CASE REPORT

Conventional transanal excision for a very low gastrointestinal stromal tumour

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SUMMARY

Gastrointestinal stromal tumour (GIST) represents a rare tumour entity, which has been more intensively investigated during the last decade. The rectum as the primary site of GIST is even uncommon. The space constraints in the pelvis renders optimal oncological surgery demanding and proximity of these lesions to the anal sphincter enhances the potential morbidity of any radical surgery. We herein report on a young patient with a >5 cm rectal GIST localised at 1 cm from the anorectal junction.

BACKGROUND

Transanal resection techniques have been increasingly applied for this 'atypical' indication in the course of the recent years, especially in combination with preoperative imatinib treatment in a neoadjuvant concept. The available few data refer mainly to modern techniques for tumours of the upper or middle third of the rectum. However, poor experience is available in the treatment of very distal rectal gastrointestinal stromal tumour (GIST), where conventional transanal techniques could be revisited.

CASE PRESENTATION

A 37-year-old Caucasian man was referred to our clinic for surgical management of a GIST situated

in the lower rectum, diagnosed after a thorough oncological consultation. Seventeen months ago the patient experienced a feeling of rectal fullness 1 year after being surgically treated for a perianal abscess. MRI of the abdomen and pelvis revealed a large, firm, oval-shaped tumour 54×49 mm of the lower rectum (figure 1A). Sigmoidoscopy confirmed the presence of a submucosal mass 1 cm from the anorectal junction causing incomplete obstruction. Endoscopic ultrasound (EUS)-guided fine-needle aspiration biopsy was compatible with a GIST. Positron emission tomography–CT confirmed the presence of the rectal tumour with corresponding metabolic activity and excluded the presence of metastatic disease (figure 1B). The patient initiated treatment with imatinib and 5 months later MRI was repeated showing a 10% decrease of the tumour size. A third MRI was performed 7 months later revealing a further 40% decrease of the GIST (30×30 mm) (figure 1C). Symptom of rectal fullness was improved but not resolved and the patient was therefore led to surgery where he underwent a transanal excision of the tumour.

TREATMENT

With the patient in modified Lloyd-Davies position, the anus was gently dilated and retracted with interrupted absorbable sutures. The inferior margin of the tumour was visualised 2 cm above the dentate line. We therefore performed a full thickness excision of the tumour down to the mesorectal fat starting from the inferior limit and proceeding circumferentially with at least 1 cm radial margin using a torsional ultrasonic scalpel. After sufficient haemostasis was obtained, the bowel wall defect was closed in a transverse manner to avoid narrowing the lumen using interrupted absorbable sutures. The specimen (figure 2A) was oriented with interrupted sutures for pathological assessment of the margins. The patient remained afebrile, experienced minimal postoperative pain and resumed regular diet and activity within 8 hours, whereas bowel transit was restored within 24 hours after surgery.

OUTCOME AND FOLLOW-UP

The patient was discharged on the third postoperative day. Pathology revealed a 45×40×35 mm GIST with free tumour margins in all directions (figure 2B). Interestingly, although preoperative EUS-obtained biopsy was positive for the CD117 c-kit proto-oncogene product, final

Figure 1 MRI of the gastrointestinal stromal tumour (GIST) at the time of diagnosis (max diameters 5.4×4.9 cm). (B) Positron emission tomography of the GIST at the time of diagnosis. (C) MRI of the GIST after completion of neoadjuvant treatment with imatinib (max diameters 3×3 cm).

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Figure 2 (A) Resected gastrointestinal stromal tumour (GIST) of the lower rectum. (B) Pathology image of the resected GIST.

immunohistochemistry was negative. He remains in excellent physical condition, without any signs of faecal incontinence, 15 months postoperatively. The patient received adjuvant treatment with imatinib during this period of time and is expected to complete therapy after a total period of 3 years postsurgery according to oncology consultation.

DISCUSSION

During the recent years, the remarkable evolution of surgical and endoscopic techniques for the treatment of anal tumours and their reported safety and feasibility broadened the indications for transanal excisions. From rectal polyps, not amenable to colonoscopic resection, to mobile lesions <3 cm, as well as benign adenomas and T1N0 adenocarcinomas, the indications were expanded to fistulas, strictures, neuroendocrine tumours, retrorectal tumours, GISTs and, in selected cases, to locally more advanced adenocarcinomas or to adenocarcinomas after neoadjuvant treatment. Transanal endoscopic microsurgery (TEM), transanal minimally invasive surgery (TAMIS), TEM plus endoscopic posterior mesorectal resection and transanal total mesorectal excision are some of the currently applied transanal methods.

The conventional transanal excision of low or middle rectal masses is feasible and could prevent a radical abdominal or abdominoperineal resection. Such an approach is advantageous in regard to shorter hospital stay, earlier recovery and lower perioperative morbidity, including a better defaecation function. Large tumour size or high location (>8 cm from anal verge) constitutes strong limitation for transanal excision. Conventional transanal surgery, although less invasive, is associated with higher recurrence rates, especially for tumours of the upper and medium rectum.¹ In these cases TEM or TAMIS appears to be a valuable alternative to radical abdominal operations. The magnified view and the possibility to use longer instruments facilitate a more accurate and radical resection of high-located rectal lesions. More specifically TAMIS has some advantages over TEM, such as the smaller diameter of the used platform and the reduced injury of internal sphincter.² Most of the recently published studies focus actually on TEM and its complications, such as incontinence and peritoneal perforation. Incontinence after TEM appears usually after resection in radiated rectum, wide full-thickness excisions (>50% of the rectal circumference) or after prolonged operative time (>2 hours). The perforation risk is highly associated with the distance of tumour from anal verge (distance >6 cm).³ Finally, the risk of bleeding appears to correlate with the tumour dimension or its lateral position.⁴

Our case underlines the feasibility of the applied conventional transanal technique for a very distal rectal GIST in combination with imatinib in the neoadjuvant setting. Surgery is the primary treatment of choice for patients with localised

or potentially resectable GIST lesions.^{5 6} Neoadjuvant therapy with imatinib may be preferred if the tumour is borderline resectable or locally advanced but non-metastatic. Selected cases of primary resectable GIST could also be pretreated with imatinib, as size reduction could decrease the postoperative morbidity. The tumour genotype should always be specified, as patients with PDGFR (platelet-derived growth factor *receptor-alpha*) D842V mutation, or a succinate dehydrogenase-deficient or neurofibromatosis-related GIST would not benefit from imatinib treatment. Nevertheless, the optimal duration of neoadjuvant treatment with imatinib remains unclear. The current practice is administration for 6–12 months before performing the surgical resection. Tumour response expressed with mass reduction indicates the favourable phase for resection. The recommended preoperative staging, restaging after neoadjuvant treatment and postoperative follow-up imaging technique for rectal GISTs is MRI. MRI confers direct multiplanar capability in determining tumour origin with precision, defines rapport to adjacent structures and delineates best the pelvic floor surgical anatomy.⁷ Furthermore, the optimal duration of adjuvant treatment for high-risk GIST is 3 years.⁸ Apart from ours, only one study reported on conventional transanal excision for rectal GIST after neoadjuvant therapy, with similar favourable outcome.⁹ Lastly, alteration of c-kit-positive tumours has been reported in the past.^{10 11} The exact cause of this phenomenon remains unclear but is speculated to be associated to the therapeutic effect of imatinib.

In conclusion, the treatment of rectal GIST depends on their size and exact location. The neoadjuvant administration of imatinib could facilitate a more conservative surgical resection.

Learning points

- Conventional transanal technique for distal rectal gastrointestinal stromal tumours (GISTs) remains nowadays a valuable curative approach.
- Size and exact location of GISTs located in the rectum have a dominant role in defining optimal treatment.
- Neoadjuvant therapy with imatinib may benefit patients with borderline resectable or locally but not distally advanced tumours.
- Preoperative administration of imatinib could help omitting a more radical surgical intervention.
- Specification of tumour genotype is essential in order to recognise patients who are more likely to benefit from imatinib administration.

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