

Spontaneous rupture of presacral epidermoid cyst mimicking a fistula-in-ano in a young girl

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Citation

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Abstract

Epidermoid cysts are benign tumors arising from ectopic epiblastic inclusions. We report a case of an epidermoid cyst in the pre-sacral region presenting as recurrent perianal sinus in a 16-year-old girl, misdiagnosed as fistula-in-ano.

CASE DETAILS

A 16-year-old girl presented with a perianal sinus discharging foul-smelling pus for the past year with a history of lower abdominal pain and constipation. Two years back she was diagnosed as a case of fistula-in-ano and operated by a surgeon elsewhere for a problem similar to the one at hand. The wound had then healed completely. She noticed discharge from the same region once again 2 months after the procedure. Histopathological reports were not available for review.

Local examination showed a single, non-tender, discharging sinus at the 6-o'clock position 3cm posterior to the anus. A scar was appreciated separately in the perianal region suggestive of prior intervention. Per rectal examination was unremarkable. The external genitalia appeared normal.

A sinogram to determine the type of fistula and its extent revealed a sinus draining a huge pre-sacral cavity (Fig.1). Ultrasound of the abdomen and retrograde urethrogram were performed to rule out communication with the genito-urinary system and were within normal limits. The patient was operated in the prone jack-knife position. The posterior (modified Kraske) approach with coccygectomy was chosen as this approach provides excellent access to the pre-sacral region. The sinus tract was found communicating with a cyst, about 8 cm in size (Fig. 2). The whole tract was excised in continuum with the cyst. The post-operative period was uneventful. At follow-up after one year, the patient showed excellent wound healing and no signs of recurrences.

Figure 1



Figure 1: Perianal fistula at 6 o'clock



Figure 2: Fistulogram showing a fistula tract communicating with a huge cavity in the presacral area



Figure 3: Cavity after removal of cyst

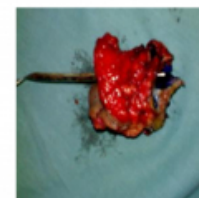


Figure 4: Excised specimen of cyst with fistula

Histopathology revealed a fluid-filled cavity with the wall lined with stratified squamous epithelium. The cyst contained dead cells suggesting pus and inflammatory infiltrate with neutrophils and lymphocytes suggesting infection.

DISCUSSION

Epidermoid cysts are benign unilocular lesions filled with clear fluid and lined by stratified squamous epithelium. They are retro-rectal in location. A pre-sacral epidermoid cyst as such is uncommon in young girls and presentation as a perianal sinus is rare. Chronic infection is the most frequent

complication occurring in 30-50% of cases and manifests as pelvic pain. These cysts rarely form local abscesses that burst within the rectum or in the perianal region resulting in a sinus with discharging pus.⁶

Fistulae-in-ano are most commonly due to ischiorectal abscesses that either rupture spontaneously into the perianal region or, more commonly, are incised. They can also be associated with Crohn's disease, ulcerative colitis, tuberculosis, actinomycosis, iatrogenic conditions and, rarely, retro-rectal cysts^{1,2}. In a study of 458 cases of fistula-in-ano, histopathology turned out to be non-specific in 90.4%, tubercular in 0.2%, post-operative or traumatic in 3.3%, anal fissure in 3.3% and ulcerative colitis in 1.5%.³ Pre-sacral epidermoid cyst as a cause of fistula-in-ano was reported in this series. Most of these conditions present in middle age and in females. Incessant fistulae are a classical feature of Crohn's disease which should be suspected when they occur in the young adult or in association with bowel symptoms. Incidence of low anal fistula in Crohn's disease is 26.7%.⁵ Another cause of non-healing fistulae is tuberculosis, a common disease in India, further emphasizing the importance of histopathology in fistulae.

Early diagnosis is necessary to prevent unwanted complications. In one study, seven patients with retro-rectal cysts had been misdiagnosed and treated as fistula-in-ano, pilonidal cysts, perianal abscess, lower back pain, post-traumatic pain, post-partum pain, and proctalgia fugax before establishing the correct diagnosis⁷. Patients in this study underwent an average of 4.1 operative procedures. All

patients underwent surgery with the para-sacrococcygeal approach and 6 out of 7 did not require coccygectomy. Physical examination in combination with CT scanning made the correct diagnosis in all the patients.

CONCLUSION

Presacral epidermoid cysts are rare entities. They are more common in middle aged women and can present as sinus in the perianal region. However, a chronic perianal sinus in the young female is commonly misdiagnosed as fistula-in-ano. A high degree of clinical suspicion is needed to diagnose the cause of the chronic or recurrent nature of such a disease, especially in the young as it may lead to significant morbidity physically and psychologically and to an unwarranted increase in procedures.

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