Endometriosis in an episiotomy scar: Review of the literature and report of case

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The perineum is a relatively uncommon site for endometriosis, with only about 50 cases reported in the literature. The possibility that endometrial fragments are implanted there during parturition or other perineal trauma has been suggested, but if this were the case perineal endometriosis might be expected to be common among multiparas. This is not true, however. A case is added here to those reported elsewhere.

The extrauterine presence of endometrium or endometrium-like tissue usually occurs on the peritoneal surfaces of the uterus and fallopian tubes, the uterosacral or round ligaments, the bladder, rectum, cul de sac of Douglas, bowel, pelvic peritoneum, and ovaries. Less common areas are the appendix, urethra, umbilicus, laparotomy and episiotomy scars, the extremities, pleura, lungs, nasal mucosa, and kidneys.

Since endometriosis was reported first by Rokitansky in 1860, three major theories of its histogenesis have been offered:

1. According to the implantation theory developed by Sampson in 1921, transtubal regurgitation of menstrual blood and endometrial particles is followed by their attachment and growth on the peritoneal surface of the pelvic and abdominal viscera.

2. According to the coelomic metaplasia doctrine proposed by Meyer in 1907, any tissue developmentally related to coelomic epithelium and mesenchyma bears the potentiality for endometrium formation.

3. According to the metastasis theory advanced by Halban, lymphatic dissemination and hematogenous spread of endometrium at the time of menstruation lead to endometriosis.

In view of the numerous locations where endometrial implantation may occur, no one theory can explain the histogenesis of the disease in all cases. Murray expressed the opinion that endometriosis in an episiotomy scar may be explained most easily on the basis of Sampson's theory of implantation, suggesting that endometrial fragments sloughed off at delivery adhere to the episiotomy site or perineal laceration.

The first case of perineal endometriosis was reported in 1923 by Schickel, 2 years after Sampson formulated his implantation theory. A review of the literature showed approximately fifty reported cases of endometriosis at an episiotomy scar. The following report adds a recently encountered case of this infrequently diagnosed disease, which was confirmed by histopathologic study.

Report of case
A 30-year-old married white woman complained of dysmenorrhea and perineal pain during menses and requested surgical sterilization. She previously had two spontaneous deliveries over left mediolateral episiotomies. She had undergone revision of an episiotomy scar a year earlier for the same pelvic symptoms, and the histologic examination at that time showed endometriosis of the scar site.

Examination at the time of menses showed a left nodular mediolateral indented episiotomy scar with active bleeding from the purplish nodule. This area was extremely tender to mild palpation. Otherwise the pelvic examination showed no abnormality.

Recurrence endometriosis of the episiotomy scar site was diagnosed. The patient was routinely prepared for elective sterilization and admitted to the hospital for surgical correction and vaginal hysterectomy. During general anesthesia, she was placed in the dorsolithotomy position, and a total vaginal hysterectomy, excision of the endometriosis, and revision of the episiotomy scar were performed. The procedure was uncomplicated, and the patient was discharged on the fifth postoperative day. Reevaluations at 6 weeks and 1 year after surgery showed the patient to be asymptomatic and completely cured.

Comments
When Prince and Abrams reviewed the literature in 1957, twenty-five cases of perineal endometriosis had been reported. The average age of patients was 38 years, with a range from 23 to 45. In 1972, Paull and Tedeschi added fifteen
cases to the forty previously recorded. The age range of their patients was from 19 to 34 years, with an average age of 28. Gordon and associates added five cases to the literature in 1976, with an age range from 27 to 37 years. Hambrick and colleagues reported four cases in 1979, with an age range from 23 to 34 years.

In all cases reviewed in the literature with the diagnosis of perineal endometriosis there was a history of perineal trauma, either obstetric or surgical, from 4 weeks to 16 years prior to the onset of symptoms. Pain, swelling, bluish subcutaneous nodules, and pruritus varying in intensity with the menstrual cycle were present in all. Every patient in the surveyed reports had noticed a tender mass in the episiotomy region. This increased in size 2 or 3 days prior to and during the menstrual period.

The clinical diagnosis of endometriosis is made on the basis of the relation of symptoms to the menses, the nature of the tissue secretion, and the nodular appearance of the lesions. Definite histopathologic diagnosis requires the presence of stroma, glandular formation, and hemosiderin pigment. Depending on the clinical findings after examination of the patient, the differential diagnosis must consider anal fistula with abscess formation, thrombosed hemorrhoids, perianal melanoma, sebaceous cyst, hidradenitis suppurativa, dermoid cyst, hematoma, tuberculosis or actinomyosis, and basal cell or squamous cell carcinoma of the perineum or the perianal skin.

The correct diagnosis was made preoperatively in 2 of the 5 cases reported by Gordon and associates. The preoperative diagnoses in the others were anal fistula, perianal abscess, and sebaceous cyst. The presumptive diagnosis in the case reported by McGivney and Mazuji was "para-anal abscess." Treatment consisting of complete surgical excision of the endometriosis should be curative. Recurrence or possibly reappearence due to incomplete removal at the initial operation was noted in several of the surveyed cases. Recurrence is usually within 1 year after excision. Symptoms of incompletely excised endometriosis may be controlled by local hormonal injections or systemic hormonal manipulation. The efficacy of danazol therapy for this disease has not been reported on, but the drug may be tried in the future.

The present case and those reviewed demonstrate that endometriosis in an episiotomy scar might be explained according to Sampson's theory of implantation of endometrial fragments sloughed off at parturition. The rarity of the lesion certainly confuses the histogenesis. The case reported here presented the typical clinical picture involving average age, obstetric history, symptomatic and physical findings, recurrence and the interval before reappearance, with histopathologic diagnosis and complete cure following total excision of the lesion.

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6. Sampson, J.A.: Perforating hemorrhagic (chocolate) cysts of the ovary. Their importance and especially their relation to pelvic adenos of endometrial type ("adenomyoma" of the uterus, rectovaginal septum, sigmoid, etc.) Arch Surg 3:245-323, Sep 21

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Precautions and Adverse Reactions:

CONTRAINDICATIONS: Meclomen should not be used in patients who have previously exhibited hypersensitivity to it. Because the potential exists for cross-sensitivity to aspirin or other nonsteroidal antiinflammatory drugs, Meclomen should not be given to patients in whom these drugs induce symptoms of bronchospasm, allergic rhinitis, or urticaria.

WARNINGS: In patients with a history of upper gastrointestinal tract disease, Meclomen should be administered with close supervision and only after consulting the Adverse Reactions section. Peptic ulceration and gastrointestinal bleeding, sometimes serious, including one fatal case, have been reported in patients receiving Meclomen.

Diarrhea, gastrointestinal irritation and abdominal pain may be associated with Meclomen therapy. Dosage reduction or temporary stopping of the drug may have generally controlled these symptoms. (See Precautions, Warnings and Adverse Reactions.)

PRECAUTIONS: General: Patients should be given nonsteroidal antiinflammatory agents, such as Meclomen, with caution. They should be evaluated periodically to ensure that the drug is still necessary and well tolerated. (See other Precautions, Warnings and Adverse Reactions.)

Decreases in hemoglobin and/or hematocrit levels have occurred in approximately 6% of patients, but rarely required discontinuation of Meclomen therapy. The clinical data revealed no evidence of increased chronic blood loss, bone marrow suppression, or hemoglobin to account for the decreases in hemoglobin or hematocrit levels. Patients who are receiving long-term Meclomen therapy should have hemoglobin and hematocrit values determined if clinical signs of anemia occur.

Ophthalmic examinations performed prior to and following Meclomen use have not shown drug-related changes. However, because of adverse eye findings in animal studies with other nonsteroidal antiinflammatory drugs, ophthalmologic studies should be carried out if any visual symptoms develop during Meclomen administration.

When Meclomen is used in combination with steroid therapy, any reduction in steroid dosage should be gradual to avoid the possible complications of sudden steroid withdrawal.

Information for Patients: Patients should be advised that nausea, vomiting, diarrhea and abdominal pain have been associated with the use of Meclomen. The patient should be made aware of a possibility of a drug connection and accordingly should consider discontinuing the drug and contacting his or her physician if any of these symptoms are severe.

Meclomen may be taken with meals or milk to control gastrointestinal complaints. Concomitant administration of an antacid (specifically, aluminum and magnesium hydroxides) does not interfere with the absorption of the drug.

Laboratory Tests: Patients receiving long-term Meclomen therapy should have hemoglobin and hematocrit values determined if signs or symptoms of anemia occur.

Low white blood cell counts were rarely observed in clinical trials. These low counts were transient and usually returned to normal while the patient continued on Meclomen therapy. Persistent leukopenia, granulocytopenia, or thrombocytopenia warrants further clinical evaluation and may require discontinuation of the drug.

When abnormal blood chemistry values are obtained, follow-up studies are indicated.

Elevated serum transaminase levels and of alkaline phosphatase levels occurred in approximately 4% of patients. An occasional patient had elevations of serum creatinine or BUN levels.

Drug Interactions: 1. Warfarin: Meclomen (meclomenate sodium) enhances the effect of warfarin. Therefore when Meclomen is given to a patient receiving warfarin, the dosage of warfarin should be reduced to prevent excessive prolongation of the prothrombin time.

2. Aspirin: Concurrent administration of aspirin may lower Meclomen plasma levels, possibly by competing for protein-binding sites. The urinary excretion of Meclomen is unaffected by aspirin, indicating no change in Meclomen absorption. Meclomen does not affect serum salicylate levels. Greater fecal blood loss results from concomitant administration of both drugs than from either drug alone.

3. Propoxyphene: The concurrent administration of propoxyphene hydrochloride does not affect the bioavailability of Meclomen (meclomenate sodium).

4. Antacids: Concomitant administration of aluminum and magnesium hydroxides does not interfere with absorption of Meclomen.

Carcinogenesis: An 18-month study in rats revealed no evidence of carcinogenicity.

Mutagenesis: Meclomen and aspirin and other nonsteroidal antiinflammatory drugs causes teratogenicity, minor skeletal malformations, eg supernumerary ribs, and delayed ossification in rodent reproduction trials, but no major teratogenicity. Similarly, it produces phalangeal hypoplasia and interferes with parturition and with normal development of young before weaning. Meclomen is not recommended for use during pregnancy, particularly in the first and second trimesters based on animal findings. There are, however, no adequate and well-controlled studies in pregnant women.

Usage in Nursing Mothers: It is not known whether Meclomen is excreted in human milk. Because of the effects on suckling rodents and the fact that many drugs are excreted in human milk, Meclomen is not recommended for nursing women.

Pediatric Use: Safety and effectiveness in children below the age of 14 have not been established.

ADVERSE REACTIONS: Incidence Greater than 1%.

The following adverse reactions were observed in clinical trials and included observations from more than 2,700 patients. 594 of whom were treated for one year and 248 for at least two years.

Gastrointestinal: The most commonly reported adverse reactions associated with Meclomen involve the gastrointestinal system. In controlled studies of up to six months duration, these disturbances occurred in the following decreasing order of frequency with the approximate incidences in parentheses: diarrhea (10-33%), nausea with or without vomiting (11%), colitis (10%), edema (10%), and peptic ulceration (1%).

In approximately 4% of the patients in controlled studies, diarrhea was severe enough to require discontinuation of the drug. The occurrence of diarrhea is dose related, generally subsides with dose reduction, and clears with termination of therapy. The incidence of diarrhea in patients with osteoarthritis is generally lower than that reported in patients with rheumatoid arthritis.

Other reactions less frequently reported were dyspepsia, flatulence, anorexia, constipation, stomatitis and pustular ulcer. The majority of the patients with peptic ulcer had either a history of ulcer disease or were receiving concomitant antiinflammatory drugs, including corticosteroids which are known to produce peptic ulceration.

Cardiovascular: edema

Integumentary: rash, urticaria, pruritus

Central Nervous System: headache, dizziness

Special senses: tinnitus

Incidence Less than 1%.

Probable Causality Related.

The following adverse reactions were reported less frequently than 1% during controlled clinical trials and through voluntary reports since marketing. The probability of a causal relationship exists between the drug and these adverse reactions:

Gastrointestinal: Bleeding with or without obvious ulcer formation

Renal: Renal failure

Hematologic: Neutropenia, thrombocytopenic purpura

Dermatologic: Erythema multiforme, Stevens-Johnson syndrome

Incidence Less than 1%.

Causal Relationship Unknown.

Other reactions have been reported but under conditions where a causal relationship could not be established. However, in these limited reported events, that possibility cannot be excluded. These observations are listed to alert physicians.

Cardiovascular: palpitations

Central Nervous System: malaise, fatigue, paresthesia, insomnia, depression

Special senses: blurred vision, taste disturbances

Incidence between 3% and 9%.

The following reactions occurring in 3-10% of patients are listed:

- Hypersensitivity reactions
- Gastrointestinal disorders
- Skin disorders
- Nervous system disorders
- Allergic reactions
- Hematologic disorders
- Renal disorders
- Respiratory disorders
- Miscellaneous disorders

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