiter CV, Vasavada SP, Barbaric Z et al. Grading pelvic prolapse and pelvic floor relaxation using dynamic magnetic resonance imaging. Urology 1999; 54: 454-457.
4. Bartolo DC, Read NW, Jarratt JA, Read MG, Donnelly TC, Johnson AG Differences in anal sphincter function and clinical presentation in patients with pelvic floor descent. Gastroenterology 1983; 85: 68-75.
5. O'Donnell LJ, Virjee J, Heaton KW. Pseudo-diarrhoea in the irritable bowel syndrome: patients' records of stool form reflect transit time while stool frequency does not. Gut 1988; 29: A1455.
6. Bartolo D, Roe A. Obstructed defaecation. Br J Hosp Med 1986; 35: 228-236.
7. Altomare DF., Spazzafumo L, Rinaldi M et al. Set-up and statistical validation of a new scoring system for obstructed defaecation syndrome. Colorectal Dis 2008; 10: 84-88.
8. Rockwood TH, Church JM, Fleshman JW et al. Patient and surgeon ranking of the severity of symptoms associated with fecal incontinence: the fecal incontinence severity index. Dis Colon Rectum 1999; 42: 1525-1532.
9. Engel AF, Kamm MA, Bartram CI et al. Relationship of symptoms in faecal incontinence to specific sphincter abnormalities. Int J Colorect Dis 1995; 10: 152-155.
10. Bump RC, Mattiasson A, Bo K et al The standardization of terminology of female pelvic organ prolapse and pelvic floor dysfunction. Am J Obstet Gynaecol 1996; 175:10-17.
11. Piloni V, Genovese M, Grassi R et al. National working team report on defecography. Radiol Med (Torino) 1993; 85: 784-893.
12. Pucciani F, Rottoli Ml, Bologna A et al. Anterior rectocele and anorectal dysfunction. Int J Colorect Dis 1996; 11: 1-9.
13. Collinson R. Rectal intussusception and unexplained faecal incontinence: findings of a proctographic study. Colorectal Dis 2009; 11: 77-83.
14. Martelli H., Devroede G, Arhan P et al. Some parameters of large bowel motility in normal man. Gastroenterology 1978; 75: 612-618.
15. Rao SS, Mudipalli RS, Stessman M, et al. Investigation of the utility of colorectal function tests and Rome II criteria in dyssynergic defecation (Anismus). Neurogastroenterol Motil 2004; 16: 589-596.
16. Gosselink MJ, Schouten MR. Rectal sensory perception in females with obstructed defecation. Dis Colon Rectum 2001; 44: 1337-44.
17. Singh K, Jakab, M, Reid et al. Three-dimensional magnetic resonance imaging assessment of levator ani morphologic features in different grades of prolapse. Am J Obstet Gynecol 2003; 188: 910-15.
18. DeLancey JOL, Hurd WW. Size of the urogenital hiatus in the levator ani muscles in normal women and women with pelvic organ prolapse. Obstet Gynecol 1988; 91: 364-68.
19. Hsu Y, Summers A, Hussain HK et al. Levator plate angle in women with pelvic organ prolapse compared to women with normal support using dynamic MR imaging. Am J Obstet Gynecol 2006; 194: 1427-433.
20. Pucciani F. Editorial. A theory of progression from obstructed defecation to fecal incontinence. Tech Coloproctol 2015; 19: 713-715.

21. Cavanaugh M, Hyman N, Osler T. Fecal incontinence severity index after fistulotomy: a predictor of quality of life. Dis Colon Rectum. 2002 Mar; 45 (3): 349-53.
22. Pucciani F. Rectal prolapse: pathophysiology. In Altomare DF, Pucciani F (eds). Rectal Prolapse. Milan, Springer; 2008: 13-19.
23. Petros P. Diagnosis. In: Petros P (ed) The female pelvic floor. Function, dysfunction and management according to the integral theory. Heidelberg: 3rd Edn. Springer; 2010: 77-117.
24. Chen BH, Wen Y, Li H, Polan ML. Collagen metabolism and turnover in women with stress urinary incontinence and pelvic prolapse. Int Urogynecol J Pelvic Floor Dysfunct 2002; 13: 80-87.
25. Mohammed SD, Lunniss PJ, Zarate N et al. Joint hypermobility and rectal evacuatory dysfunction: an etiological link in abnormal connective tissue? Neurogastroenterol Motil 2010; 22: 1085-e283.
26. Murad-Regadas SM, Fernandes GO, Regadas FS. Assessment of pubovisceral muscle defects and levator hiatal dimensions in women with faecal incontinence after vaginal delivery: is there a correlation with severity of symptoms? Colorectal Dis 2014; 16: 1010-1018.
27. Pucciani F, Boni D, Perna F et al. Descending perineum syndrome: are abdominal hysterectomy and bowel habits linked? Dis Colon Rectum 2005; 48: 2094-2099.
28. Varpe P, Huhtinen H, Rantala A et al. Quality of life after surgery for rectal cancer with special reference to pelvic floor dysfunction. Colorectal Dis 2011; 13: 399-405.
29. Alavi K, Chan S, Wise P et al. Fecal Incontinence: Etiology, Diagnosis, and Management. J Gastrointest Surg 2015; 19: 1910-1921.
30. Sunesen KG, Nørgaard M, Lundby L et al. Long-term anorectal, urinary and sexual dysfunction causing distress after radiotherapy for anal cancer: a Danish multicentre cross-sectional questionnaire study. Colorectal Dis 2015; 17: O230-O239.
31. Townsend MK, Matthews CA, Whitehead WE et al. Risk factors for fecal incontinence in older women. Am J Gastroenterol 2013; 108: 113-119.

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Multidisciplinary UroGyneProcto Editorial Comment

To improve the integration among the three segments of the pelvic floor, some of the articles published in *Pelviperineology* 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.

Gynecologist. According to the work of Henry[1], confirmed by Beco[2], an abnormal perineal (anal margin) descent could be defined as more than 1.5 cm using a perineocaliper in lying position. In such a case, a descending perineum syndrome (DPS) can occur. Levator plate sagging, the descending perineum syndrome key feature, can occur because of long term dyschesia and/or puborectalis damage during delivery[3].

For many years, proctologist were the only physicians aware of this syndrome because gynecologist and urologist, even specialized in prolapse treatments, forgot to look at anal margin descent while straining. During their clinical examination, they searched for cystocele, rectocele, uterine prolapse and urinary incontinence while straining but they forgot to look at anal margin descent. It explains why most of the works about the descending perineum syndrome were done by colo-proctologist only looking at dyschesia and anal incontinence (induced by pudendal nerve stretching).

Abnormal levator plate sagging is a key feature in perineology. The absence of backpressure explains partly the difficulty to defecate[4]. Levator hiatus widening favors genital prolapse and stress urinary incontinence. Anal descent by itself explains the appearance of stretch pudendal neuropathy which disturbs anal and puborectalis innervation (vicious circle) and induces a real pudendal syndrome. Loss of anorectal angle, together with obstructive defecation, favors rectal intussusception with its side effects (vicious circle). Last but not least, the absence of support below uterus creates overstretching of the utero-sacral ligaments with posterior fornix symptoms[5].

In his paper, Pucciani showed the importance of descending perineum syndrome in perineology. As a proctologist, he underscored the link between DPS and obstructive defecation. Like Henry[1], he demonstrated changes in external sphincter muscle (with consecutive ano-rectal manometry anomalies), consistent with damage to its nerve supply and probably induced by pudendal nerve stretching. Almost all the patients had urogynecological problems (96.7%) thus showing that a global perineological approach is indispensable to treat correctly the patient.

REFERENCES

1. Henry MM, Parks AG, Swash M. The pelvic floor musculature in the descending perineum syndrome. Br J Surg 1982; 69 (8): 470-472.
2. Beco J. Interest of retro-anal levator plate myorrhaphy in selected cases of descending perineum syndrome with positive anti-sagging test. BMC Surg 2008; 8 (1): 13.
3. Roche B, Robert-Yap J, Skala K, Zufferey G: La désinsertion du muscle pubo-rectal est-elle la cause des rectocèles? In. Genêve (Switzerland): Société Suisse de Chirurgie. 2011.
4. Beco J, Antolak S: Posterior perineal support during defecation, descending perineum syndrome, pudendal neuropathy and anal fissures. Tech Coloproctol 2010; 14 (2): 193-194; author reply 195.
5. Goeschen K: Role of uterosacral ligaments in the causation and cure of chronic pelvic pain syndrome. Pelviperineology 2015; 34: 2-20.

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Colorectal surgeon. In essence this paper can be regarded as a hypothesis. It is compiling a set of observations made under test conditions in order to construct an understanding of how abnormal pelvic anatomy and physiology may lead to the presenting symptoms. An analysis of this manuscript is better performed by starting with the conclusion and working backwards.

The conclusion states that, "*faecal incontinence inevitably will materialise with the passage of time in patients affected by descending perineum syndrome, characterised by hypotonia of the pelvic floor and associated pudendal neuropathy…*" It continues to state that many factors may influence the evolution towards faecal incontinence, however, these particular factors were all excluded from the study.

For a hypothesis to become established as a Theory it requires testing. Unfortunately, there was no testing of pudendal nerve function in the methodology and this conclusion cannot therefore be stated as part of the hypothesis. Furthermore, only external sphincter and internal sphincter muscle resting tone and contraction was tested. There was no attempt to look at the function or tone in the more major pelvic floor muscles, including puborectalis, pubococcygeus and ileococcygeus/levator plate. These muscles play a fundamentally important role in pelvic floor function in both emptying and continence and cannot be excluded from testing, if it is hypothesised that subsequent failure of muscle function is a causative factor behind the development of symptoms. Although the hypothesis stands, in this paper it remains untested and therefore un-validated.

Nonetheless, the paper contains important data. Most important is the almost global coexistence of urogynaecological symptoms and other structural abnormalities lending further weight to the importance of the Pescatori Iceberg in the assessment of these cases[1,2].

Over the last few years the surgical literature has contained increasing numbers of publications that demonstrate that pelvic ligament augmentation in the presence of prolapse and the associated symptom complexes produces cures in excess of 80% (symptoms and prolapse)[3-6]. Virtually all of these papers involve cases that are individual presentations based on a well-documented spectrum of pelvic floor dysfunction and structural abnormality, the vast majority of which conform to the observations, testing and publications in accordance with the Integral Theory. It should be noted that since its initial publication almost 30 years ago not a single observation has been published that moves toward invalidating the Integral Theory. The data presented in this manuscript does more to confer further validation of the Integral Theory than it does to support the alternative hypothesis that it presents[7].

REFERENCES

1. Pescatori, M., M. Spyrou, and A. Pulvirenti d'Urso, A prospective evaluation of occult disorders in obstructed defecation using the 'iceberg diagram'. Colorectal Dis, 2007, 9 (5), 452-6.
2. Goeschen, K. and Gold, D.M., Surgical cure of chronic pelvic pain, associated bladder & bowel symptoms by posterior sling in 198 patients validates the Pescatori Iceberg principle of pelvic symptom co-occurrence. Pelviperineology, 2017, p. 84.
3. Liedl, B. et al., Is overactive bladder in the female surgically curable by ligament repair? Cent European J Urol, 2017, 70 (1), 53-59.
4. Inoue, H. et al., Repair of damaged ligaments with tissue fixation system minisling is sufficient to cure major prolapse in all three compartments: 5-year data. J Obstet Gynaecol Res, 2017, 43 (10), 1570-1577.
5. Sekiguchi, Y., et al., Is Chronic Pelvic Pain in the female surgically curable by uterosacral/cardinal ligament repair? Pelviperineology, 2017, 74.
6. F. Wagenlehner, I.-A.M.-F., G. Perletti, B. Abendstein, K. Goeschen, H. Inoue, Y. Sekiguchi, A. Sivaslioglu, M. Haverfield, P. Richardson, Vaginal apical prolapse repair using two different slingtechniques improves chronic pelvic pain, urgency and nocturia – a multicentre study of 1420 patients. Pelviperineology, 2016, 35 (4), 99-104.
7. Liedl, B. et al., Update of the Integral Theory and System for Management of Pelvic Floor Dysfunction in Females. European Urology Supplements, 2017.

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Letter to the Editor

Dear Sir,

the correlation between pelvic floor muscles and abdominal muscles is increasingly considered in the research of uro-gynecological rehabilitation. The aim of Martinho and Coll. study in the December 2017 issue of Pelviperineology[1], is to demonstrate the mutual and synergic influences between the transverse abdominal and the pelvic floor muscles. They have evaluated the changes in bladder neck position, genital hiatus and puborectalis thickness during the voluntary contraction of the transverse abdominal muscles in 31 selected patients with stress urinary incontinence. The results were compared with the parameters obtained during the maximal contraction of the only pelvic floor muscles through Kegel's exercises in the same patients. The research concludes that during a maximal contraction of the transverse abdominal muscles, only puborectalis muscle thickness was found increased, while pelvic floor maximal contraction changes all parameters.

An important starting point has been stated with this research to determine whether the goals of pelvic floor rehabilitation can be reached by abdominal muscles strengthening. An increasing number of physiotherapists, in fact, suggest low pressure abdominal exercises as a rehabilitation path in pelvic floor disorders like stress urinary incontinence and mild grade prolapse. Abdominal tone is one of the goals to be achieved in this type of pathology, but the way the therapist choose to train abdominal muscles becomes of the utmost importance: intra-abdominal pressure is greatly increased during classical abdominal exercises (flexion of the trunk on the lower limbs in supine position) and this pressure can damage the connective tissue of the pelvic floor. It is demonstrated that the most frequent pelvic floor disorders are related to changes in connective tissue. A new rehabilitative approach has been developed by Caufriez[2] to manage intra-abdominal pressures through global postural exercises targeted to tone up abdominal muscles.

These studies actually open new points of view in the research to avoid that wrong rehabilitation protocols become harmful to the patient: pelvic floor, vertebral column, thoracic diaphragm and abdomen must be considered as one functional unit[3]. We are working with this aim and results seem to be promising.

REFERENCES

1. Martinho N, Botelho S, Nagib A et al. The effects of pelvic floor and transverse abdominal muscles' maximal voluntary contractions on pelvic floor ultrasound biometric parameters in women with stress urinary incontinence: preliminary results. Pelviperineology 2017, 36 (4), 125-128.
2. Caufriez M, Pinsach P, Fernandez-Dominguez JC, 2010; Abdominaux et Perinée. Mithes et Realitées. MC Editions Mallorca.
3. Frighetto E, Una metodica innovativa nell'ambito della riabilitazione del pavimento pelvico: la Ginnastica Addominale Ipopressiva. Pelviperineologia 2017, 36 (4), 59-61.

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Response to comment on

"*THE EFFECTS OF PELVIC FLOOR AND TRANSVERSE ABDOMINAL MUSCLES' MAXIMAL VOLUNTARY CONTRACTIONS ON PELVIC FLOOR ULTRASOUND BIOMETRIC PARAMETERS IN WOMEN WITH STRESS URINARY INCONTINENCE: PRELIMINARY RESULTS*"

We read with interest the comments by Elena Frighetto on our article, which investigated the effects of pelvic floor and transverse abdominal muscles' maximal voluntary contractions on pelvic floor ultrasound biometric parameters in women with stress urinary incontinence[1]. Our findings reinforce the Sapsford's theory[2], that suggests that the transverse abdominal muscle (TrA) correlates with pelvic floor muscles (PFM), since TrA contraction increases puborectalis muscle thickness, assessed by 3D/4D translabial ultrasound. However, our results also demonstrated that TrA contraction does not seem to be as effective as direct PFM contraction, since it did not have a significant effect on genital hiatus measurements as well as on bladder neck position.

Imaging studies have shown that bladder neck funneling and descend during Valsalva maneuver, as well as urethral hypermobility and low closure pressure appear to be the main predictors of stress urinary incontinence (SUI)[3-7]. So, PFM functional rehabilitation in women with SUI should be based on PFM training, in such a way that PFM voluntary contraction can help to stabilize the bladder neck and to increase the urethral closure pressure during stressful activities that promotes involuntary loss of urine[8-10]. Therefore, TrA contraction seems not to be the best mechanism to reach these objectives.

Despite this, we share the same idea that abdominopelvic muscles act as a single functional unit and, in this way, a global training must be carried out during rehabilitation. However, in order to avoid inappropriate interventions in clinical practice, we must be careful about the results obtained and we have to keep researching in order to elucidate the functional relationship between these muscles, favoring an evidence-based clinical practice. Soon, the complete results of our research will be published and, thus, we hope to contribute expanding the scientific knowledge in our area.

REFERENCES

1. Martinho N, Botelho S, Nagib A, Marques J, Silva V, Jales R et al. The effects of pelvic floor and transverse abdominal muscles' maximal voluntary contractions on pelvic floor ultrasound biometric parameters in women with stress urinary incontinence: preliminary results. Pelviperineology. 2017; 36 (4): 125-128.
2. Sapsford R, Hodges PW, Richardson CA, Cooper DH, Markwell SJ, Jull GA. Co-activation of the abdominal and pelvic floor muscles during voluntary exercises. Neurourol Urodyn. 2001; 20 (1): 31-42.
3. Dietz HP, Clarke B, Herbison P. Bladder neck mobility and urethral closure pressure as predictors of genuine stress incontinence. Int Urogynecol J Pelvic Floor Dysfunct. 2002; 13 (5): 289-93.
4. Shek KL, Dietz HP. The urethral motion profile: a novel method to evaluate urethral support and mobility. Aust N Z J Obstet Gynaecol. 2008; 48: 337-42.
5. Digesu GA, Calandrini N, Derpapas A, Gallo P, Ahmed S, Khullar V. Intraobserver and interobserver reliability of the three-dimensional ultrasound imaging of female urethral sphincter using a translabial technique. Int Urogynecol J. 2012; 23 (8): 1063-8.
6. Wlazlak E, Surkont G, Shek KL, Dietz HP. Can we predict urinary stress incontinence by using demographic, clinical, imaging and urodynamic data? Eur J Obstet Gynecol Reprod Biol. 2015; 193: 114-7.
7. Pizzoferrato AC, Fauconnier A, Fritel X, Bader G, Dompeyre P. Urethral closure pressure at stress: A predictive measure for the diagnosis and severity of urinary incontinence in women. Int Neurourol J. 2017; 21 (2): 121-127.
8. Miller JM, Perucchini D, Carchidi LT, DeLancey JOL, Ashton-Miller J. Pelvic floor muscle contraction during a cough and decreased vesical neck mobility. Obstet Gynecol 2001; 97 (2): 255-60.
9. Miller JM, Sampselle C, Ashton-Miller JA, Hong GR, DeLancey JO. Clarification and confirmation of the Knack maneuver: the effect of volitional pelvic floor muscle contraction to preempt expected stress incontinence. Int Urogynecol J Pelvic Floor Dysfunct. 2008; 19 (6): 773-82.
10. Junginger B, Baessler K, Sapsford R, Hodges PW. Effect of abdominal and pelvic floor tasks on muscle activity, abdominal pressure and bladder neck. International Urogynecological Journal. 2010; 21 (1): 69-77.

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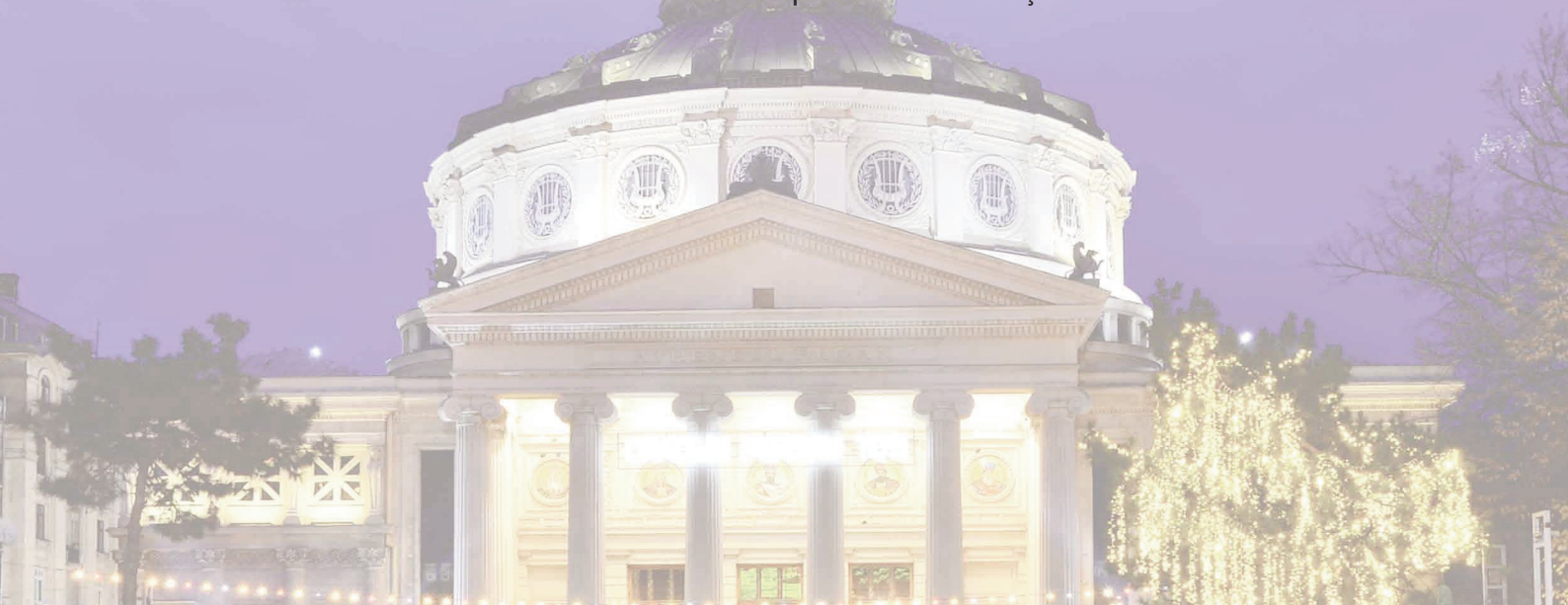
Prof. Yakup Kumtepe (Turkey) / Prof. Erkut Attar (Turkey)
Prof. Balint Farkas (Germany)

Pre-congress Workshop: Aesthetic Gynecology Workshop

Course directors: Prof. Sivaslıoğlu / Prof. Eray Caliskan
Speakers: Prof. Akin Sivaslıoğlu / Prof. Eray Caliskan
Asist. Prof. Dr. Özkan Özdamar / Asist. Prof. Aşkı Ellibeş Kaya
Prof. Dr. Sevtap Hamdemir Kılıç



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