URETHRAL DIVERTICULUM IN WOMEN: DIVERSE PRESENTATIONS RESULTING IN DIAGNOSTIC DELAY AND MISMANAGEMENT

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ABSTRACT

Purpose: We describe various clinical presentations of urethral diverticulum, which may mimic other pelvic floor disorders and result in diagnostic delay. Management and outcome results are reported.

Materials and Methods: We reviewed retrospectively 46 consecutive cases of urethral diverticulum. Patient characteristics, history, clinical evaluation, management and long-term followup are reported.

Results: Mean patient age plus or minus standard deviation was 36.3 ± 11.7 years. Most (83%) cases were referred as diagnostic dilemmas with symptoms present for 3 months to 27 years. Mean interval between onset of symptoms to diagnosis was 5.2 years. The most common symptoms were pain (48% of cases), urinary incontinence (35%), dyspareunia (24%) and frequency/urgency (22%). The number of physicians previously consulted ranged from 3 to 20 and prior therapies included oral and/or vaginal medications, anti-incontinence surgery and psychotherapy. The diverticulum was palpable on examination in 24 patients (52%), in only 6 of whom was it possible to "milk" contents per meatus. Of these 24 palpable diverticula 2 contained malignancy, and 2 others contained endometriosis and stones, respectively. Diagnosis was made by voiding cystourethrography in 30 cases (65%), double balloon urethrography in 5 (11%) and transvaginal ultrasound or magnetic resonance imaging in 7 (15%). Diverticula were incidental findings during vaginal surgery in 4 cases (9%). Treatment consisted of diverticulectomy and/or Martius flap, pubovaginal sling and urethral reconstructive procedures when indicated in 35 cases (76%), and 2 other patients underwent radical surgery for diverticular malignancy. Subsequently all but 2 patients with pain were cured. In another patient de novo stress incontinence developed postoperatively. None of the patients who underwent concomitant pubovaginal sling had postoperative incontinence.

Conclusions: The symptoms of urethral diverticulum may mimic other disorders. This condition should be considered in women with pelvic pain, urinary incontinence and irritative voiding symptoms not responding to therapy. Surgical treatment is usually effective in alleviating associated symptoms.

Key Words: diverticulum, urethra, pelvic floor, urinary incontinence

Urethral diverticulum in women is uncommon and usually presents between the decades 3 and 5, although it has been reported in neonates and young women.1 Urethral diverticulum may be congenital or acquired. Most cases in women are acquired, resulting from infection of the paraurethral glands with subsequent rupture into the urethral lumen. Other etiologies include urethral injury during childbirth or surgery and repetitive trauma secondary to catheterization.2 The pathognomonic presentation of post-void dribbling, urethral pain, tender periurethral mass and/or expression of pus from the urethra on physical examination is uncommon. Most patients present with nonspecific, refractory, lower urinary tract symptoms, unrelated to the diverticulum size or number, and undergo extensive evaluation and empirical treatments before correct diagnosis is established. Therefore, clinical awareness and a high index of clinical suspicion are essential in making the definitive diagnosis and formulating a treatment plan. We report our experience with 46 symptomatic women with proved urethral diverticulum, most of whom did not have any of the typical symptoms and/or physical findings.

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MATERIALS AND METHODS

This study entailed a retrospective analysis of 46 consecutive symptomatic women with urethral diverticulum seen during a 6-year period. Most (83%) cases were diagnostic dilemmas referred for evaluation of persistent refractory lower urinary tract symptoms. Clinical evaluation included history (specifically, duration and nature of symptoms, prior diagnoses and prior therapeutic regimens), physical examination, urinalysis and culture, 24-hour voiding diary, 24-hour pad test in incontinent patients, video urodynamics and urethrocystoscopy. Some patients underwent positive pressure urethrography using double balloon catheters or, more recently, transvaginal ultrasound and/or magnetic resonance imaging (MRI) as adjunctive diagnostic measures.

Management was tailored according to the clinical findings and the need for concomitant procedures (anti-incontinence surgery, urethral reconstruction, Martius flap interposition). Various treatment modalities have been previously advocated for symptomatic urethral diverticulum but only transvaginal diverticulectomy has been found to be highly effective.1,3 Surgical technique varied according to the anatomy, and consisted of dissection of the diverticular sac, identification of the urethral ostia when possible, excision of part or all of the diverticular sac, closure of the ostia when identified and layered closure of the defect, often using remnants of the
diverticular sac to reinforce closure. Indications for concomitant pubovaginal sling included dissection which undermined the bladder neck at the time of diverticular excision and/or preoperative diagnosis of sphincteric incontinence. Martius flap interposition was done routinely when there was a large defect after excision of the diverticular sac, a pubovaginal sling was performed or urethral reconstruction was necessary. Urethral reconstruction with local vaginal, labial or bladder wall flaps was done when tension-free primary closure of the urethra could not be accomplished.

The women were evaluated postoperatively at 1, 6 and 12 months and thereafter on an as needed basis. All were assessed for symptom relief, anatomical result and postoperative continence status. Patients opting not to have surgery were advised to return biannually for reexamination.

RESULTS

The study included 28 white (61%), 9 black (20%), 7 Hispanic (15%) and 2 Asian (4%) women. Mean patient age was $36.3 \pm 11.7$ years (range 17 to 67) and mean parity was $2.2 \pm 1.4$ (range 0 to 4).

Symptoms. The most common presenting symptoms were chronic, often intermittent pain in 22 (48%), urinary incontinence in 16 (35%), dyspareunia in 11 (24%) and frequency/urgency in 10 (22%) patients (see table). Of the patients 8 (14%) had onset of symptoms associated with pregnancy, and diagnosis was made before delivery in 2, post partum in 5 and 15 years after delivery in 1. Pain was variously described as a burning, sticking, pressure, aching or spasm, located in the vagina, pubic bone, groin, bladder, rectum or lower back, and usually not associated with voiding. Many of the patients with pain syndromes described intermittent symptoms from 6 months to 27 years, with at best temporary response to various therapies. Of the patients with urinary incontinence only 2 described post-void dribbling, a symptom which is traditionally considered a common finding in women with urethral diverticulum.

Mean interval between onset of symptoms and definitive diagnosis was 5.2 years (range 3 months to 27 years), and was significantly longer for nonpalpable compared with palpable diverticula ($7.1 \pm 5.8$ versus $3.2 \pm 4.2$ years, respectively, $p = 0.01$). The number of physicians (gynecologists, urologists, internists and family practitioners) previously consulted for these symptoms ranged from 3 to 20 (mean 9), and diagnoses included vulvodynia or vulvovestibulitis in 42%, interstitial cystitis in 19%, urethral syndrome in 19% and idiopathic chronic pelvic pain in 27% of the cases.

Diagnosis. The diverticula were distinctly palpable in 24 (52%) of the 46 patients, and in only 6 (25%) was it possible to “milk” the contents of the diverticulum via the urethral meatus, which was another sign considered to be pathognomonic of urethral diverticulum. Of the palpable diverticula cases 3 (13%) were previously misdiagnosed as cystocele, 1 of which contained 20 stones (fig. 1), and 1 presented with exacerbating vaginal pain and urinary retention, and endometriosis was inside the diverticulum.

In 2 patients malignancy was inside the diverticulum. In 1 patient who presented with acute urinary retention and a vaginal mass invasive adenocarcinoma was inside the diverticulum. She underwent anterior pelvic exenteration, continent diversion and vaginal reconstruction. The other patient had 20-year history of urinary frequency, urgency and incontinence, and had not been sexually active for years because of vaginal discomfort. She was evaluated by several urologists and referred for urethral diverticulectomy after another urologist attempted diverticulectomy but could not complete the procedure. At surgery the urethral diverticula was firm and adherent to the bladder neck. A wedge biopsy was sent for frozen section and was consistent with invasive squamous cell carcinoma. The patient underwent anterior pelvic exenteration, continent diversion and vaginal reconstruction.

Of 41 patients who underwent diagnostic cystourethroscopy the diverticular ostium was visible in only 6 (15%) (fig. 2). Definitive diagnosis of urethral diverticulum was established by voiding cystourethrography (figs. 3 and 4) in 30 cases (65%), double balloon positive pressure urethrography in 5 (11%) and by transvaginal ultrasound or MRI (fig. 5) in 7 (15%). In 4 patients (9%) diagnosis was made incidentally during vaginal operations for urinary incontinence, urethral reconstruction or exploration of a vaginal mass. Concomitant urodynamic diagnoses included sphincteric incontinence in 10 patients, detrusor instability in 5 (3 of whom had mixed stress and urge incontinence) and bladder outlet obstruction in 2.

Management and outcome results. Transvaginal excision of the diverticulum with concomitant anti-incontinence and reconstructive procedures when indicated was performed in 35 cases (76%). At surgery all diverticula except 2, were large (2 to 8 cm in diameter). Followup has ranged from 6 months to 6 years. After excision all but 2 patients with pain symptoms became symptom-free. Martius graft was used in 21 women, 1 of whom had persistent postoperative dyspareunia and labial point tenderness on the side of the graft harvest. Concomitant pubovaginal sling was performed in 14 women, all of whom were continent postoperatively. Urethral reconstruction was required in 7 patients, involving use of local vaginal wall flaps in 6 and construction of a Tanagho anterior bladder tube in 1. The patient who underwent Tanagho reconstruction had urge incontinence postoperatively. One woman, whose diverticulum was an unexpected finding during emergency surgery, required extensive dissection in the area of the bladder neck to complete the excision. Pubovaginal sling was not performed at primary surgery and postoperative sphincteric incontinence developed. Of the 2 patients who underwent radical surgery for diverticular malignancy 1 is disease-free at 5 years (squamous cell) and the other had a recurrence 6 months after surgery (adenocarcinoma).

Five patients (11%) declined surgical excision. These patients had minimally symptomatic diverticula in the vicinity of the bladder neck and after counseling opted not to undertake the risks inherent in surgery and related procedures. Four other patients were diagnosed only recently and are not candidates for surgical intervention. Two patients were diagnosed during pregnancy before delivery. There were no signs of any inflammatory process at diagnosis. Patients delivered vaginally and were observed post partum during which time both diverticula decreased significantly in size (from 5 to 6 to 1 to 2 cm.). However, despite this reduction in size symptoms persisted and surgical intervention was necessary.

<table>
<thead>
<tr>
<th>TABLE 1. Patient characteristics</th>
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<tr>
<td><strong>No. cases</strong></td>
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<tr>
<td>Mean age ± SD</td>
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<tr>
<td>No. race (%):</td>
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<tr>
<td>White</td>
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<tr>
<td>Black</td>
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<tr>
<td>Hispanic</td>
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<tr>
<td>Asian</td>
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<tr>
<td>Mean interval to diagnosis ± SD (yrs.)</td>
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<td>3.2 ± 4.2*</td>
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<tr>
<td>No. symptoms (%):</td>
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<tr>
<td>Pain</td>
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<tr>
<td>Dyspareunia</td>
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<tr>
<td>Incontinence</td>
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<tr>
<td>Frequenturgency</td>
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<tr>
<td>Vaginal mass</td>
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<tr>
<td>Dysuria</td>
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<td>Post-void dribbling</td>
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<td>Urinary retention</td>
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<tr>
<td>Recurrent urinary tract infection</td>
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<td>Voiding difficulties</td>
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* Significantly different from nonpalpable diverticula ($p = 0.01$).
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FIG. 1. A, large diverticulum fills introitus, and stones were palpable (and audible) within it. B, at excision there was no proximal cystocele. C, total of 20 stones were harbored within diverticulum.

FIG. 2. A, cystoscopic evaluation shows bladder neck to left side, and opening of large, wide mouth diverticulum on right side. B, voiding cystourethrography reveals complex bilateral urethral defects.

FIG. 3. Voiding cystourethrography reveals large pantaloon diverticulum anterior to urethra, which was nonpalpable on examination.

DISCUSSION

Urethral diverticula are believed to arise from the complex array of perirectal glands. Although they have been described in infants, the vast majority are probably acquired and may be the site of infection, stone formation or, rarely, carcinoma or endometriosis. Prior studies indicated that urethral diverticulum occurs in up to 5% of women, is most prevalent in decades 3 to 7 and is more common in black than white women. However, a more recent series showed no such racial predilection. Our study also showed a preponderance of white patients but is undoubtedly skewed by the characteristics of our referral pattern.

In a 1956 review of 66 patients with urethral diverticulum Wharton and Telinde stated, “the condition has not been generally recognized by the profession, and there is no doubt that many women are unnecessarily suffering from it today, even though they have repeatedly consulted gynecologists and urologists.” Spence and Duckett in 1970 similarly noted that “the diagnostic possibility of a diverticulum should be kept in mind in the woman with chronic or recurrent lower urinary tract complaints which baffle explanation and remain unresponsive to customary treatment.” Our series, some 30 years later, underscores the perplexity and persistence of this issue.

Most of our cases presented as diagnostic dilemmas. They had been diagnosed by previous physicians with stress urinary incontinence, urge incontinence, chronic cystitis, trigonitis, urethral syndrome, vulvovestibulitis, cystocele, sensory urgency, idiopathic chronic pelvic pain and psychosomatic disorder. Treatments, most of which were given for many years, included multiple courses of long and short-term antibiotics, antifungal, antibacterial and emollient vaginal preparations, anticholinergics, tricyclic antidepressants, anti-incontinence surgery, hydrodistention, dimethylsulfox-
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FIG. 4. Urethral diverticulum noted during video urodynamics. A, defect is seen only during voiding. B, normal post-void film.

FIG. 5. A, voiding cystourethrography reveals normal urethral contour. B, MRI shows distinct, thick walled unilocular diverticulum with ostium in area of mid urethra.

ide irrigation, urethral dilation and psychotherapy, all with persistent and/or recurrent symptoms.

In their comprehensive review Leach and Bavendam listed vaginal mass, dysuria, frequency/urgency, recurrent infections, dyspareunia, urethral pain, hypogastric pain, post-void dribbling or pain, hematuria, stress and urge incontinence, and urethral discharge as symptoms published previously during a 30-year period. In our experience many diverticula present with subtle, transient signs, and a salient feature is the episodic nature of the symptoms, with many patients reporting months to years of quiescence between flares. Obstetricians should be vigilant regarding the possibility of this condition in pregnant or postpartum women with pain, incontinence or voiding difficulty complaints.

We concur with Davis and Telinde who stated, “The most important single diagnostic instrument for the discovery of suburethral diverticula is a high index of suspicion.”12 Initial diagnostic measures for the evaluation of a patient with a suspected urethral diverticulum usually include target vaginal examination, cystourethroscopy and voiding cystourethrography. On physical examination the most pathognomonic finding is a tender cystic swelling in the anterior vaginal wall. However, only 52% of our patients had urethral diverticula that were palpable on examination. Furthermore, in only 6 of the palpable diverticula were we able to express the contents per meatus. Regardless of size or complexity, diverticula may or may not be either palpable or tender, and may arise at any point along the circumference and length of the urethra. In our series those diverticula that were palpable ranged from a slight bogginess of the urethra or parurethral tissues, to discrete cystic lesions. There were 3 cases of a large diverticulum that had been previously diagnosed as a cystocele, including 1 which was a large suburethral mass that on palpation contained multiple stones and no associated cystocele (fig. 1).

Cystourethroscopy may allow direct visualization of the diverticulum orifice and, occasionally, expression of pus or retained urine by digital compression of the mass.13 However, the orifice may be missed, even in the hands of experienced endoscopists, particularly when there is an inflammation process or obstructed orifice. The orifice was cystoscopically visible in only 6 of our patients (15%). It has been previously suggested that transvaginal digital compression of the vesical neck at cystoscopy may maximize visualization by distending the urethral mucosal folds.14

Voiding cystourethrography is considered to be a useful diagnostic tool. Ganabathi et al reported a series of 63 women with urethral diverticulum, and voiding cystourethrography adequately demonstrated the diverticulum in 95.2% of the cases.15 However, other investigators reported far less favorable detection rates.16 Voiding cystourethrography may also reveal “paradoxical stress incontinence” caused by loss of retained urine in the diverticulum during coughing.16 Diagnosis was made or confirmed with voiding cystourethrography in 65% of our cases. We routinely perform voiding cystourethrography during urodynamic evaluation. Urodynamics lends a comprehensive evaluation of bladder function, and allows us to evaluate any reproduction of symptoms in relation to bladder filling and voiding. We perform video urodynamics even when the diverticulum is obvious on physical examination to assess the extent and complexity of the defect, and to evaluate for the presence of space occupying lesions within the lumen. It is important that radiographic pictures be exposed during voiding, as some diverticula empty at the end of micturition and will be missed if only filling and post-void films are used (fig. 4). Others have advocated urethral pressure profiles as useful in the diagnosis of this disorder, with a biphasic curve being the tip-off that a diverticulum may be present.13,17

In patients in whom urethral diverticulum is strongly suspected and voiding cystourethrography is equivocal or non-confirmatory positive pressure urethrography via double balloon catheter,18 ultrasound19,20 or MRI21–23 may be useful adjuncts, particularly when the neck of the diverticulum is functionally occluded and passive filling of the defect during
voiding is not possible. Detection of urethral diverticulum on ultrasound has been improved with the introduction of high resolution transvaginal and transperineal transducers. Transvaginal ultrasound examination may detect urethral diverticulum that does not fill with contrast material, and may further provide data regarding the size, number, location, structure, content and wall thickness of the diverticulum. However, other cystic lesions, such as Gardner's cysts, vaginal inclusion cysts, ectopic ureteroceles and endometriosis, cannot be differentiated from urethral diverticulum by ultrasound examination solely. MRI, although high in cost, is an excellent imaging modality for demonstrating urethral diverticulum because of its multiplanar capabilities, excellent tissue contrast and lack of ionizing radiation. Kim et al compared urethrocystographic, urethrographic and MRI findings in 13 patients who underwent surgery because of suspected urethral diverticulum. MRI correctly showed the presence or absence of diverticula in all 13 patients (100%), whereas urethrography was correct in 9 (69%) and urethrocystography in 10 (70%). Compared with surgical findings (20 diverticula in 12 patients), MRI depicted 14 (70%), and urethrography and urethroscopy each depicted 11 (55%) of the 20 diverticula. Therefore, MRI is highly recommended when clinical findings strongly suggest a urethral diverticulum but all other imaging modalities are nonconclusive.

Of our patients 5 declined surgical excision. We cannot make any definitive statements regarding outcome of non-surgical management, as followup has been too short and the number of patients too few for meaningful analysis. All 5 patients are minimally symptomatic from the diverticula and do not wish to undergo the risks inherent in excision. Prior studies have alluded to patients with minimally symptomatic diverticula without discussing how they were managed. In their comprehensive review of 121 women with diverticula Davis and Telinde noted that "...nine (7.4%) of the patients denied any complaints whatsoever referable to the urinary system, a sizable diverticulum constituting an incidental physical finding." Likewise, Adams performed positive pressure urethromgrams in 129 women without urinary tract symptoms and 6 (4.7%) had diverticula. Davis and Robinson reviewed 120 cases diagnosed during a 10-year period, 10 (8.3%) were not treated and followup of these patients was not given. Future research should focus on the outcomes of patients with diverticula who decline surgical repair so that meaningful comparison can be given to patients regarding management options.

Various treatment modalities have been advocated for symptomatic urethral diverticulum but only transvaginal diverticulectomy was found to be highly effective. However, surgical excision can put the patient at significant risk for sphincteric incontinence and/or urethrovaginal fistula. Our preferred technique involves creation of a vaginal wall flap over the defect, excision of part or all of the sac, identification of the ostium from the urethra and closure of the urethral defect in layers under no tension, as one would a fistula. To prevent urethrovaginal fistula formation we advocate the liberal use of Martius grafts. For anti-incontinence we routinely place a pubovaginal sling of rectus fascia. Urethral reconstruction with vaginal, labial or bladder wall flap techniques is used when tension-free primary closure of the urethral defect is not possible.

CONCLUSIONS

Symptoms of urethral diverticulum may mimic a wide variety of conditions and may be intermittent in nature, which can contribute to delay in diagnosis. Even large diverticula may not be clinically obvious, especially when in the bladder neck area or anterior to the urethra. When suspected a variety of imaging and endoscopic techniques may be necessary to confirm the diagnosis, beginning with voiding cystourethrography. For surgical excision the need for concomitant anti-incontinence surgery, liberal use of Martius graft interposition and ability to use a variety of urethral reconstruction techniques should be considered. Surgical excision involves risks which may not be acceptable to minimally symptomatic or asymptomatic patients. Careful observation with time, which to our knowledge has not been delineated in any prior study on urethral diverticulum, may be a reasonable option in patients who are able to understand the risks of delaying excision, have ready access to care and are willing to come for regular followup evaluation including imaging of the defect.

REFERENCES

The need for meticulous dissection and reconstruction is absolutely critical to preventing recurrence and/or urethral dysfunction. Tension-free closure using local flaps when required may be necessary to ensure a tension-free closure using local flaps when required. Lesions available for comparison. The authors clearly describe measurements of midline periurethral fascia defects and urethral pseudodiverticula (reference 3 in article). It is not clear whether the authors found a similar correlation in this group of patients. Additionally, a description of the diverticula, including size, location and number, would have clarified the specific types of lesions that eluded earlier diagnosis. It is somewhat disconcerting that a palpable diverticulum (55%) or worse, one associated with purulent contents on digital examination (22%), should have constituted a diagnostic dilemma to a urologist or urogynecologist. It may be that these were new findings identified at patient presentation to the authors.

The authors required multiple imaging modalities to document the urethral diverticula. Definitely, if a high level of suspicion is present then additional studies are indicated. However, it is unclear whether the decreased sensitivity of voiding cystourethrography (65%) in this study and that of Jacoby and Rowbotham (44%), in comparison to the report of Ganabathi et al. (95%) (reference 1 in article) is a function of the type of diverticula or study technique. This difference in sensitivity also emphasizes why classification of the lesion is critical to comparing significant differences in the sensitivity of diagnostic techniques between reports.

Anecdotally, one would think that a greater percentage of urethral diverticula are related to anti-incontinence procedures or other periurethral surgery than 50 years ago. Some of the procedures associated with the development of pseudodiverticula, such as needle suspensions, are more recent additions to the incontinence surgery armamentarium. However, to confirm this all authors of this subject stated that the bladder had dropped, had that diverticulum when symptoms began. It is self-evident that another patient with recurrent clitoral abscesses for 8 years, who underwent 3 to 4 incisions and drainage annually for abscess recurrence before diagnosis, and in whom we found a nonpalpable, stellate, scarred periurethral diverticulum with a subclitoral loculation on positive pressure urethrography, had those symptoms because of a urethral diverticulum and for no other reason. In other words, it is obvious that these diverticula, being either difficult or impossible to palpate, or presenting with symptoms more frequently associated with other conditions, were simply not diagnosed because of the intermittent nature of the symptoms, as well as insufficient clinical awareness. In this group of patients detailed histories were obtained to ascertain all predisposing factors. Prior incontinence surgery was not a risk factor, with only 3 prior incontinence procedures among them. However, we acknowledge, as common sense dictates, that in theory any prior trauma in the vicinity of the urethra may weaken periurethral fascia and induce a true or pseudo diverticular defect.

Comparative studies indicate a higher diagnostic sensitivity with MRI compared to other contrast based radiological techniques. Recent data showing high sensitivity using voiding cystourethrography alone did not compare this technique to either MRI or sonography. It is possible that the women in that series did not have diverticula with edematous, scarred or pinpoint ostia, and passive flow diversion of the contrast stream was enough to ensure precise diagnosis. Our series included diverticula varying widely in configuration, location and local tissue quality. In some instances more than 1 diagnostic technique was needed to secure the diagnosis.

Is it reasonable to expect one imaging method to be the best test for all cases of diverticula? Is MRI the new gold standard? What if the radiology facility is not facile with MRI of deep pelvic soft tissues? Are voiding cystourethrography and urethrography now passe? Is a combination of imaging tests necessary? The answers to these questions vary with each patient and each geographical region. Although we routinely screen with voiding cystourethrography, we believe that MRI has distinct advantages. Properly applied, it can detect minute amounts of fluid collection in soft tissue. It provides a Polaroid type image, allowing even a nonradiologist to ascertain quickly the relationship of abnormal anatomy to normal structures. Voids cystourethrography, on the other hand, requires that a diverticular ostium open widely enough for rapid transit contrast material to divert the stream into the defect. Not all diverticula have such cooperative, patent ostia. Double balloon urethrography may allow one to "force" contrast material into a diverticular ostium by creating a relatively closed urethral system in which contrast passes into the defect via concentric pressure rather than opportunistic stream diversion, and yet it too is imperfect.

Despite an increasing array of imaging techniques, the most important diagnostic tool remains a high level of clinical awareness. Once suspected, judicious application of diagnostic tests, as dictated by clinical presentation and regional variations of imaging facilities, reduces the likelihood that a diverticulum will go undiagnosed.