Efficacy of magnetic resonance imaging in the diagnosis of perianal hidradenitis suppurativa, complicated by anal fistulae: A report of two cases and review of the literature

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A B S T R A C T
BACKGROUND: Perianal hidradenitis suppurativa (PHS) is a chronic recurrent inflammatory disease of the apocrine glands present in the skin and soft tissue adjacent to the anus. It is often misdiagnosed or treatment is delayed, resulting in the formation of an abscess or, in the worst case, leading to sepsis. It is difficult to treat perianal lesions merged with fistulae completely due to high recurrence rate. Therefore, we should diagnose it correctly and treat it with appropriate methods.

PRESENTATION OF CASE: We report two cases of PHS with anal fistulae that were examined preoperatively using magnetic resonance imaging (MRI) and treated safely by surgery without any recurrence.

DISCUSSION: The anal sphincter area cannot be visualized and evaluated directly by fistulography. Also CT has only limited resolution, making it difficult to distinguish between soft tissues and inflammatory streaks. Endosonography is not suitable for the examination of supra-sphincteric or extra-sphincteric extensions, as it is limited by insufficient penetration of the ultrasonic beams. MRI can demonstrate the entire course of the fistulae owing to its high contrast resolution.

CONCLUSION: Our findings support the idea that PHS with complicated anal fistulae can be diagnosed accurately using MRI and treated safely and completely with surgery.

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1. Background

Hidradenitis suppurativa (HS) represents chronic, recurrent, deep-seated folliculitis resulting in abscess formation, followed by sinus tracts and scar tissue formations. It is an uncommon disorder but frequently involves the perianal region and coexists with anal fistulae [1].

If left untreated, it may progress to a severe, persistent state with systemic infection or sepsis with the possibility of malignant alteration [2]. Complete treatment of this condition is difficult and may require complicated surgical intervention, with a recurrence rate of up to 25% [3]. One of the major reasons for this high recurrence rate is an incomplete resection of the HS and fistulae resulting from inaccurate identification or underestimation of the condition [4]. Therefore, definite preoperative recognition of the location of fistulae spread will help decrease the risk of recurrence after surgery. Since it has been well known that magnetic resonance imaging (MRI) is useful for detecting anal fistulae, MRI in HS may not only help define disease extent but assess coexisting fistulae. There have been only two published reports that highlight the efficacy of MRI on HS [5,6]. Here we report two cases of HS diagnosed using MRI for identification of the exact location of the fistulae.

2. Presentation of case

2.1. Case 1

A 43-year-old male patient was admitted to the hospital with perianal HS. The patient presented with swelling of the gluteal region and pain and pyrexia for 7 years but had been treated only by antibiotics and had never undergone any surgical
Fig. 1. (A) Hypertrophic scarring and hyperpigmentation in the anogenital lesion after 7-year history of widespread recurrent tender, erythematous nodules, abscesses, and sinus tract formation. (B) Perianal fistula tract extension, confirmed with black ties in situ, after wide excision. (C) Surgical site closed with split-thickness skin grafts. (D) Postoperative view, 6 months after successful treatment, showing no evidence of anal fistula recurrence. (Case 1).

Fig. 2. (A) Fistulography showing complicated webbing fistula in the left gluteal region. (Case 1). (B) Computed tomography showing solid hyperdense tissue in the gluteal lesion, between the intergluteal fold and the rectum (shown by arrows). However, the exact anatomic location of fistula still remains unclear. (Case 1). (C) T1-weighted MRI image, showing several separate branching fistula tracts (shown by arrows) on both sides of the posterior space, close to the anal sphincter and the surrounding inflammatory tissues. (Case 1).
Fig. 3. (A) Preoperative images of the case 2. The skin was indurated with hypertrophic scar tissue and several openings were seen from the gluteal region to the inner thigh. (B) Image after wide excision of the skin and complete removal of the tracts. (C) Surgical site closed with split-thickness skin grafts. (Case 2).

Fig. 4. Comparison of CT images (upper row) and T2-weighted MR images (lower row). MR images clearly show two main trans-sphincter fistulae with horseshoe loops, and another main fistula spreading toward the cocci (shown by arrows). These fistulae cannot be easily distinguished from soft tissues in the CT images. (Case 2).

Fig. 5. Gadolinium-enhanced T1 fat-saturated images. (Fistulae were shown by arrows). (A) A fistula extending above the left levator plate (shown by a broken line) was revealed. (B) Another fistula spreading along the right levator plate was clearly shown.
treatment. Hypertrophic scar tissue, hyperpigmentation, several visible external openings and malodorous exudate from the abscess, throughout the entire gluteal area, were observed (Fig. 1A). The internal opening was palpable beyond the dentate line by digital rectal examination. Escherichia coli was identified by culture of the exudate, and no malignancy was found by cytology. Blood examinations revealed a high white blood cell count (WBC) of 24,400/μL (range: 3,500–9,200/μL) and elevated C-reactive protein (CRP) of 17.0 mg/dL (0.0–0.3 mg/dL). The patient did not have any comorbidities. A fistulogram was performed which revealed complex interconnections of the fistulae (Fig. 2A). Computed tomography (CT) showed solid, hyperdense tissue in the gluteal region between the intergluteal fold and the rectum (Fig. 2B). Further, MRI was performed which revealed the exact location of the left gluteal abscess that communicated with the perirectal anal fistula. Four main communications of the trans-sphincter fistula with the anal canal were seen that could not be identified by CT or fistulogram (Fig. 2C).

We performed surgical excision of the involved area, above the normal tissue layer. We were able to remove all perianal fistula tracts that were identified by the preliminary MRI (Fig. 1B). The skin defect was covered with split-thickness skin grafts concurrently (Fig. 1C), and diverting colostomy was performed. Since the postoperative course was uneventful, the colostomy was closed 5 months after operation and no complication or incontinence has been seen from then on. The patient has not had any recurrence for 4 years after surgery (Fig. 1D).

2.2. Case 2

Our second case was a 46-year-old male patient who had been suffering from HS for 30 years. He had refused any surgical procedures, and had chosen a treatment with antibiotics only. The patient’s skin was indurated with hypertrophic scar tissue and several openings were seen from the gluteal region to the inner thigh (Fig 3A). Blood tests showed anaemia with haemoglobin level of 8.7 g/dL (range: 13.8–16.6 g/dL), high WBC count of 17,600/μL (3,500–9,200/μL) and elevated CRP of 8.62 mg/L (0.0–0.3 mg/dL). The patient did not have any comorbidities. Diagnostic imaging methods were performed same as in case 1. CT showed solid, hyperdense tissue extending from the gluteal subcutaneous region to the levator ani muscle, but the routes of fistulae were still ambiguous. MRI detected two main communications of the trans-sphincter fistula, one penetrating through the levator ani muscle, and the other extending towards the coccyx, neither of which could be seen with any other imaging modalities (Fig 4).

Surgical excision of the involved gluteal region and skin transplantation from the dorsal thigh was performed (Fig 3B and C). A diverting colostomy was also performed. We were able to identify the narrow fistula penetrating the levator ani muscle, as well as the abscesses connected to the minor tracts, just as shown by the preoperative MRI (Fig 5).

Though the operation was successful, skin graft infection by Pseudomonas aeruginosa was confirmed on day 4. The patient was treated with antibiotics, and the wound was irrigated with normal saline every day. Consequently, the skin graft was successful, with no deficit or necrosis. No recurrence was observed 10 months after surgery and now the patient is ready for closure of the colostomy.

3. Discussion

Hidradenitis suppurativa (HS) represents a recurrent chronic inflammatory skin disease involving the axillary, sub-mammary, inguinal, genital and perianal regions. It is characterized by abscess, sinus and undetermined ulceration formations. HS occurs in both sexes with equal frequency, although women show a greater tendency for axillary involvement, whereas in men there is a greater inclination for perianal involvement. Mean age at diagnosis was reported to be 32.9 years (range: 9.9–78.5 years) [1]. Jackman reported that out of 388 patients with HS, the perianal region was involved in 125 patients (32%) [7].

The exact aetiology of this condition remains unclear. However, it is thought to be associated with poor personal hygiene, obesity, androgen levels, cigarette smoking and presence of anal fistulae [8].

It has been reported that 64% (16/26 cases) of the patients with PHS exhibit involvement of anal fistulae [9]. One of the major reasons for relapse is incomplete removal of the fistulae. Deep-seated, remnant fistulae around the anal canal or insufficiently resected fistulae spreading into the perianal HS lead to re-growth [4]. This indicates the necessity of assessing the exact location of all fistulae of perianal lesions in patients with PHS. Several techniques have been proposed for preoperative assessment of complicated anal fistulae, such as fistulography, CT and endosonography. However, the anal sphincter area cannot be visualized and evaluated directly by fistulography [10,11]. Also CT has only limited resolution, making it difficult to distinguish between soft tissues and inflammatory streaks [12]. Moreover, we cannot anatomically identify the levator ani muscle using CT. Endosonography is not suitable for the examination of supra-sphincteric or extra-sphincteric extensions, as it is limited by insufficient penetration of the ultrasonic beams. Moreover, a recent study has shown that endosonography is not superior to digital examination for the evaluation of anal fistulae [13]. MRI can demonstrate the entire course of the fistulae, including the portion in the wall of the anal canal, owing to its high contrast resolution. We can also identify the anatomical location of the levator plate and deep fistulae. Therefore, it can be a useful method for preoperative assessment of complicated anal fistulae. It has been reported that preoperative MRI can reveal important additional information in 21% of the patients, as compared with surgery alone [14]. Gadolinium enhanced T1 images can also highlight the walls of the tracts and abscesses [6].

Aggressive and definitive surgical treatment must be performed in conjunction with medical management. Incision and drainage of individual lesions may lead to temporary control of symptoms, but is best avoided as chronically relapsed PHS may lead to severe complications such as sepsis, anaemia, arthropathy and malignant alternation. Donsky and Mendelson reported that cancer arises in 3.2% of patients with HS [15]. Lavogez et al. reported that the average time period from the onset of HS to the diagnosis of cancer was 25.3 years (range 7–40 years) [2].

We think that sufficient and drastic surgical excision is the only way to completely cure patients in the chronic, recurrent stage of the disease so that wide and deep resection of the inflammatory area were taken. Skin flap repair is frequently performed and suitable after those wide resection to reduce the risk of recurrence [16]. Performing diverting colostomy in patients with severe PHS affecting the entire perianal area was proposed by Ching and Stahlhagen [17]. We performed temporary colostomy to avoid faecal complications, and this led to successful engraftment.

4. Conclusion

Our findings support the idea that PHS with complicated anal fistulae can be diagnosed accurately using MRI and can be treated safely and completely with surgery. To avoid recurrence or any complications after operation, strategies for preoperative assessment of anal fistulae will be necessary.
Conflict of interest

No conflict of interest related to this manuscript.

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Authors’ contributions

HT, YT and SK mainly designed the study and drafted the manuscript. TK participated in the diagnostic imaging analysis. KK, DY, TM and ES participated in the operation. KY, KO, TN, TT, JT, KH and HN conceived of the study, participated in its design and coordination, and helped to draft the manuscript. HY, SI, ES and WT were responsible for this paper. All authors read and approved the final manuscript.

Consent

Written informed consent was obtained from the patient for publication of this Case report and any accompanying images.

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