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Fistula-unexpected complication after rectal cancer surgery due to Zinner syndrome

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ABSTRACT

Zinner syndrome is an uncommon developmental defect of men's urogenital system, described as a triad of unilateral kidney underdevelopment, seminal vesicle cysts, and ejaculatory duct blockage. In our report, a patient had a diagnosis of Zinner syndrome only after treating recurrent rectal cancer. The patient presented with a postoperative perineal abscess and fistula. During the surgery, the abnormally extended ureter was damaged. The complications could be avoided if the presence of Zinner syndrome was confirmed before the surgery. The specific radiological signs of magnetic resonance and computed tomography must be considered. They could have predicted the patient's surgical outcomes.

1. Introduction

Zinner syndrome is a rare innate abnormality in men's genitourinary system.[1] It manifests with a symptom triad of renal agenesis, seminal vesicle cyst, and obstruction of the ejaculatory duct.[1,2] In most cases, these aberrances are unilateral.[1] Magnetic resonance imaging (MRI) is an irreplaceable modality to visualize soft tissues; it helps detect minor deformities of seminal vesicle tissue and adjacent parts of men's internal genitalia and evaluates the development of the kidneys.[3] These abnormalities are uncommon in the pediatric age and may not cause any discomfort until the second to the fourth decade of the patient's life.[1] Patients may present with pain in the perianal area, dysuria, frequent micturition, painful ejaculation, or primary infertility.[3] However, sometimes these symptoms are not evident, and the patient may not be aware of his condition until he is tested for another disease in the pelvic area. In this report, we will present the case of a patient incidentally diagnosed with Zinner syndrome after the management of recurrent rectal cancer and discuss its outcomes and postoperative complications.

2. Case presentation

A 62-year-old man with a medical history of rectal cancer was referred to the urologist consultation due to discomfort in the genitourinary region. The patient underwent chemoradiation therapy and surgery due to rectal cancer two years ago. A few months ago, a local rectal cancer recurrence was diagnosed, and a pelvis magnetic resonance tomography (MRI) was performed (Fig. 1A and B).

Later, he underwent rectal resection. The postoperative course was smooth. Two months after the surgery, the man presented to the emergency department due to pain in the perianal region and swelling. A "bump" appeared in the anus area. The patient was hospitalized, and drainage of the perianal abscess was performed.

A control abdomen and pelvis computed tomography (CT) was performed shortly later. The CT demonstrated a tubular structure that communicates with the remaining fluid in the postoperative area (Fig. 2). The radiologist differentiated the lesion between abscess and injury in the right ureter during the previous surgery.

An MRI of the pelvis was conducted one month later to detect changes in the pelvis. The MRI confirmed the right seminal vesicle cysts and postoperative fistula (Fig. 3A and B).

The previously performed CT scan confirmed right kidney agenesis (Fig. 3C).

We presented the case of a 62-year-old man who suffered from a postoperative fistula. The diagnosis of Zinner syndrome was confirmed after surgery for recurrent rectal cancer when complications occurred.

3. Discussion

Zinner syndrome is a very rare developmental anomaly characterized by ipsilateral renal agenesis, malformations of seminal vesicles, and

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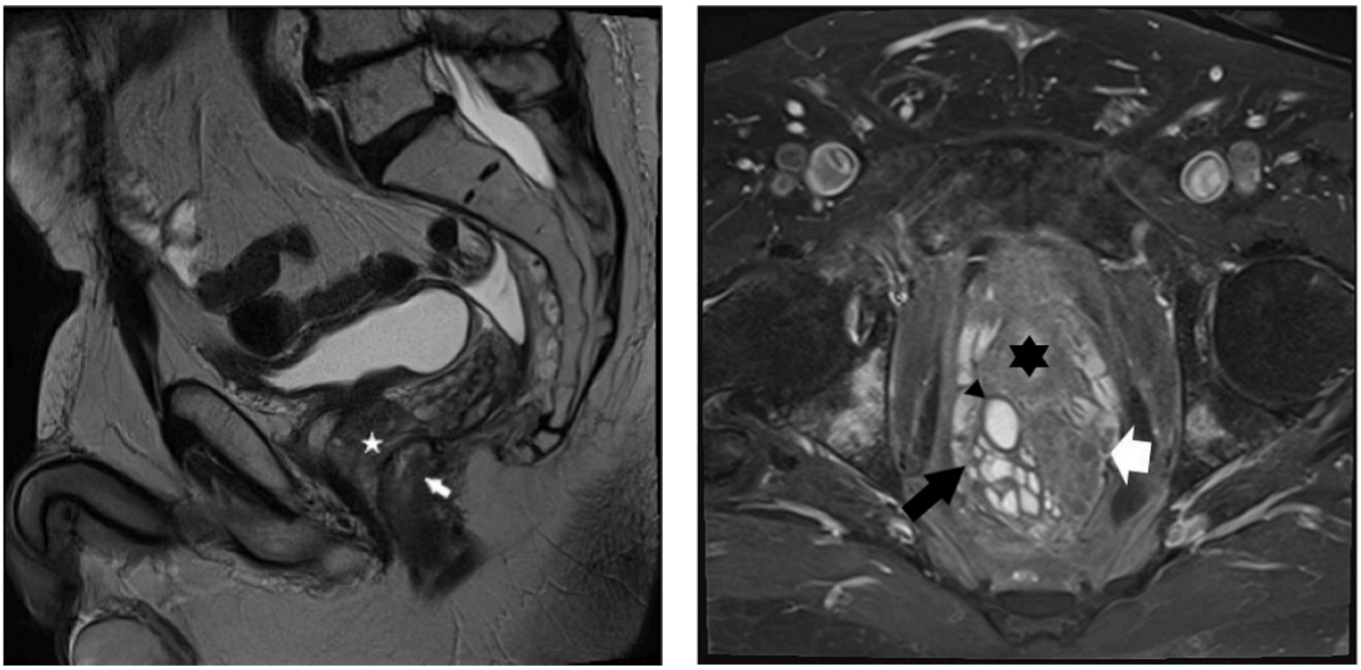

Fig. 1. **A** – T2w magnetic resonance imaging showing rectal cancer recurrence (arrow), the prostate is also marked (star); **B** – axial T1w after contrast media showing hyperintense right remnant ureter (black arrowhead), which is draining into a seminal vesicle cyst (black arrow). The left seminal vesicle is normal looking (short white arrow), the prostate is marked (star).

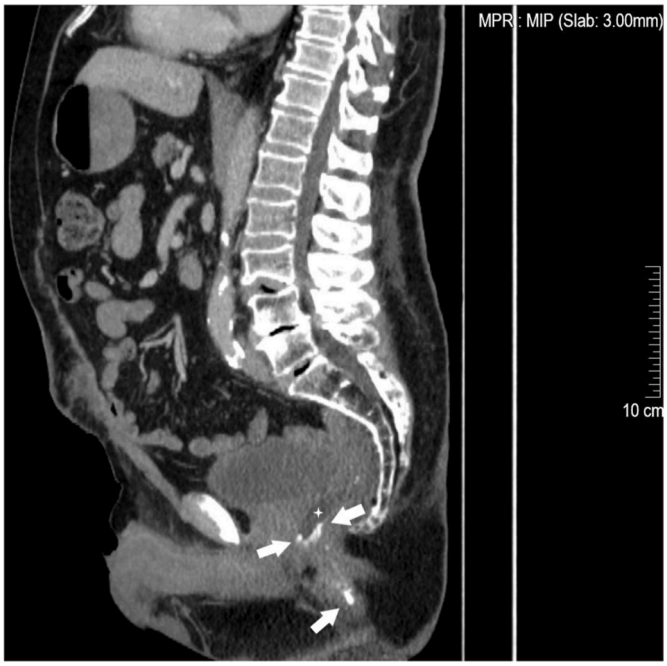

Fig. 2. Computed tomography sagittal imaging showing postoperative clips after rectal cancer surgery (arrow) and retrovesical hypodense tubular structure (star).

ejaculatory duct obstruction.[2] Diagnosis is usually based on intravenous urography, MRI, and cystoscopy. Radiological examinations play a crucial role in making a final diagnosis. Complications and comorbidities should be identified accordingly.[2]

Infectious complications have the highest incidence rate after rectal surgeries because of the complexity of keeping the area uncontaminated.[4] Fistulas between the bowel lumen and adjacent pelvic structures may develop in the late postoperative period. There is up to a 0.28% chance of ureter damage during colorectal surgeries. Rectal cancer patients are at the highest risk of intraoperative ureter damage.[5] Our patient underwent two rectal cancer surgeries. The first one was smooth, but the residual ureter in the underdeveloped kidney was injured during the second surgery, resulting in a perineal abscess and fistula formation. The patient had three risk factors contributing to his adverse postoperative outcomes: preoperative radiotherapy for rectal cancer, immunosuppression due to chemotherapy, and Zinner syndrome.

Syndrome diagnosis in our case was complicated because the focus was on diagnosing, staging, and treating rectal cancer. Zinner syndrome was silent for the patient and showed no clinical symptoms such as dysuria, perineal pain, epididymitis, and pain following ejaculation [2].

The case is didactic. The patient was diagnosed with Zinner syndrome after rectum cancer management. We should remember that a man with ipsilateral renal agenesis and tubular structure near seminal vesicles should be aware of Zinner syndrome presentation. A timely diagnosis of Zinner syndrome would have prevented the patient from developing a fistula and associated complications that worsened the patient's quality of life.

4. Conclusion

In conclusion, CT and MRI allow proper investigation of renal and seminal vesicle anomalies. MRI overtakes CT in the abdomen and pelvis due to the absence of ionizing radiation and soft tissue contrast, which are essential features in assessing the association between pelvic organs and structures. Compared to other cases of Zinner syndrome described, our case is unique in that a fistula complicated the syndrome. Additionally, the diagnosis was complicated by postoperative changes, and the patient underwent only pelvic MRI examinations for the spread of the rectum disease, which hindered the assessment of renal agenesis.

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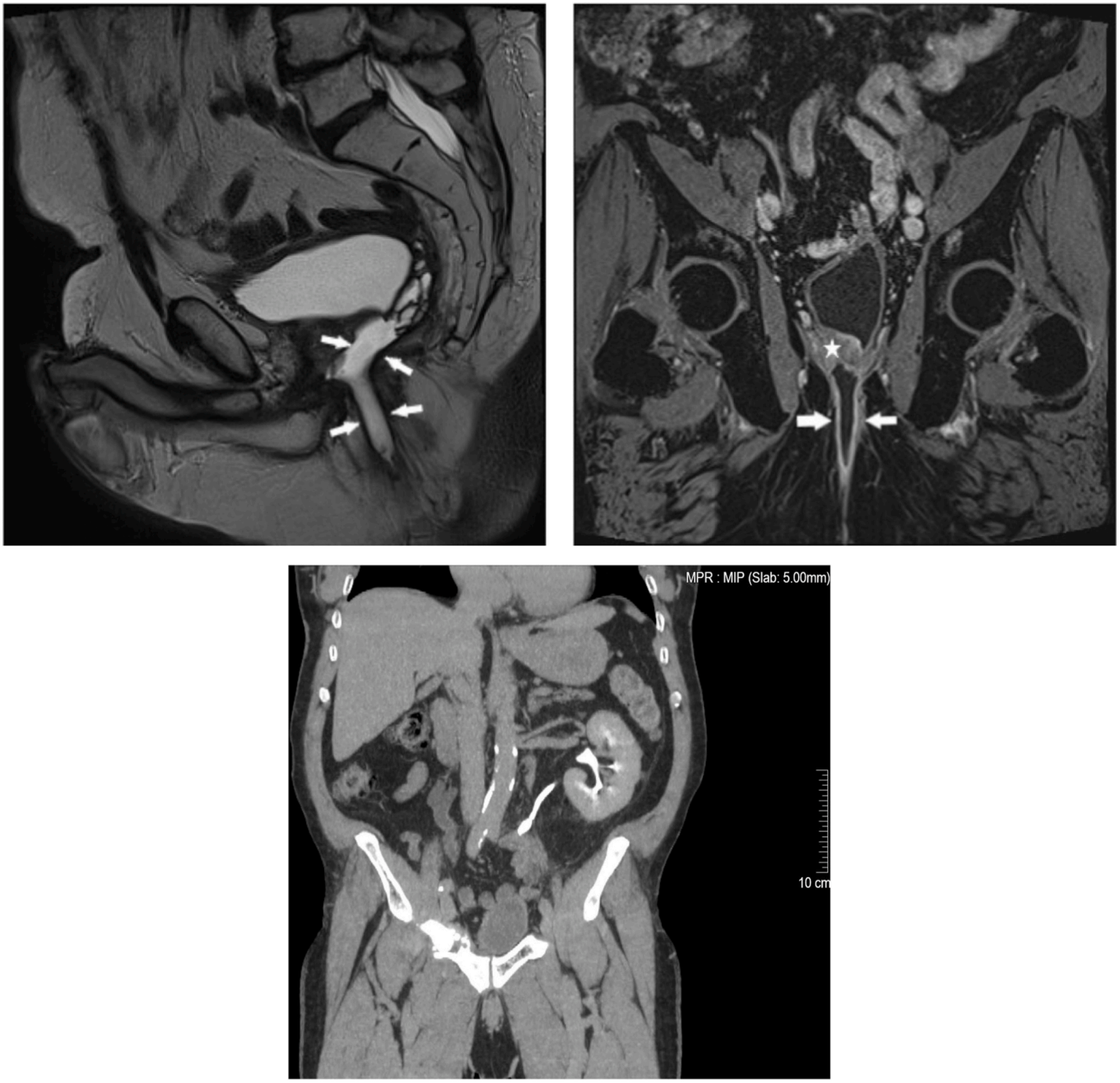

Fig. 3. Magnetic resonance imaging **A** – T2w sagittal view and **B** – T1w coronal view after contrast media are showing postoperative fistula (arrows) and prostate (star); **C** – computed tomography presented the right kidney agenesis.

Declaration of competing interest

The authors declare no conflict of interest.

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