

A case report of isolated presacral squamous cell carcinoma developed four years after gastrectomy

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ABSTRACT Chemoradiation therapy and a transsacral resection were performed to treat isolated squamous cell carcinoma that occurred in presacral tissues in the pelvis four years after gastrectomy due to early gastric cancer, with the prognosis continuing to be favorable. The patient was a 57-year-old woman, who came to our hospital having symptoms of anemia four years after gastrectomy. After a rectal examination, a tumor mass the size of a sparrow-egg was discovered on the left rectal wall. An abdominal CT showed a tumor, 3.7cm × 3.7cm in size, on the outer left wall of the upper rectum. After a CT-guided biopsy, squamous cell carcinoma was detected. Irradiation (total 40 Gy) and chemotherapy (MMC+5-FU) were performed, mass shrinkage was confirmed, and a transsacral tumor resection was performed. According to the histopathological examination, a very small but viable cancer was found to be remaining. 4 years after the tumor removal, no recurrence has been discovered. Squamous cell carcinoma in the pelvis often originates from the vagina. However, the patient didn't have any malignant findings from a genital examination at the time the symptoms appeared, and this case was diagnosed as isolated squamous cell carcinoma. A transsacral approach to remove such a tumor is considered to be useful because it is relatively low invasion and preserves anal functions.

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Key words : Presacral tumor, Squamous cell carcinoma, Transsacral resection, Epidermoid cyst

INTRODUCTION

Malignant tumors in the presacral space often originate from the rectum, uterus, adnexa, vagina, bladder, or have metastasized from a tumor in another part of the body. This is a case report

of squamous cell carcinoma developed in the presacral space four years after gastrectomy due to early gastric cancer. The cancer was treated by chemoradiation therapy and transsacral resection.

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CASE REPORT

A 57 year-old female who had undergone distal gastrectomy for gastric cancer (in the body of stomach, poorly differentiated adenocarcinoma, sm2, no lymph node metastasis) four years ago visited our hospital for anemia. She had received follow-up treatments after the gastrectomy, but by her own decision she stopped receiving the treatments three years after the gastrectomy. In March of the fourth year after her operation, she felt lightheaded and saw a nearby doctor. In April, she came to our hospital to have a more detailed examination.

No abnormalities were found in either the head and neck area or the chest. No superficial lymph node was enlarged. The abdomen was flat and soft, with no abnormal bowel sound. A tumor mass the size of a sparrow egg (1.5-2 cm) was detected by a digital rectal examination on the outside of the left wall of the rectum. Blood examination revealed no remarkable abnormalities except moderate anemia. CEA, CA19-9 and SCC were within the

normal ranges. Abdominal CT showed a tumor mass approximately 3.7 cm × 3.7 cm in size, with necrosis in it, on the outside of the left rectum above the peritoneal reflection. The rim of the tumor mass was moderately enhanced by a dynamic contrast-enhanced CT (Fig. 1). The tumor mass was touching the uterine corpus, the rectum above the peritoneal reflection and the pelvic wall. Invasion couldn't be ruled out, but additional obvious metastasis to other parts of the body including the lymph nodes was not found. CTs from the time of her stomach cancer 4 years prior were re-examined retrospectively and a nodular shadow of low density approximately 1.8 × 1.2 cm was found in the same area. Pelvic MRI revealed the lesion was partially adjacent to the uterine corpus and posterior vaginal fornix, but it was not successive and both ovaries were normal. However, the lesion was strongly adhered to the rectum above the peritoneal reflection and the left anterior sacrum, and severe adhesion and/or invasion was suspected (Fig. 2). Upper gastrointestinal

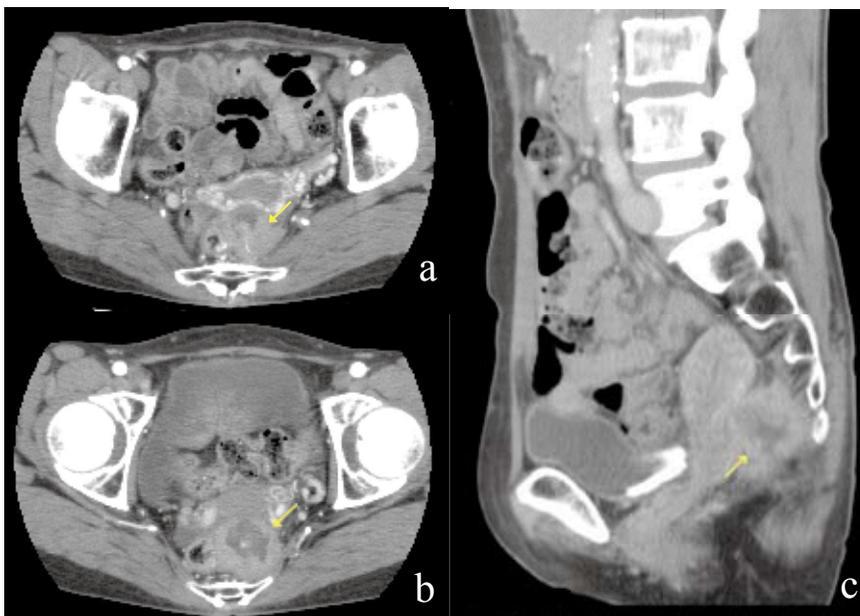


Fig.1. Dynamic contrast-enhanced CT (Yellow arrows show the moderately enhanced tumor)
a: Axial view revealed the tumor was touching the uterine corpus and rectum.
b: Axial view revealed the tumor was touching the sacrum.
c: Sagittal view revealed the tumor was touching the uterine corpus.

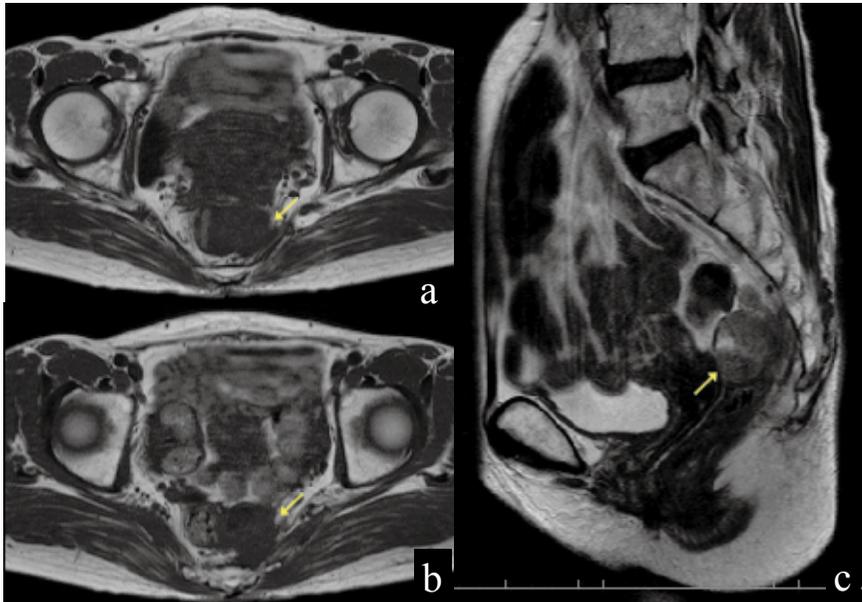


Fig.2. Pelvic MRI (Yellow arrows show the presacral tumor)

a: The tumor was partially adjacent to the uterine corpus and posterior vaginal fornix, but it was not successive and both ovaries were normal.

b: The tumor was strongly adhered to the rectum and the left anterior sacrum, and severe adhesion and/or invasion was suspected.

c: Sagittal view revealed the tumor adhered to the rectum above the peritoneal reflection.

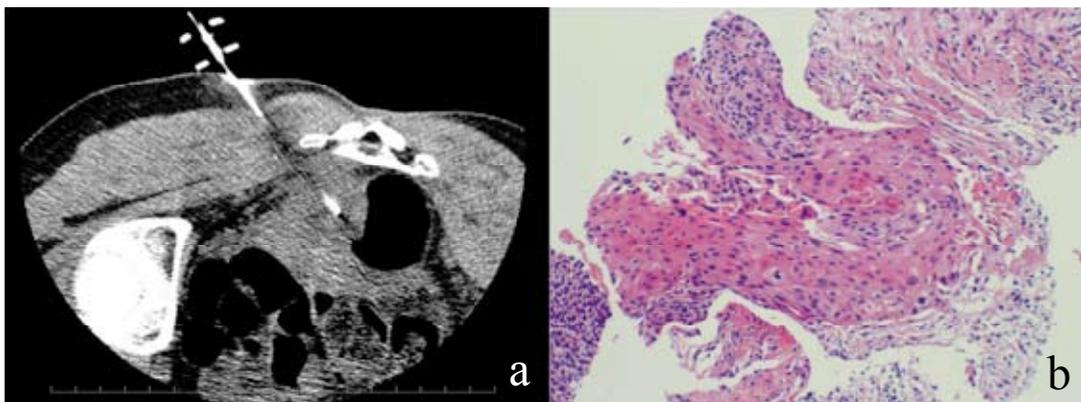


Fig.3.

a: CT-guided needle biopsy was performed using a 20 G cutting needle placed in the prone position.

b: Squamous cell carcinoma was detected after a histopathological examination.

endoscopy showed no significant abnormalities in either the remaining stomach or duodenum. Colorectal endoscopy showed a dolichocolon. A 3 cm-sized mild bulge that seemed to be caused by the pressure on the outer wall was found on the left anterior wall of the rectum above the peritoneal

reflection, 10 cm above the anal verge. Vaginal exfoliative cytodiagnosis revealed no malignant finding on the vaginal mucous membrane.

CT-guided needle biopsy was performed using a 20 G cutting needle placed in the prone position (Fig. 3a). Squamous cell carcinoma was detected

after a histopathological examination (Fig. 3b). The lesion was diagnosed as squamous cell carcinoma originating from the pelvis. Irradiation (10MV x-ray, 2 Gy parallel opposing portals per treatment, 5 times a week, total 40 Gy) and chemotherapy (MMC 100 mg/m², D1+5-FU 1,000 mg/m², D1-4) were started based on standard anal canal cancer treatment. A month and two weeks after chemoradiation was finished, the tumor was confirmed by abdominal CT to have shrunk to 1.2 cm.

2 months after chemoradiation was finished, a transsacral resection was performed. With the patient in the prone jackknife position and the buttocks were taped apart, the procedure started with an angled incision from the left of the fourth sacral vertebra, through the tip of the coccyx, to 3 cm away from the anal verge. Then the gluteus maximus muscle was resected at the point of connection of both sides of the coccyx. The coccyx was resected at its head (Fig. 4a) and the anococcygeal rache was resected as well. A tumor mass the size of a chicken egg that was fixed to the left wall of the rectum was found digitally. The uppermost portion of the tumor mass was adhered to the peritoneal reflection so the pelvic cavity and the abdominal cavity were opened at the time of removal. Dorsally, while bleeding from the venous plexus in the anterior sacrum was

stopped, and the tumor was removed from the surrounding tissues, leaving only its connection to the invaded rectal wall. We resected the mesorectum around the invaded area and exposed the rectal adventitia. Then with an electric scalpel, we resected the adventitia and muscularis propria and exposed the submucosa. Using an automatic suturing device, we resected the invaded part of the rectal wall and removed the tumor along with the rectal wall (Fig. 4b). A sample of the resection stump was taken to frozen section pathology and its malignancy was confirmed negative. Histopathological examination revealed hyalinized connective tissue among the pelvic tissue had increased, and cornified cells that lapsed into necrosis in the connective tissues were also found. There was foam cell infiltration around the cornified cells (Fig. 5a). Isolated and scattered vesicles of dysplastic cells were found and very little yet viable squamous cancer remained (Fig. 5b). Immunostaining was p16-positive (Fig. 5c). The radiotherapy treatment effectiveness as measured by histopathological criteria was Grade 2. No tumor-cell invasion in the rectum was found histologically.

The postoperative course was uneventful, and the patient was discharged 14 days after the operation. She is receiving out-patient follow-up treatments and no reoccurrence has been found after 4 years.

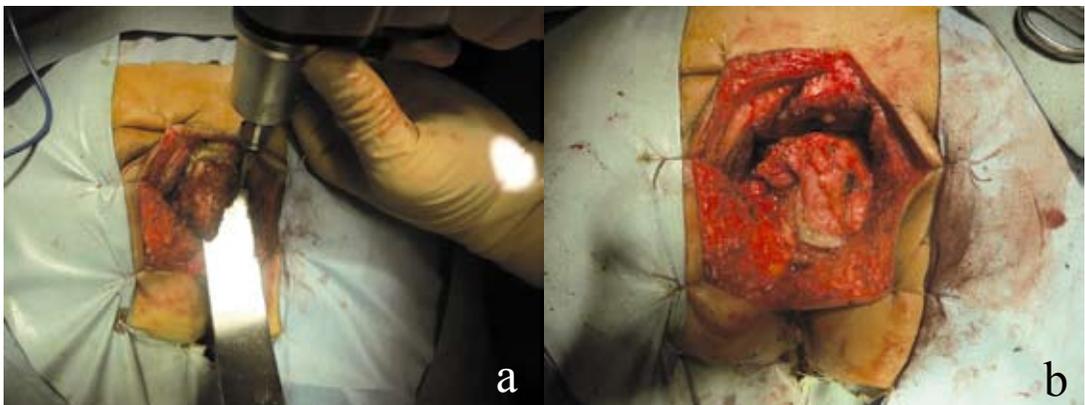


Fig.4. Operative findings

a: The coccyx was resected at its head.

b: Using an automatic suturing device, we resected the invaded part of the rectal wall and removed the tumor along with the rectal wall.

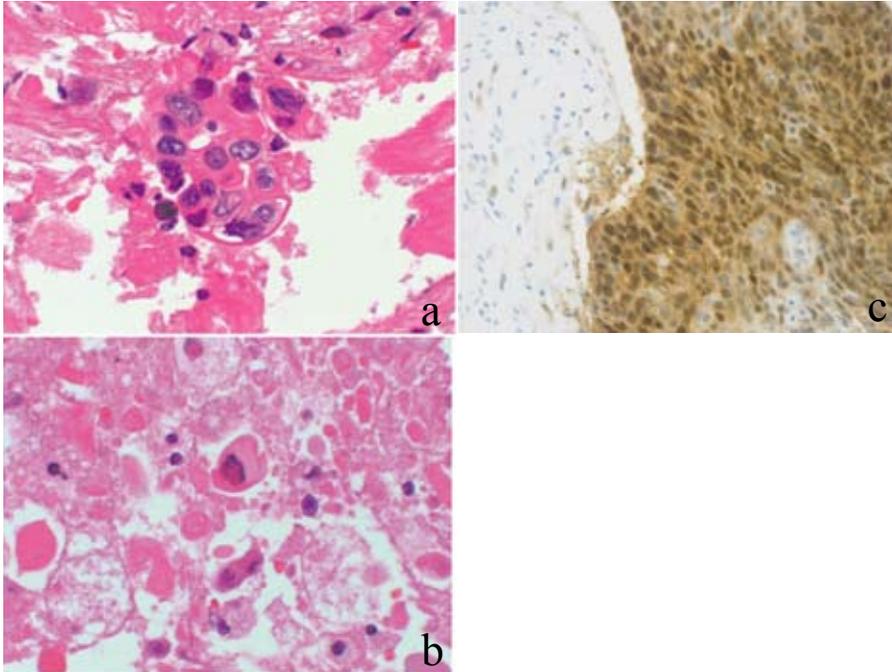


Fig.5. Histopathological findings

a: The foam cell infiltration around the cornified cells.

b: Isolated and scattered vesicles of dysplastic cells were found and very little yet viable squamous cancer remained.

c: Immunostaining revealed p16 was positive.

DISCUSSION

The presacral space is bounded anteriorly by the rectum, posteriorly by the sacrum and coccyx, superiorly by the peritoneal reflection (generally below the S2 level), and inferiorly by the pelvic floor muscles. This area is a caudal end, the site of fusion of the embryologic hindgut, neuroectoderm and contains many types of tissue and totipotent cells that can lead to the development of various tumors; neurofibrosarcoma, neurilemmoma, leiomyoma, chordoma, teratoid, lipoma, desmoids, GIST (Gastrointestinal stromal tumor) and metastatic tumors. Although the true incidence of such tumors is unknown, it is estimated at one in every 40,000 hospital admissions^{1,2)}. In general, they are asymptomatic over prolonged periods and are often discovered incidently^{3,4)}. Digital rectal examination is very important for diagnosis of presacral tumors, Testini *et al.*⁵⁾ reported that 76 %

- 90 % of patients presented with palpable tumors in the digital rectal examination. CT and/or MRI have become the best diagnostic modalities for presacral tumors. CT can be used to assess size, upper extent, behavior, and whether surrounding structures are involved⁶⁾. MRI is highly recommended in presacral tumors, because of its multiplanar capacity and particularly useful in delineating soft-tissue planes and bony and nerve invasion⁷⁾. Generally, it is said that needle biopsy for presacral tumors should be avoided because complications including meningitis after puncture of a meningocele, infection of a cystic tumor, malignant extension through the biopsy tract or bleeding can occur⁸⁾. But in our case, histological diagnosis was required to distinguish whether originating or metastatic tumor.

In this case, the patient had had stomach cancer in the past. However, the cancer was at an early stage and histologically classified as adenocarcinoma.

Therefore, the squamous cell carcinoma was not likely metastasized from the stomach cancer. We could find no report of squamous cell carcinoma originating from the presacral space, other than malignant regeneration of epidermoid cyst, which is very rare. Hawkins *et al.*⁹⁾ recognized that cystic tumor masses were generated as a developmental cyst by developmental error during the fetal stage, and made 3 histological classifications: dermoid cysts, epidermoid cysts and mucus secreting cysts. Among the three, epidermoid cysts consist of stratified squamous epithelia that do not include cutaneous appendages, and there are also some reports that the masses turned malignant^{10,11)}. Moreover, our patient's cyst from 4 years ago showed low density, meaning it was a cystic node. Therefore, it can be said that there is a high possibility that an epidermoid cyst was the cause of the tumor in this case. Also, immunostaining for the tumor from this patient was p16-positive. p16 is characteristically positive when there is a high-grade squamous intraepithelial lesion (HSIL) related to an infection of high-risk human papillomavirus in the uterine cervix, or when there is an adenocarcinoma in situ (AIS). This polity was found from the thorough gynecological examinations before and after the surgery. Thus, the positive result is considered significant when classifying reactive squamous atypia or not. No abnormality was diagnosed with isolated squamous cancer derived from the tissues in the vaginal portion of the cervix, not a metastatic lesion.

Excision of a presacral tumor is essential, even in asymptomatic patients for the possibility of malignancy, potential dystocia in women of childbearing age, future malignant transformation or infection. The four main approaches to excision of presacral tumors are as follows: the transsacral approach with or without coccygeal excision, the transabdominal approach, the combined transsacral/transabdominal approach, and the transrectal

approach. For early-stage cancer in the rectum below the peritoneal reflection, a trans-sacral approach for local rectal excision can be performed to preserve anal sphincter functions. As stated earlier, isolated presacral squamous cell carcinoma is rare and there is no standard surgical procedure. The surgical approach and extent of resection for presacral lesion is determined by the location, size and its malignant potential while taking into account whether or not the sacrum or adjacent viscera is involved¹²⁾. Localio¹³⁾ suggested that when treating a presacral tumor, a transsacral approach should be chosen when a tumor is benign and smaller than 8 cm, and an abdominosacral approach should be chosen when the tumor has a larger diameter or is malignant and could cause an infection. Recently, some reports revealed if the tumor is positioned below the level of the S3 vertebra, transsacral approach can be considered. But tumors that extend above S3 require an abdominal or combined approach^{12,14)}.

In our case, irradiation (10MV x-ray, 2 Gy parallel opposing portals per treatment, 5 times a week, total 40 Gy) and chemotherapy (MMC 100 mg/m², D1+5-FU 1,000 mg/m², D1-4) were started based on standard treatment for anal canal cancer, which is generally squamous cell carcinoma^{15,16)}. Metastasis to neighboring lymph nodes was not found before the surgery, and apparent shrinkage was recognized after the radiotherapy but before the surgery. Therefore, removing the tumor was possible because the resection stump was negative, including the rectal adventitia that trans-sacrally adhered to the tumor. Bowel movement function after the surgery was good and currently there is no sign of recurrence or metastasis including in the local areas. Therefore, it can be concluded that the approach in this report to treat isolated presacral squamous cell carcinoma was effective.

REFERENCES

- 1) Whittaker LD, Pemberton JD. Tumors ventral to the

- sacrum. *Am Surg* 107: 96-106, 1938
- 2) Wolpet A, Beer-Gabel M, Lifscitz O, Zbar AP. The management of presacral masses in the adult. *Tech Coloproctol* 6: 43-49, 2002
 - 3) Buchs N, Taylor S, Roche B. The posterior approach for low retrorectal tumors in adults. *Int J Colorectal Dis* 22: 381-385, 2007
 - 4) Glasgow SC, Dieyz DW. Retrorectal tumors. *Clin Colon Rectal Surg* 19: 61-68, 2006
 - 5) Testini M, Catalano G Jr, Macarini L, Paccione F. Diagnosis and surgical treatment of retroperitoneal tumors. *Int Surg* 81: 88-93, 1996
 - 6) Ludwig KA, Kalady MF. Trans-sacral approaches for presacral cyst/rectal tumor. *Oper Tech Gen Surg* :126-136, 2005
 - 7) Wetzel LH, Levine E. Pictorial essay: MR imaging of sacral and presacral lesions. *Am J Roentgenol* 154: 771-775, 1990
 - 8) Aranda-Narváez JM, González-Sánchez AJ, Montiel-Casado C, Sánchez-Pérez B, Jiménez-Mazure C, Valle-Carbajo M, Santoyo-Santoyo J. Posterior approach (Kraske procedure) for surgical treatment of presacral tumors. *World Gastrointest Surg* 4: 126-130, 2012
 - 9) Hawins WJ, Jackman RJ. Developmental cysts as a source of perianal abscesses, sinuses and fistulas. *Am J Surg* 86: 678-683, 1953
 - 10) Nagano Y, Yoshimoto N, Nakajima S, Hosoi H. Squamous cell carcinoma arising from presacral epidermoid cyst in an adult-Report of a case-. *Nihon Rinsyou Geka Gakkaisi* 59: 2695-2699, 1998 (Article in Japanese)
 - 11) Kanno A, Naito H, Takahashi M, Goto S, Ueno T, Hakozaiki H. A case of squamous cell carcinoma develops in presacral epidermoid cyst. *Geka* 68: 338-342, 2006 (Article in Japanese)
 - 12) Woodfield JC, Charmers AG, Phillips N, Sagar PM. Algorithms for the surgical management of retrorectal tumors. *Br J Surg* 95: 214-221, 2008
 - 13) Localio SA, Eng K, Ranson JH. Abdominosacral approach for retrorectal tumors. *Ann Surg* 191: 555-560, 1980
 - 14) Hassan I, Wietfeldt ED. Presacral tumors: diagnosis and management. *Clin Colon Rectal Surg* 22: 84-93, 2009
 - 15) Flam M, John M, Pajak TF, Petrelli N, Myerson R, Doggett S, Quivey J, Rotman M, Kerman H, Coia L, Murray K. Role of mitomycin in combination with fluorouracil and radiotherapy, and of salvage chemoradiation in the definitive nonsurgical treatment of epidermoid carcinoma of the anal canal: results of a phase III randomized intergroup study. *J Clin Oncol* 14: 2527-2539, 1996
 - 16) Bartelink H, Roelofsen F, Eschwege F, Rougier P, Bosset JF, Gonzalez DG, Peiffert D, vanGlabbeke M, Pierart M. Concomitant radiotherapy and chemotherapy is superior to radiotherapy alone in the treatment of locally advanced anal cancer: results of a phase III randomized trial of the European Organization for Research and Treatment of Cancer Radiotherapy and Gastrointestinal Cooperative Groups. *J Clin Oncol* 15: 2040-2049, 1997

