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Recurrence of anal canal duplication with abscess formation

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ABSTRACT

Anal canal duplication, a rare congenital disease, is often noticed in fistulas behind the native anus and is sometimes accompanied by abscesses and cysts. The perineal approach and posterior sagittal approach are considered surgical options. A 6-year-old girl with anal canal duplication with abscess formation underwent surgery using the perineal approach but experienced relapse. The foci were completely removed using a posterior sagittal approach, and her condition was cured completely. To prevent the risk of abscess formation, fistulation, and malignancy, it is important to completely remove the fistula/abscess cavity using the posterior sagittal approach during the first treatment or by switching from the perineal approach to the posterior sagittal approach when necessary.

1. Introduction

Anal canal duplication is a rare congenital disease that is often noticed in fistulas behind the native anus; often, it is a simple luminal structure. Occasionally, cases are accompanied by caustic entities in the deep part of the fistula or abscesses. Surgery is performed as minimally invasively as possible in the perineum; however, some approaches involve a sagittal sinus incision. Unfortunately, there is no policy regarding which surgical procedure to choose. A case of recurrence with abscess formation after surgery using the perineal approach and complete recovery of the lesion with a posterior sagittal incision is reported. We examined the characteristics of cysts and abscesses and the surgical method reported in the literature.

2. Case report

A 6-year-old girl reported fever and excretion from the anus. Her history revealed treatment for constipation beginning at age 3 years. At an age of 5 years, fever of 38.5 °C, swelling, redness of the buttocks, and pain in and excretion from the anus were observed. During the first examination, a fistula appeared in the 5 o'clock position of the anus; the depth of the fistula was 1 cm. No excretion from the fistula was observed. On computed tomography during initial examination, an abscess approximately 4 cm in diameter was observed in the posterior rectum that led to the right anus at the bottom and right ischiorectal fossa (Fig. 1A and B). Anal canal duplication with abscess formation was diagnosed. Although antibiotic drugs were started, there was no improvement; therefore, drainage was performed under general anesthesia on day 3 of hospitalization. After disappearance of the abscess cavity, anal canal duplication and the abscess scar were extirpated using the perineal approach. Visual confirmation was accomplished by folding the cords as much as possible and excising the cords at their limits. Incomplete resection was a possibility, but the infection was mild; therefore, we used the perineal approach. Pathological examination revealed squamous epithelium/multicore column epithelium with a length of 17 mm in luminal tissue. Postoperative wound infection was noted, but it improved with conservative treatment. She was discharged thereafter. However, the same symptoms recurred 6 months after discharge. A fistula appeared in the 5 o'clock position of the anus, and excretion from the fistula was observed. Fever of 38.5 °C, swelling, redness of the buttocks, and pain were observed. On magnetic resonance imaging, in the axial view, a cord-like cystic mass with multiple cavities squeezing the rectum from the right rear side was confirmed. In the coronal view, a cord-like cystic mass reached the coccygeus deeply on the cranial side (Fig. 1C and D). Abscess recurrence was diagnosed and reoperation was performed using the posterior sagittal approach. Elongated cord-like cystic tissue enveloping the abscess extended from the tailbone ventral side to the middle between the

Abbreviations: CT, computed tomography; MRI, magnetic resonance imaging

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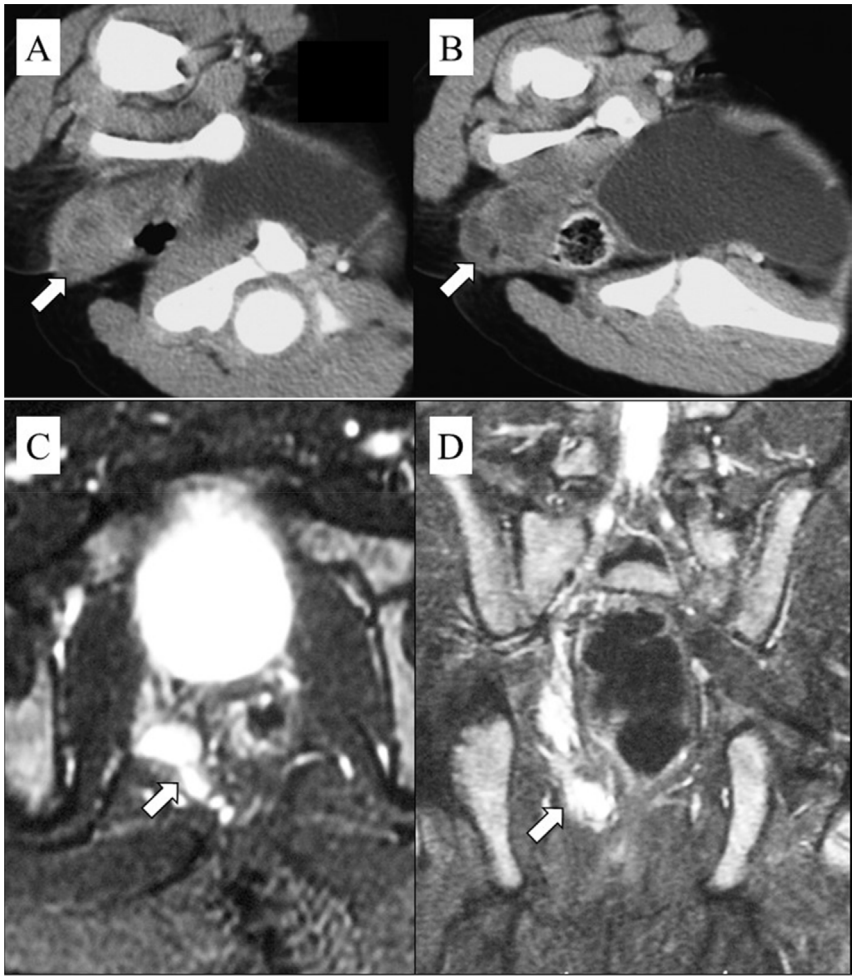

Fig. 1. Computed tomography and magnetic resonance imaging findings. A, B: An abscess with a diameter of approximately 4 cm was observed in the posterior rectum. It led to the right anus at the bottom and the right ischiorectal fossa during initial treatment. C: Axial view. A cord-like cystic mass with multiple cavities squeezing the rectum from the right rear side was confirmed. D: Coronal view. A cord-like cystic mass reached the coccygeus deeply on the cranial side during the second treatment. White arrow: abscess.

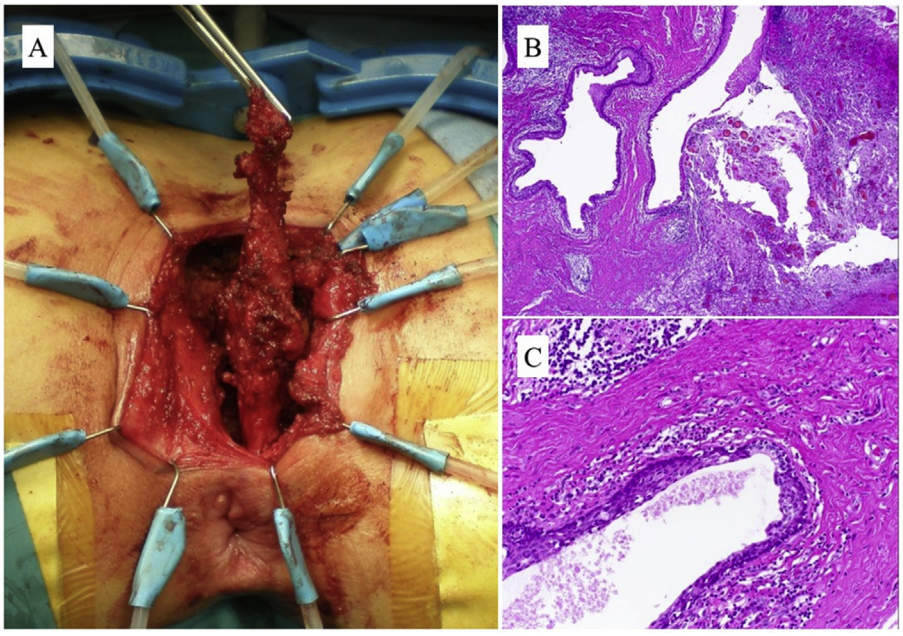

Fig. 2. Surgical and histological findings. A: Along the rectum posterior wall slightly to the right and along the rectum, elongated cord-like cystic tissue enveloping the abscess was confirmed extending from the tailbone on the ventral side to the middle between the anal margin and the dentate line. B, C: Excised specimens were granulation tissue, squamous epithelium on the anal side, and cavity formation in the squamous lining or transitional lining on the oral side.