Use of Anal Endosonography in Diagnosis of Endometriosis of the External Anal Sphincter

Report of a Case

Heinz Bacher, M.D., Wolfgang Schweiger, M.D., Herwig Cerwenka, M.D., Hans-Jörg Mischinger, M.D.

From the Department of Surgery, Karl-Franzens-University Graz, Graz, Austria

PURPOSE: Perianal endometriosis is an infrequent form of extragenital endometriosis and is usually situated in episiotomy scars. METHODS: We report a rare case involving the external anal sphincter in a 24 year-old female. The precise anatomical location of the endometriotic lesion was confirmed using preoperative and intraoperative anal endosonography. CONCLUSION: We believe this procedure to be essential when history, digital examination, and proctoscopy are not conclusive in the differential diagnosis of perianal pain or mass. Although horinonal suppression often is the therapy of choice in extrapelvic endometriosis, we think surgical excision, respecting the anatomical fiber architecture of the anal sphincter, is the best treatment for perianal endometriosis. Surgical excision is required for histological diagnosis, which is imperative in view of the albeit rare development of malignancy in extragenital endometriosis. [Key words: Extrapelvic endometriosis; External anal sphincter; Surgery; Anal endosonography]

Bacher H, Schweiger W, Cerwenka H, Mischinger H-J. Use of anal endosonography in diagnosis of endometriosis of the external anal sphincter: report of a case. Dis Colon Rectum 1999;42:680-682.

Extrapelvic endometriosis is defined as endometriotic tissue outside the uterus. Several theories[1-6] of causation have been propounded. The most likely is the autologous transplantation theory of vital endometrial cells, which includes tubarian reflux to pelvic sites during menstruation and direct wound implantation resulting from cesarean section or vaginal delivery (especially when manual uterine exploration and postpartum curettage is performed). This explains endometriosis of abdominal and episiotomy scars. Hematogenous or lymphatic spread could be the cause of rare extrapelvic occurrences. On the other hand, the metaplasia theory would explain endometriosis in females with primary infertility or males treated with estrogens by postulating metaplastic transformation of cells into endometrial tissue incertain hormonal milieux. Low concentrations of hormones in the peritoneal cavity after anovulatoric cycles (high levels after normal ovulation) may play an important role in the nesting of uterine mucosa cells. Deficiency of cellular immunity may also favor autologous transplantation. The most common anatomical locations of endometriosis are the ovaries, the pelvic peritoneum, the uterine ligaments, and the rectovaginal septum. Less common sites[2,7] are the bladder, kidney, sigmoid colon, rectum, vagina, umbilicus, and inguinal hernia sacs. Rare extrapelvic localizations are pleura, lungs,[8] breasts, extremities, abdominal scars, and the perianal region, especially in episiotomy scars.[3-6, 9-13] Symptoms in endometriosis are caused by the growth of endometrial tissue, responding to ovarian hormones during the menstrual cycle, followed by breakdown and bleeding at the end of the cycle. Nonetheless, clinical diagnosis remains difficult. Clinical findings depend on the anatomical location and may include pelvic pain, infertility, dyspareunia, dysuria, hematuria, intestinal obstruction, hemoptysis, cutaneous nodules, and painful perianal mass. Patients often have been examined in several clinics and have been given conflicting diagnoses, often of mental instability, before the correct diagnosis is finally made. This should always be confirmed histologically. The maximum incidence is during the ages of 30 to 40 years in women.[12]

Address reprint requests to Dr. Bacher: Karl-Franzens-University Graz, Department of Surgery, Auenbruggerplatz 29, A-8036 Graz, Austria.

REPORT OF A CASE

A 24-year-old female, referred by her physician to our proctological unit, reported fluctuating pain in the right anterior perianal region during the past two years. The patient did not recognize any relationship of the perianal pain to her menses. The intensity of the pain was described as slight to almost unbearable. There were never any pathological secretions or in-

flammatory signs. Her past medical history included one vaginal delivery with episiotomy seven years ago. She underwent several courses of antibiotic treatment for "incipient abscess". Perianal examination showed a narrow episiotomy scar in the right mediolateral position; no inflammatory signs, nodules, or fistula orifices were found. In the right anterior position, pain was enhanced by rectal digital examination, but no mass could be discerned. Proctoscopy revealed a normal anal canal and no evidence of internal fistula orifice. Owing to these contradictory findings, endosonography of the anal canal was performed (5 MHz rectal probe, Hitachi Medical Corporation, Tokyo, Japan). In the right anterior position we found a sharply defined lesion, 15.9 mm in diameter, with echo-free contents and dorsal enhancement. The lesion was seen to be unequivocally at a distance of 4 mm from the hypoechogenic line of the internal sphincter (Fig. 1), in contrast to frequently observed intersphincteric abscesses, which usually reach the internal sphincter. Thus, the lesion was demonstrably located in the external sphincter. Laboratory findings (leukocytes and C-reactive protein) excluded inflammation. Surgery was performed under general anesthesia, and anal endosonography was repeated intraoperatively. After curved incision of the skin, the superficial part of the external sphincter was exposed, and the fibers were divided anatomically, until a firmly rounded bluish nodule could be excised. The transected specimen revealed a fibrotic cyst, filled with chocolate-colored fluid (Fig. 2). The external sphincter was loosely reconstructed by interrupted sutures (Vicryl™ 2-0, Ethicon, Norderstedt, Germany), and the skin was primarily closed. The patient was discharged on the fourth postoperative day with normal sphincter function and in good general condition. Gynecologic examination gave no evidence of pelvic endometriosis. Histologic examination showed radically resected endometrial glands with typical strorna, blood, and hemosiderin-laden macrophages (Fig. 3). Postoperative follow-up examinations were performed at sixmonth intervals, and the patient was doing well without recurrence after 26 months.

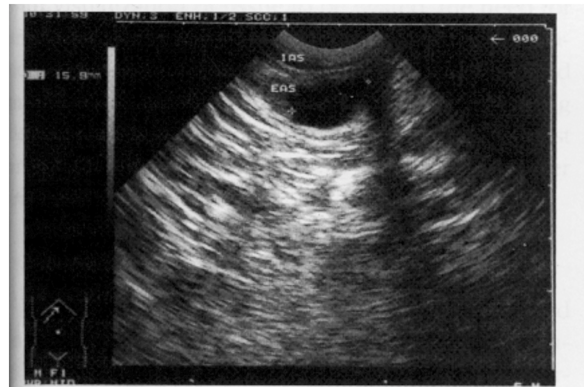

Figure 1. Transverse anal endosonographic scan showing an echo-free lesion within the mixed echogenic structure of the external anal sphincter (EAS) and the hypoechogenic line of the internal anal sphincter (IAS).

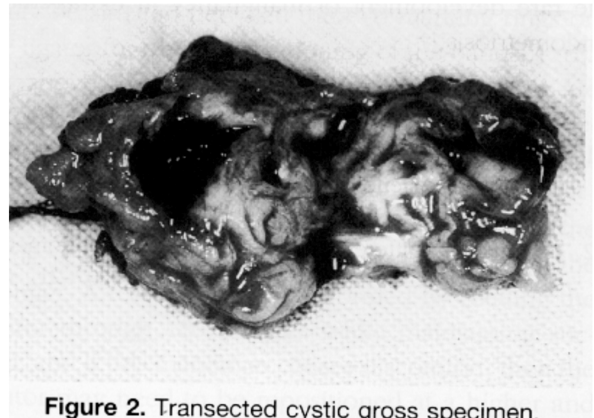

Figure 2. Transected cystic gross specimen.

DISCUSSION

Perianal endometriosis is rare, especially involving the anal sphincters.[12, 13] Autologous transplantation of endometrial cells in open episiotomy wounds or perineal ruptures during vaginal delivery, manual uterine exploration, or postpartum curettage seem to be the pathogenic mechanisms. Nevertheless, rare cases of infertile women without any perineal trauma have been described.[5] Endometriosis should be considered when cyclic perianal pain is present in women, especially in episiotomy scars. The onset of symptoms may be years after delivery.[3,5] If a precise

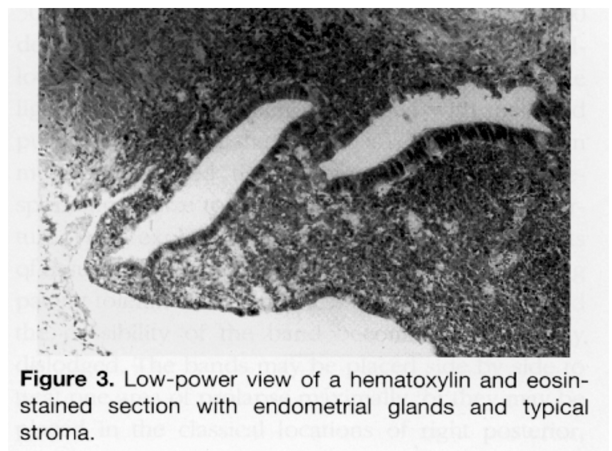

Figure 3. Low-power view of a hematoxylin and eosin-stained section with endometrial glands and typical stroma.

clinical history, physical examination, and proctoscopy cannot provide a diagnosis of perianal pain, endosonography of the anal canal is essential, in our opinion.[9] The exact anatomic relationship of any lesion in the internal and external sphincter can be determined, substantially influencing diagnosis and operative management. In our patient, conventional exploration of the intersphincteric and extrasphincteric plane yielded negative results, whereas only endosonography revealed the intrasphincteric lesion. Needle aspiration cytology and bacteriology may be of value in some cases but will hardly change the need for surgery. Limited availability and the costs of computerized tomography or magnetic resonance imaging prohibit their use in routine examinations.

CONCLUSION

Although hormonal suppression[8] often is the therapy of choice for extrapelvic endometriosis, we think surgical excision, respecting the anatomical fiber architecture of the anal sphincter, is the best treatment for perianal endometriosis.[6,9,12,13] Only surgery can provide the histologic specimens required to rule out the rare development of malignancy in extragenital endometriosis.[14]

REFERENCES

1. Dmowski WP, Radwanska E. Current concepts on pathology, histogenesis and etiology of endometriosis. Acta Obstet Gynecol Scand Suppl 1984;123:29-33.
2. Bergqvist A. Different types of extragenital endometriosis: a review. Gynecol Endocrinol 1993;7:207-21.
3. Salamalekis E, Vasiliadis TX, Kairi P, Zourlas PA. Perineal endometriosis. Int J Gynaecol Obstet 1990;31: 75-80.
4. Ljubojevic N, Trosic A, Vamai M. Endometriosis in an episiotomy scar. Jugosl Ginekol Opstet 1982;22:129-30.
5. Pollack R, Gordon PH, Ferency A, Tulandi T. Perineal endometriosis. A case report. J Reprod Med 1990;35: 109-12.
6. Cheng DL, Heller DS, Oh C. Endometriosis of the perineum: a report of two new cases and a review of literature. Eur J Obstet Gynecol Reprod Biol 1991;42:81-4.
7. Marczell A, Rosen H. Extragenital endometriosisabdominal manifestations and complications. Wien Klin Wochenschr 1987;99:273-6.
8. Espaulella J, Armengol J, Bella F, Lain JM, Calaf J. Pulmonary endometriosis: conservative treatment with GnRH agonists. Obstet Gynecol 1991;78:535-7.
9. Liang CC, Tsai CC, Chen TC, Soong YK. Management of perineal endometriosis. Int J Gynaecol Obstet 1996;53: 261-5.
10. Ramsey WH. Endometrioma involving the perineal tissues: report of a case. Dis Colon Rectum 1971;14:366-7.
11. Paull T, Tedeschi LG. Perineal endometriosis at the site of episiotomy scar. Obstet Gynecol 1972;40:28-34.
12. Sayfan J, Benosh L, Segal M, Orda R. Endometriosis in episiotomy scar with anal sphincter involvement. Dis Colon Rectum 1991;34:713-16.
13. Gordon PH, Schottler JL, Balcos EG, Goldberg SM. Perianal endometrioma: report of five cases. Dis Colon Rectum 1976;19:260-5.
14. Chumas JC, Thanning L, Mann WJ. Malignant mixed mullerian tumor arising in extragenital endometriosis: report of a case and review of the literature. Gynecol Oncol 1986;23:227-33.