

Laparoscopic Approach to Presacral Recurrence of Sacrococcygeal Teratoma

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A 6-month-old girl was presented to our hospital due to a presacral mass found 5 months after surgery of sacrococcygeal teratoma. The original tumor was a 63 x 50 mm sized round cyst connecting to the coccyx, observed with computed tomography. The initial operation was performed with *en bloc* removal of the tumor along with the coccyx in the prone position. During a routine follow up, ultrasonography indicated a possible local recurrence, 5 months after the initial operation. The magnetic resonance imaging revealed a polycystic formation with a diameter of 20 x 11 x 17 mm in the presacral space. The laparoscopic operation was conducted with the patient in the lithotomy and Trendelenburg position. The broad ligament of uterus was fixed to the abdominal skin and the rectum was mobilized to identify the tumor, which was resected laparoscopically. A histopathological examination showed the tumor to be a mature cystic teratoma. We observed her without any additional treatment and no recurrence is seen after 6 months.

Key words: Laparoscopic resection, presacral recurrence, retrorectal tumor, Sacrococcygeal teratoma, teratoma in infant

INTRODUCTION

Presacral recurrence of sacrococcygeal teratoma are often extended to the retrorectal space. There is a tendency toward malignant change with increasing age in the pediatric sacrococcygeal teratoma. Complete resection of the tumor is necessary to provide an excellent prognosis. The surgical resection of a retrorectal tumor can be performed using several approaches, depending on the location and size of the tumor. Complete resection for the retrorectal tumors are difficult with either open laparotomy or posterior approach. Here, we report our experience with a successful excision by laparoscopic surgery of a recurrent sacrococcygeal teratoma in the retrorectal space.

CASE REPORT

A 6-month-old girl was presented to our hospital due to a presacral mass found 5 months after surgery of sacrococcygeal teratoma. The original tumor was a 63 x 50 mm sized round cyst connecting to the coccyx with computed tomography (Fig. 1A). Laboratory tests returned normal findings. Alfa-fetoprotein (AFP) level was 34,563 ng/ml, considered to be normal in newborn infant. Initial operation was performed with *en bloc* removal of the tumor along with the coccyx in the prone position. There were no complications during the operation and postoperative period. The histopathological examination showed a mature cystic teratoma with negative margins. During a routine follow up, ultrasonography indicated a possible local

recurrence, 5 months after the initial operation. The magnetic resonance imaging revealed a polycystic formation with a diameter of 20 x 11 x 17 mm in the presacral space (Fig. 1B). At this point, AFP level was decreased to 82.2 ng/ml.

The laparoscopic operation was conducted with the patient in the lithotomy and Trendelenburg position. The patient was placed transversely at the end of the operating table. The surgeon stood at the head of the patient and the monitor was placed between the patient legs. A 12 mm trocar was inserted into the umbilicus for the camera, and two 5 mm trocars were inserted into the both side of flank, respectively. The broad ligament of uterus was fixed to the abdominal skin using 4-0 nonabsorbable suture at the inguinal area to secure a working space (Fig. 1C). The mesorectum was dissected free and the rectum was tied with a vessel tape (Fig. 1D). An additional 3 mm trocar was inserted into the right suprapubic area for the traction of the mobilized rectum by the assistant sitting between the legs under the monitor. The cystic lesion was identified after the further dissection of retroperitoneum (Fig. 1E). Although the cyst was tightly adherent to the presacral fascia, a total dissection of the cyst was carried out carefully from the surrounding tissues by using monopolar L-hook device and bipolar scissors (Fig. 1F). The pelvic floor was reconstructed with absorbable suture. The histopathological examination showed a benign mature cystic teratoma and the complete resection was achieved with negative margins. The postoperative course was uneventful and the patient

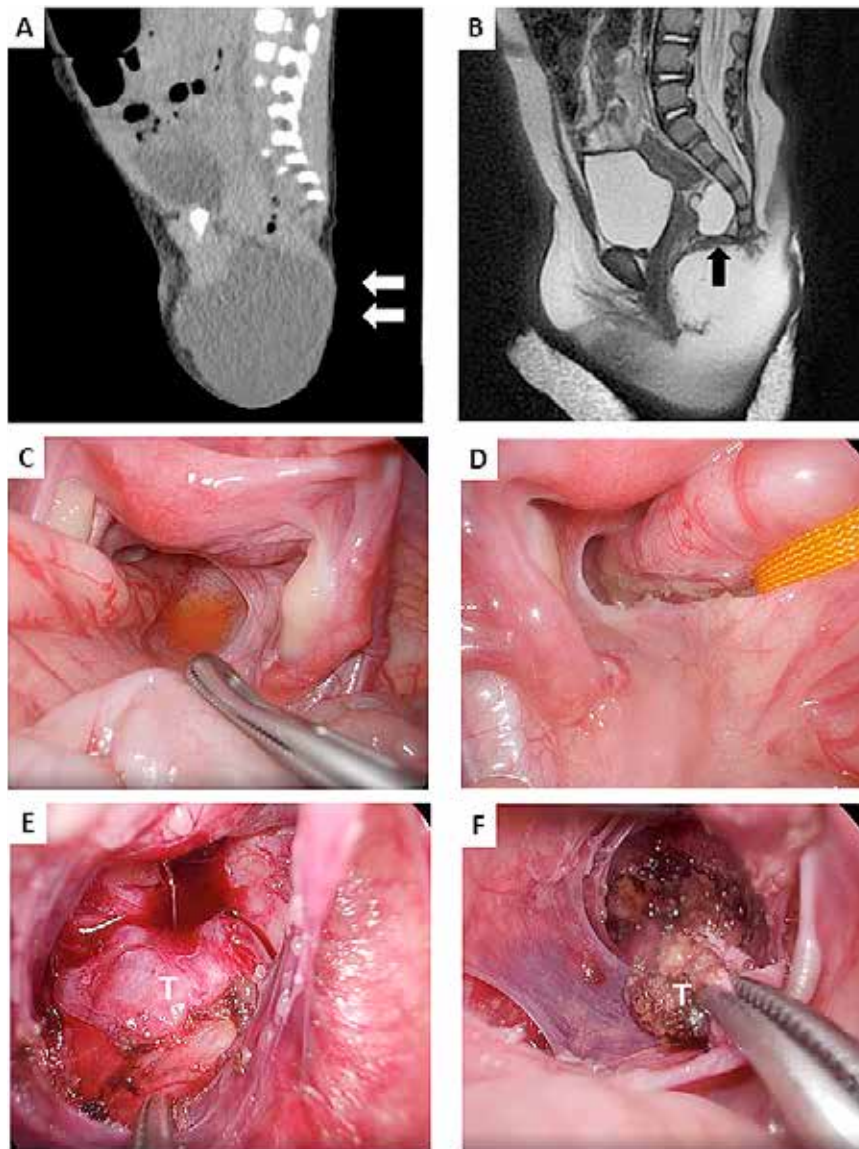


Fig. 1 (A) Computed tomography revealing a round cystic lesion with a diameter of 63 x 50 mm connected to coccyx.
 (B) Magnetic resonance imaging revealing a polycystic formation with a diameter of 20 x 11 x 17 mm in the presacral space.
 (C) A wide working space obtained by securing the broad ligament of uterus.
 (D) Rectal mobilization
 (E) Identification and dissection of the tumor (T)
 (F) Removal of the tumor (T)

remained free of recurrence during regular clinical follow ups for 6 months.

DISCUSSION

Although Sacrococcygeal teratoma is an unusual tumor, it is still the most common germ cell tumor of infancy [1]. Survival rates higher than 80% are now achieved, but the risk of late recurrences or second malignancies persists. Recurrent tumors are usually local and occur in 4-21% of sacrococcygeal teratomas, which are often located within the pelvis. Incomplete resection, tumor spillage, and residual coccyx have been considered to be the main risk factors for recurrence [2, 3]. A mean recurrence rate of mature teratoma is approximately 10% and immature and malignant teratoma are 33% and 18%, respectively [4]. There is a tendency toward malignant change with

increasing age in the pediatric sacrococcygeal teratoma [5]. In our case the recurrent tumor was discovered 5 months after the initial surgery, suggesting that there might be a remnant or spillage of the tumor. However, when the initial operation was performed, the invasion of the tumor to the abdominal cavity was not expected. Complete resection is the key treatment for achieving an excellent prognosis. The surgical approaches generally include open laparotomy or posterior approach, depending on the size and position of the tumor.

We present a case of successful laparoscopic removal of a presacral recurrence of sacrococcygeal teratoma without any complication. With either open laparotomy or posterior approach, the presacral view is extremely limited. We may cause bleeding and damage to the surrounding tissues and nerves with blind dissection. A laparoscopic approach provided a

good view of the narrow pelvic space and allowed safe resection of the tumor without damaging any residual tissues. However, securing a working space with the laparoscopic approach is not easily accomplished in an infant. The patient was fixed carefully enough with the Trendelenburg position. The mobilization of broad ligament of uterus and rectum were the technical point to obtain a wide working space.

Laparoscopic-assisted excision of initial sacrococcygeal teratoma has been reported in the previous papers [6, 7]. However, to our knowledge, this is the first report of a patient with the laparoscopic approach to presacral recurrence of sacrococcygeal teratoma. The laparoscopic surgery allows safe resection of tumor with early recovery and excellent aesthetic outcomes. This method provides better visualization of the retrorectal space than with either a laparotomy or posterior approach. However, indication needs to be carefully considered with tumor size and location.

DISCLOSURE

The authors declare no conflicts of interest.

AUTHOR CONTRIBUTIONS

E.T. wrote the manuscript. T.W. contributed to the conception, and revised the manuscript. H.H. and T.W. are the primary assistance during the surgery. All authors read and approved the final manuscript.

HUMAN ETHICS

This case report is for academic communication only and not for other purposes. The patient provided informed consent, and patient anonymity was preserved. The approval of our institutional ethics committee was unnecessary for a clinical case report.

REFERENCES

- 1) Rescorla FJ, Sawin RS, Coran AG, Dillon PW, Azizkhan RG. Long-term outcome for infants and children with sacrococcygeal teratoma: a report from the Childrens Cancer Group. *J Pediatr Surg*. 1998; 33(2): 171-6.
- 2) Derikx JP, De Backer A, van de Schoot L, Aronson DC, de Langen ZJ, van den Hoonaard TL *et al.* Factors associated with recurrence and metastasis in sacrococcygeal teratoma. *Br J Surg*. 2006; 93(12): 1543-8.
- 3) Yao W, Li K, Zheng S, Dong K, Xiao X. Analysis of recurrence risks for sacrococcygeal teratoma in children. *J Pediatr Surg*. 2014; 49(12): 1839-42.
- 4) Schneider DT, Wessalowski R, Calaminus G, Pape H, Bamberg M, Engert J *et al.* Treatment of recurrent malignant sacrococcygeal germ cell tumors: analysis of 22 patients registered in the German protocols MAKEI 83/86, 89, and 96. *J Clin Oncol*. 2001; 19(7): 1951-60.
- 5) Monteiro M, Cunha TM, Catarino A, Tomé V. Case report: sacrococcygeal teratoma with malignant transformation in an adult female: CT and MRI findings. *Br J Radiol*. 2002; 75(895): 620-3.
- 6) Lee KH, Tam YH, Chan KW, Cheung ST, Sihoe J, Yeung CK. Laparoscopic-assisted excision of sacrococcygeal teratoma in children. *J Laparoendosc Adv Surg Tech A*. 2008; 18(2): 296-301.
- 7) Bax NM, van der Zee DC. The laparoscopic approach to sacrococcygeal teratomas. *Surg Endosc*. 2004 Jan; 18(1): 128-30.