



Case report

Extraspincteric anal fistula with intrarectal opening extended upto thigh, successfully treated with a minimally invasive, novel surgical technique- a rare case report

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ABSTRACT

Introduction: Fistula in ano is a complex disease, and the treatment for it is still a big challenge for surgeons because of the high recurrence rate (7 %–50 %) and incontinence, and to minimize these complications numerous surgical interventions are emerging daily in the conventional system of medicine.

Presentation of case: A 48- year- old male patient came with complaints of pus discharge from an external opening in the inner aspect of his right thigh, located about 22 to 25 cm away from the anal verge for the last 15 years and was diagnosed as long extraspincteric fistula with intrarectal opening based on clinical and MRI findings. We successfully treated this case with a minimally invasive novel surgical technique, RetroGrade Probing and Application of KharaSutra and Division of the Fistulous Tract (RGPAKS- DFT).

Discussion: Ksharasutra is a well-known method in the treatment of anal fistula for preserving continence and a low recurrence rate. Performing retrograde probing in every single case of anal fistula can address the involved anal gland under direct vision, which is essential for preventing recurrence and pairing retrograde probing with the division of the fistulous tract significantly reduces the treatment duration with minimal tissue loss.

Conclusion: In fistula surgery, successful treatment relies on identifying the internal opening and eradicating the involved anal glands. Based on the same principle, this rare and complex anal fistula was effectively treated with this RGPAKS-DFT, resulting in no recurrence and incontinence in two years of follow-up after complete recovery.

1. Introduction

An abnormal hollow tract that connects an internal opening in the anal canal to an external opening in the perianal skin is known as an anal fistula. Cryptoglandular infection, which originates in the intersphincteric space and spreads in multiple directions, is thought to be the primary cause of anal fistulas in the adult population [1].

Treatment of complex anal fistulas is still a challenge to surgeons as these fistulas involve a significant involvement of the sphincter complex and eradicating the entire fistulous tract is associated with a high risk of incontinence. Additionally, failure to remove or excise the primary and all the secondary tracts may result in the persistence or recurrence of fistulas [2], and this rate of recurrence ranges from 7 % to 50 % in various types of anal fistulae [3–5]. To minimize the rate of recurrence and preserve continence, various surgical procedures are emerging daily in conventional medicine.

Sushruta described anal fistula as Bhagandara under eight grave disorders i.e., Ashtamahagdas in Ayurveda [6] and he (500 BCE) was the first surgeon who explained the surgical excision and ksharasutra in anal fistula management [7]. In ancient Indian texts, Ksharasutra is mentioned as a treatment modality in anal fistula [8]. It is a “medicated seton” made from different medicinal plant extracts and latex impregnated in multiple layers onto a linen thread. Different Ksharas/ alkali will be used for the preparation of different ksharasutras. These ksharas [9] have anti-inflammatory and chemical debriding properties [10]. It works as a cutting, draining seton and simultaneously promotes healing and is a well-accepted treatment procedure for continence preservation and low recurrence rate. A study on the effectiveness of Ksharasutra reported a low (3.33 %) recurrence rate over a 4-year follow-up period [11].

To prevent an abnormally high recurrence rate, accurate identification of the internal opening is an essential step of fistula surgery [12,13].

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If an accurate internal opening fails to be identified and addressed, recurrence is certain. This is just because the source of sepsis in these cases won't be eliminated [14]. Hence, identification of the correct original internal opening and eradication of the involved crypts is the ultimate solution to eradicate fistula in ano. This can be achieved with the current, minimally invasive, innovative procedure, RetroGrade Probing and Application of KharaSutra and Division of the Fistulous Tract (RGPAKS- DFT). Here we treated this complex, rare anal fistula in our hospital by using the above-said procedure. This work has been reported in line with the SCARE criteria [15].

2. Presentation of case

A 48-year-old male patient presented to our outpatient unit with a complaint of pus discharge from an external opening in the inner aspect of his right thigh over the past 15 years. He had no history of chronic medical conditions such as diabetes mellitus or hypertension. He is a chronic smoker since 30 years and has normal bowel movements. He is from a middle level of socioeconomic status. The patient reported being asymptomatic 15 years ago and he suddenly developed an abscess on the upper inner aspect of his thigh, which spontaneously ruptured and healed on its own without treatment. Over two years (2008–2009, 2009–2010), he had recurrent attacks of abscess at the same site for which he was operated four times with incision and drainage. The patient was diagnosed as a case of anal fistula and was informed about the potential risk of incontinence and recurrence postoperatively. Due to concerns about these complications, the patient opted not to undergo surgical treatment, resulting in continuous suffering and recurrent abscess episodes over the fifteen years. He sought treatment at our hospital on August 24th, 2021.

On clinical examination, an external opening on the upper posteromedial aspect of the right thigh, approximately 20 to 25 cm away from the anal verge, was observed with purulent discharge and previous visible operative scars (Fig. 1A). No perianal external opening was present. Digital rectal examination revealed three internal openings, with two at the level of the dentate line at 6 o' and 7 o'clock positions, and the third approximately 2 cm above the anorectal ring at 9 o'clock position. Based on the clinical findings (Fig. 1B), the case was diagnosed as extrasphincteric fistula secondary to transphincteric fistula; Type 4, Park's classification [16].

MRI Pelvis revealed (Fig. 2A–D) a long tortuous fistulous tract with an external opening in the posteromedial aspect of the upper right thigh. The tract was observed to course anteriorly, superiorly, and right laterally up to the right gluteus maximus muscle and then slightly posteriorly, medially along the right gluteus maximus muscle with a possible intramuscular component. From there, it courses anteriorly, superiorly and slightly medially, piercing the right puborectalis muscle

at approximately 8 o'clock position, about 45 mm above the anal verge. There is noticeable surrounding fibrosis or scarring in this area. There is a small irregularity with a focal outpouching involving the anorectum approximately at 9 o'clock position, possibly indicating the internal opening. Additionally, there are other ill-defined fibrosed or sinus tracts seen in the right ischioanal fossa. There was no evidence of osteitis in pelvic bones. MRI pelvis, indicated a traceable intrarectal opening at 9 o'clock position, but the intra-anal openings were not traceable. The patient was effectively communicated about the prognosis and the anticipated treatment duration of approximately 8 to 9 months taking into account the presence of an intrarectal opening.

After obtaining written informed consent, the patient was taken for surgery under spinal anaesthesia on 1st September 2021. Surgery was performed by the corresponding author and is experienced with the technique for the last 11 years. After placing the patient in lithotomy position, an intraoperative inspection and digital rectal examination were conducted. The surgical findings are aligned with the preoperative assessment (Fig. 3A–C). Methylene blue dye was flushed from the external opening to confirm the accuracy of the visible internal openings and dye was expelled from all three internal openings (Fig. 3D). The shape of the internal (6 and 7 o'clock) openings was noted, with both directed towards 7 o'clock. Subsequently, a probe and curved fine artery forceps were cautiously introduced into these internal openings to ascertain the nature of the fistulous tracts, whether separate or interconnected (Fig. 4A). After confirming that both tracts were separate, a rigid, brass probe was inserted through the 6 o'clock internal opening at the level of the dentate line. The probe effortlessly navigated towards 7 o'clock and travelled approximately 2–3 cm away from the anal verge. At this point, division of the fistulous tract (DFT) was performed using Metzenbaum scissors and created a new external opening, through which the probe was brought out and a plain linen thread no. 20 was applied to this proximal tract (RGPAKS) (Figs. 4B–D, 5A, B). The same procedure was repeated through the 7 o'clock internal opening, and a thread was applied by bringing out the probe from the same newly created external opening. In this case, since both tracts were separate, separate threads were applied. No surgical intervention was performed for the intrarectal opening. To ensure proper drainage of the distal tract, probing was carried out from the existing external opening. However, due to the tortuosity of the fistulous tract, the probe was non-negotiable up to the newly made external opening or DFT wound. Therefore, an opening was created till where the probe was negotiable (Fig. 5C, D) and an infant feeding tube no. 8 was applied (Fig. 6A) for adequate drainage.

Preoperative prophylactic single dose and postoperative Intravenous antibiotics including inj. Ceftriaxone 1 g, intravenous, 12th hourly; inj. Gentamycin 80 mg, intravenous, 12th hourly; inj. Diclofenac sodium 50 mg, intramuscular, 8th hourly; and inj. Ranitidine, intravenous, 12th hourly was administered for 48 h. From 3rd day onwards, suggested to



Fig. 1. Preoperative findings.

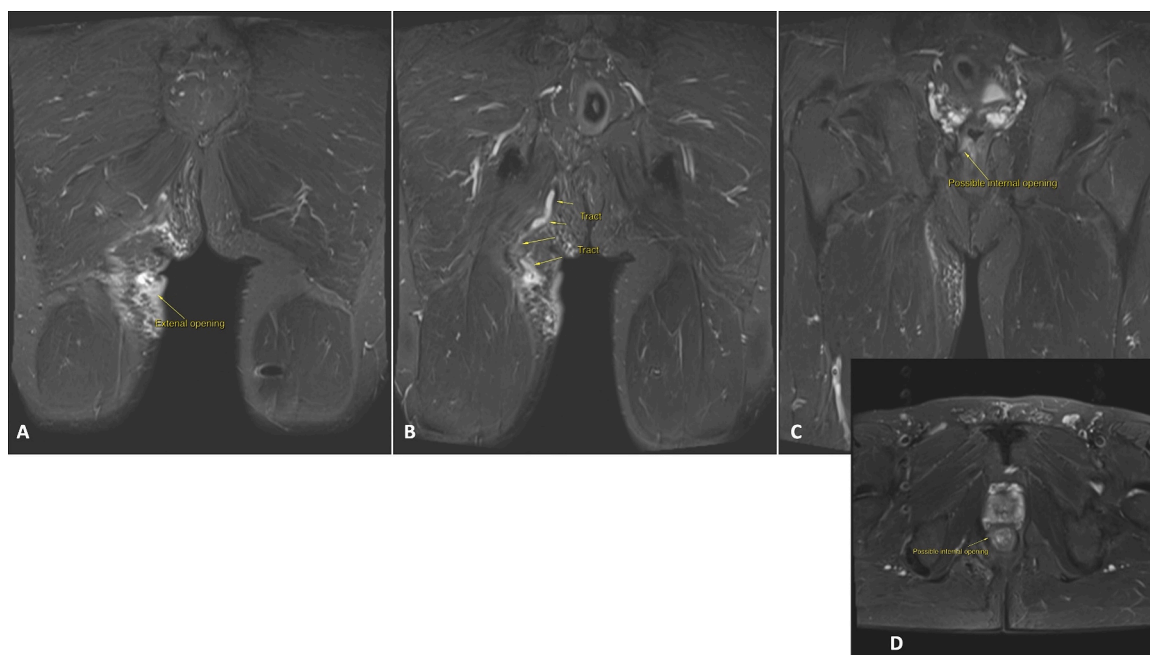


Fig. 2. MRI pelvis findings (25/08/2021).

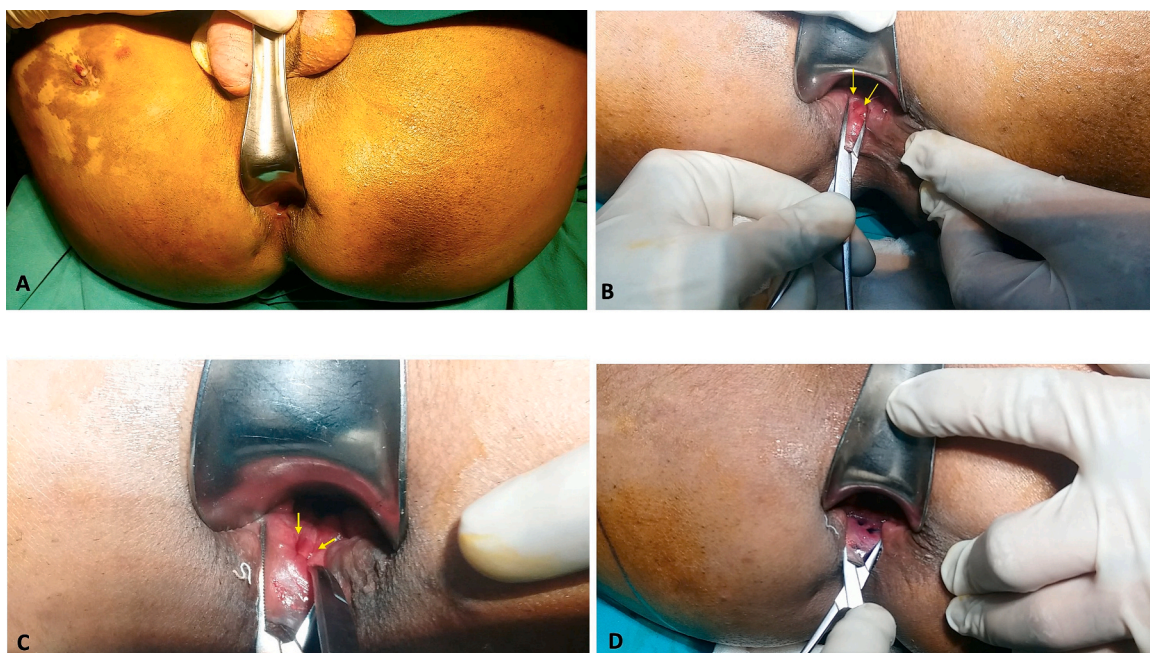


Fig. 3. Intraoperative findings (01-09-2021).

perform daily regular chores and initiated ayurvedic conservative treatment such as oral administration of anti-inflammatory drug, tablet triphala guggulu, 1 g, 12th hourly, after meals; isabgol husk 2tsf with lukewarm water at bedtime; for a month. Patient was instructed, to have a warm water sitz bath by adding 3–4 pinches of purified borax (Tankan) powder, once daily after defecation; wound dressing with Jatyadi tail smeared wick or gauze daily; and weekly once visit.

The patient was kept on plain linen thread no. 20 (Fig. 6B) for four months to aid the healing of an intrarectal opening and other ramifications. After confirming the healing of the intrarectal opening by digital rectal examination (four months after surgery) treatment with apamarg ksharasutra was started for cutting and healing of the fistulous tract. The

Ksharasutra was changed and tightened once a week for next five weeks and the tract was eventually laid opened with Ksharasutra. To ensure proper drainage of the distal tract, an infant feeding tube was kept in place without being changed over for these five months. On 22nd of February, after inserting Sims speculum into the anal canal, the floor of the wound was examined for remnant tracts and a ramification was identified at 6 o'clock position, below the level of the dentate line. To address this issue patient was taken for surgery under local anaesthesia on 24th of February 2022 and RGPaks was performed in the ramification and the infant feeding tube in the distal tract was removed and replaced with plain linen thread no. 20 for drainage. Subsequently, the ksharasutra was changed once a week for three weeks and the tract was

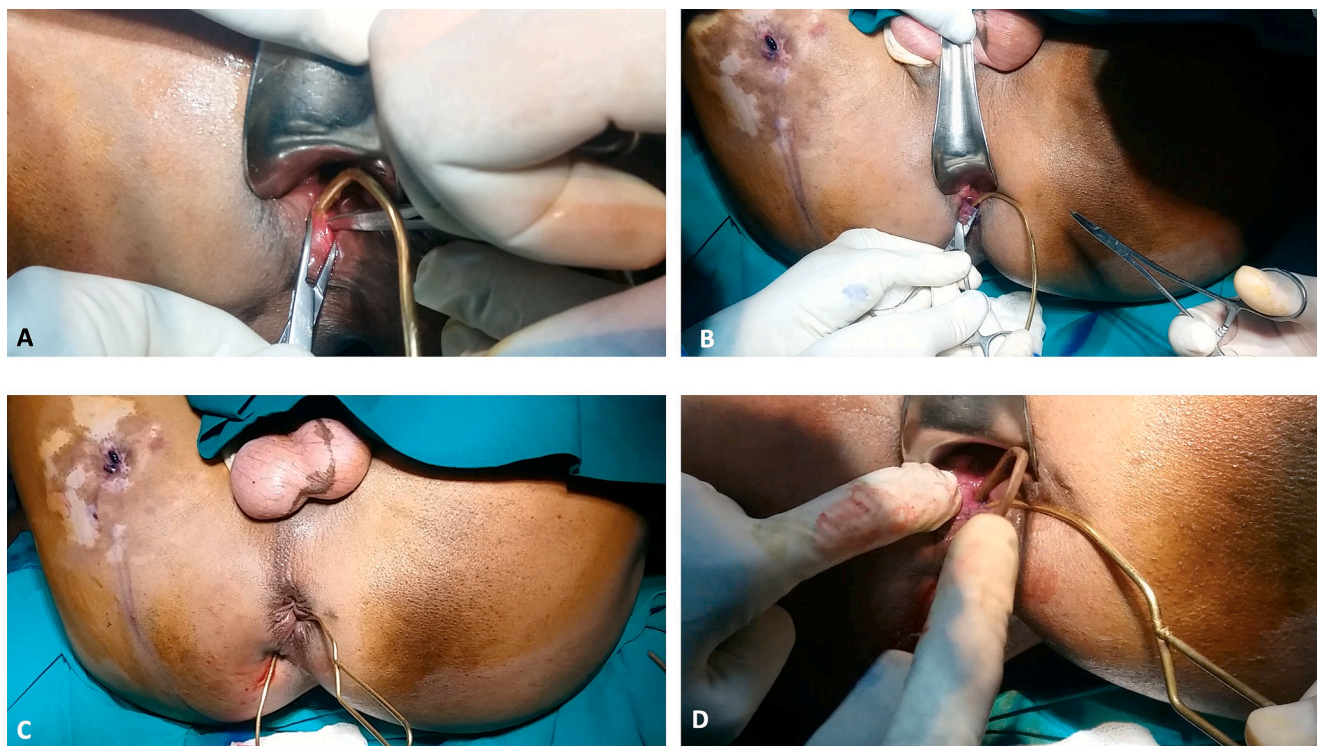


Fig. 4. Steps of operative procedure (01-09-2021).

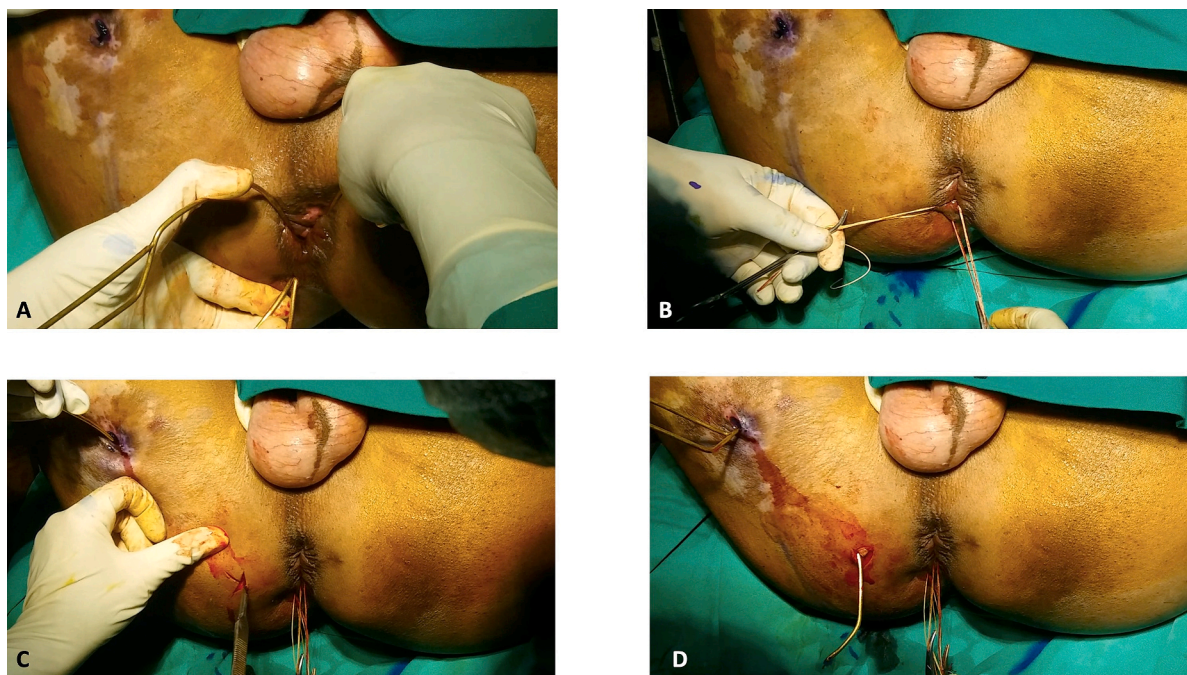


Fig. 5. Steps of operative procedure (01-09-2021).

successfully laid open with ksharasutra. On 19th of April 2022 the floor of the wound was re-examined (Fig. 6C) and after confirming the floor to be healthy, plain linen thread from the distal tract was removed. Notably, no curettage or coring was performed for the distal tract, and it got healed completely (Fig. 6D) within the subsequent ten days.

The patient has not experienced any postoperative complications such as incontinence to flatus, liquids, and stool (as per Wexner's continence score), and the operative procedure is well tolerated by the

patient with high satisfaction score. There are no signs of recurrence after two years of follow-up after recovery. The patient expressed overwhelming gratitude for the treatment, stating, "I am excited and overjoyed with my complete recovery and I feel like I got a new life after suffering for fifteen long years."

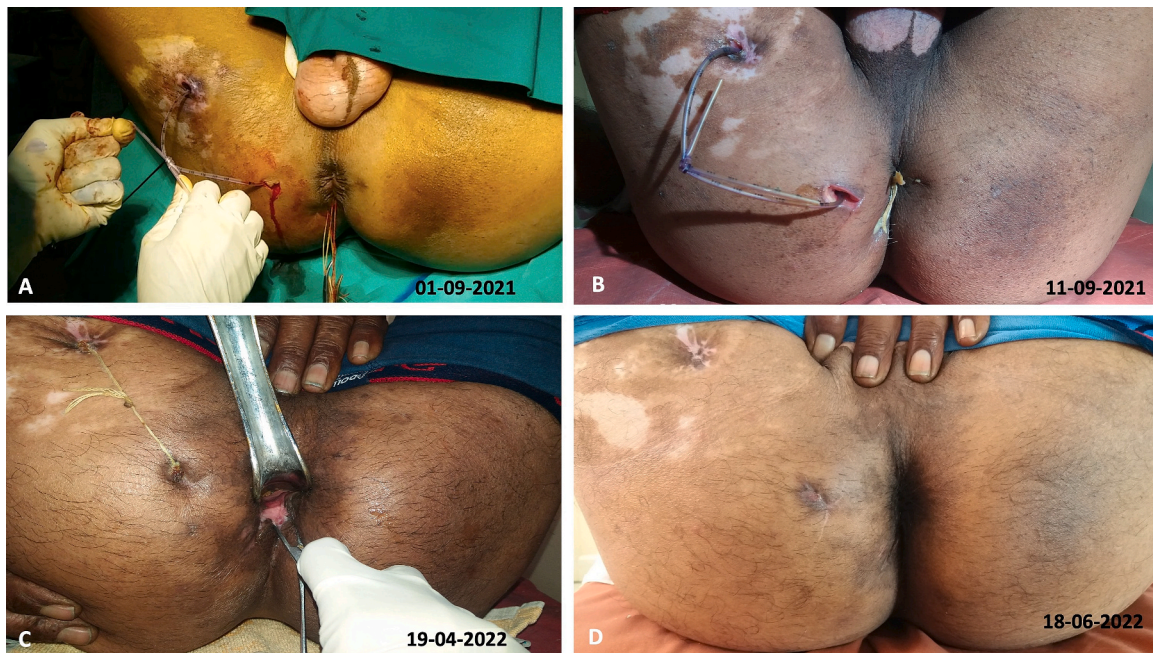


Fig. 6. Images captured during treatment and follow - up.

3. Discussion

The current surgical procedures available for the extrasphincteric anal fistulas with an intrarectal opening often involves a temporary diversion colostomy or the placement of a seton from an intrarectal opening, which has a high risk of incontinence or resection of the fistulous tract followed by surgical closure of the intrarectal opening with unsatisfactory results.

The use of ksharasutra in the treatment of anal fistula is an established and safe technique for preserving continence. Ksharasutra is a medicated thread made from linen no. 20 (Barbour) and is coated with various herbal medicines and herbal extracted alkali (Kshara). These ksharas have anti-inflammatory and anti-debriding properties with which draining, cutting and healing of the tracts will be achieved simultaneously. After the application of ksharasutra in the fistulous tract, it remains in direct contact with the tract and curettes out the inner epithelial lining chemically and physically, allowing the fistulous tract to cut and heal simultaneously [17,18]. Ksharasutra treatment involves the tightening and changing of the medicated thread every seven days. This process facilitates the cutting of the muscle complex, while the medicinal properties of the thread support the healing of the wound at the same time [19]. Moreover, Ksharasutra aids in draining and promoting the healing of the ramifications too. In this particular case, apamarga ksharasutra was used for the treatment, where 21 coatings of different herbal medicines were done on thread, such as 11 coatings of latex of *Euphorbia nerifolia* Linn. (snuhi ksheer), 7 coatings of *Achyranthes aspera* Linn. plant extracted alkali and 3 coatings of turmeric powder [20].

Retrograde probing is a technique where probing will be performed from inside the anal canal through the internal opening and surgeons usually choose this procedure when probing is failed from the external opening or where an external opening is absent. But by performing retrograde probing in every single case of anal fistula, surgeons can carefully examine the shape, direction, site, number, and nature of internal openings under direct vision, which is essential for preventing recurrence.

In the RGPaks procedure, the initial step involves thorough identification of the internal opening's shape, direction, site, nature, and number. Subsequently, a rigid probe is carefully passed through the

opening. Once the probe enters the original fistulous tract it navigates effortlessly into the fistulous tract as per its course. In small fistulous tracts, this procedure alone can be performed whereas in long fistulous tracts (more than 4 cm), to minimize the tissue loss and to reduce the overall duration of the treatment, RGPaks should be paired with DFT.

In DFT, division of the fistulous tract (DFT) will be done approximately 2 to 4 cm away from the anal verge (according to the course of the tract) considering this point as a new external opening/ DFT wound. The fistulous tract towards the anal verge is the proximal tract and treat it with Ksharasutra, the tracts away from the DFT wound are the distal tracts. If the DFT wound is kept well patent for adequate drainage, no intervention such as applying thread/ feeding tube in the distal tracts is necessary. Once the source of sepsis is removed, the distal tracts will heal in the next 10 to 15 days, without performing coring or curettage.

The following are some of the crucial points that contributed to the complete recovery and the prevention of recurrence in this case:

1. Addressing the correct internal openings according to their shape and nature
2. Providing adequate drainage healed the intrarectal opening, all the ramifications, and secondary tracts.
3. Identifying the nature of multiple internal openings as separate and treating them with ksharasutra
4. Examining the floor for remnant tracts
5. Addressing the ramification with ksharasutra

The advantages of this procedure are, that it is safe and cost-effective, causes only minimal tissue loss, allows the patient to resume daily activities from 2nd or 3rd postoperative day, preserves continence, leaves a minimal scar and maintains the integrity, anatomy and contour of the perineum with a single disadvantage, i.e., burning pain, which lasts for ten minutes to two days after changing ksharasutra every week, till the anoderm is laid open. During this period patients will be suggested to take ice compression and oral analgesics (if the pain is intolerable).

In every fistula surgery, identifying the original internal opening is critical in minimising the risk of a high recurrence rate. To effectively address this issue, it is important to perform retrograde probing through the internal opening based on its shape, number, and nature to eliminate

the involved anal crypt and gland. This rare, complex extrasphincteric and long anal fistula with the intrarectal opening was successfully treated with this minimally invasive, innovative surgical procedure, without causing any postoperative complications such as incontinence to liquids, gases and stool and there was no sign of recurrence in two years of follow-up.

4. Conclusion

Complex fistulae are a challenge to surgeons due to their high recurrence rate and incontinence, making them difficult to manage. Surgery to address the extrasphincteric fistulas with an intrarectal opening often involves a temporary diversion colostomy or the placement of a seton from an intrarectal opening or resection of the fistulous tract followed by surgical closure of the intrarectal opening. However, in this long, complex extrasphincteric fistula secondary to transphincteric fistula with intrarectal opening, eradicating the involved anal gland (primary focus) with Ksharasutra and providing adequate drainage had healed up the intrarectal opening, multiple ramifications and secondary tracts. Hence, RGPaks alone or paired with DFT can be considered a safe, cost-effective surgical procedure in treating anal fistulas.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written informed consent is available for review by the editor-in-chief of this journal on request.

Ethical approval

Case reports are exempted from ethical approval in our institute, National Institute of Ayurveda Hospital, Jaipur, Rajasthan, India.

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Author contribution

Dr. Swapna Bopparathi, performed surgery, care of this patient, conceptualization, design, acquisition, analysis and interpretation of data, and drafting this case report.

Dr. K. V. Narasimha Raju reviewed and revised the draft.

Both authors approved the final version of the manuscript.

Guarantor

Dr. Swapna Bopparathi.

Research registration number

N/A.

Conflict of interest statement

None.

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