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Original Article

Sacrococcygeal Teratoma: A Survey by the Korean Association of Pediatric Surgeons in 2018

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Conflict of Interest
No potential conflict of interest relevant to this article was reported.

ABSTRACT

Purpose: The Korean Association of Pediatric Surgeons (KAPS) performed a nationwide survey on sacrococcygeal teratoma in 2018.
Methods: The authors reviewed and analyzed the clinical data of patients who had been treated for sacrococcygeal teratoma by KAPS members from 2008 to 2017.
Results: A total of 189 patients from 18 institutes were registered for the study, which was the first national survey of this disease dealing with a large number of patients in Korea. The results were discussed at the 34th annual meeting of KAPS, which was held in Jeonju on June 21–22, 2018.
Conclusions: We believe that this study could be utilized as a guideline for the treatment of sacrococcygeal teratoma to diminish pediatric surgeons' difficulties in treating this disease and thus lead to better outcomes.

Keywords: Teratoma; Sacrococcygeal region; Surveys and questionnaires

INTRODUCTION

Since 1991, the Korean Association of Pediatric Surgeons (KAPS) has performed annual nationwide studies, each year addressing a different topic relating to pediatric diseases, and the results of these studies are discussed at each respective annual meeting of KAPS. They are also summarized and published in *Advances in Pediatric Surgery*, the official journal of the KAPS. The list of study topics is summarized in **Table 1**. The 34th annual meeting of KAPS was held in Jeonju on June 21–22, 2018, and the topic was sacrococcygeal teratoma, which was discussed for the first time at an annual meeting of KAPS.

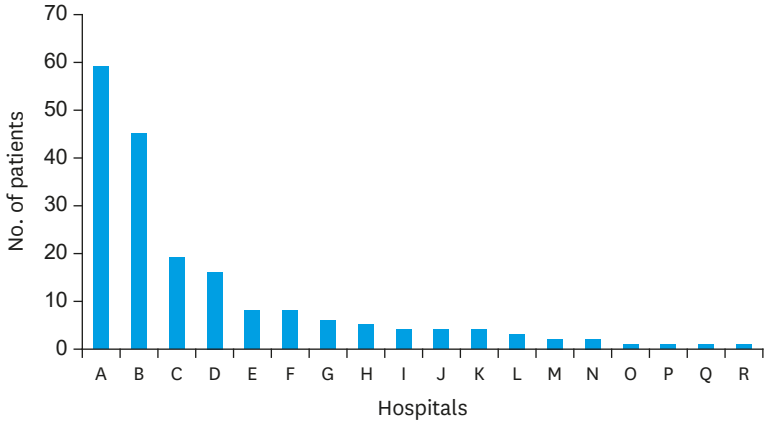

Fig. 1. Hospital distribution of the patients with sacrococcygeal teratoma who underwent surgical treatment.

METHODS

The authors reviewed and analyzed the clinical data of patients who had been treated for sacrococcygeal teratoma by KAPS members from 2008 to 2017. We used Microsoft Access 2016® (Microsoft, Redmond, WA, USA) for the patient registry and data collection. All the data were analyzed using IBM SPSS version 23 statistical software (IBM Co., Armonk, NY, USA), and p-values <0.05 were considered statistically significant.

RESULTS

1. Demographics

A total of 189 patients with sacrococcygeal teratoma from 18 institutes were registered for the study (**Fig. 1**). The top 4 institutes treated 73.5% of all the patients during the study period. The patients' demographics are summarized in **Table 2**. The disease was found to have occurred predominantly in females (M:F=1:2.71). A total of 37 accompanying malformations were present in 29 patients (15.3%), and 15 of these patients were diagnosed with Currarino syndrome. Most patients (n=137, 72.5%) were diagnosed and underwent surgery during the neonatal period, but 27 (14.3%) patients underwent surgery beyond the age of 1 year. In this

Table 1. The list of topics addressed at each annual meeting of the Korean Association of Pediatric Surgeons since 1991

Year and topic			
1991	Current situation in Korean pediatric surgery	2005[a)]	Necrotizing enterocolitis
1992	Inguinal hernia	2006[a)]	Acute appendicitis
1993	Hirschsprung disease	2007	Prospect of pediatric surgery
1994	Anorectal malformation	2008	Inguinal hernia
1995[a)]	Esophageal atresia and tracheoesophageal fistula	2009	Hirschsprung disease
1996[a)]	Branchial anomalies	2010[a)]	Intestinal atresia
1997[a)]	Infantile hypertrophic pyloric stenosis	2011[a)]	Biliary atresia
1998[a)]	Intestinal atresia	2012	Statistics of pediatric surgery disease
1999[a)]	Anorectal malformations	2013[a)]	Minimally invasive surgery
2000[a)]	Index cases in pediatric surgery	2014[a)]	Newborns surgery with congenital anomalies
2001[a)]	Biliary atresia	2015[a)]	Neonate congenital Bochdalek hernia
2002[a)]	Choledochal cyst	2016	Esophageal atresia with tracheoesophageal fistula
2003[a)]	Congenital posterolateral diaphragmatic hernia	2017	Choledochal cyst
2004	Trend of pediatric surgery disease	2018[a)]	Sacrococcygeal teratoma

[a)]These studies were published in *Advances in Pediatric Surgery*.

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Table 2. Patient demographics

Characteristic	Value
Sex (M:F)	1:2.71 (51:138)
Gestational age (wk, n=176)	37.9±2.8
Birth weight (kg, n=179)	3.18±0.52 (0.97–4.71)
Mode of delivery	
Normal spontaneous vaginal delivery	76 (40.2)
C-section	100 (52.9)
Unknown	23
Accompanied malformation	29/189[a] (15.3)
Cardiovascular	8
Gastrointestinal	5
Genitourinary	4
Musculoskeletal	4
Chromosomal	2
Other	6
Currarino syndrome	15
Age at time of surgery	
<29 day	137
29 day–2 mo	6
2–3 mo	7
3–12 mo	12
>12 mo	27

Values are presented as mean±standard deviation or number (%).
[a]Including multiple selection.

study, we compared the results of the neonates who were younger than 29 days with those of the "old-age" group, which comprised patients who were older than 1 year.

2. Preoperative evaluation and treatment

The most common symptom was sacral mass, and 85.6% of the neonates were diagnosed prenatally. Ultrasonography was the most common diagnostic tool in the prenatal period, but magnetic resonance imaging (MRI) was the most popular study after birth. Eleven patients underwent an in-utero procedure. Preoperative serum levels of α-fetoprotein (AFP) were evaluated in 138 patients, and it was the most common tumor marker, followed by β-human chorionic gonadotropin and carcinoembryonic antigen. Almost all the patients underwent surgical treatment without preoperative treatment, with only 8 patients requiring preoperative chemotherapy or embolization. Anterior displacement of the rectum and obstructive hydronephrosis were the most common preoperative complications because of the mass effect of the tumor (**Table 3**).

3. Operative treatment

Almost all the neonatal patients underwent surgical treatment without delay. Their median age at the time of surgery was 4 days after birth, and the median body weight was 3.0 kg. Most of the patients underwent surgery once, but 22.2% required an operation more than one time. The perineal approach as a surgical method was so common that 93.7% of the operations were performed using only a perineal approach. A total of 84.1% of the patients underwent complete excision of the tumor, and 21 patients had 24 intra-operative complications among them. Of these, intraoperative bleeding was the most common complication, followed by cerebrospinal fluid leakage. Postoperatively, 15.9% had complications. Problems with the wound constituted the most common postoperative complication and the most common cause of reoperation during the postoperative hospitalization period (**Table 4**).

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Table 3. Preoperative evaluation and treatment

Characteristic	Total	Neonate	Old-age
Clinical presentation[a]	(n=182)	(n=132)	(n=26)
Abdominal pain/distension	13 (7.1)	5 (3.8)	2 (7.7)
Mass	145 (79.7)	120 (90.9)	11 (42.3)
Constipation	10 (5.5)	0	5 (19.2)
No symptoms	9 (4.9)	7 (5.3)	1 (3.8)
Other	14 (7.7)	1 (0.8)	9 (34.6)
Prenatal diagnosis in neonate[a]		113/135 (85.6)	
Prenatal US		113	
Prenatal MRI		3 (2.2)	
In-utero procedure		11/135 (8.1)	
RFA		8	
Aspiration		2	
Cystic-amniotic shunt		1	
Diagnostic work-up after delivery[a]	(n=182)	(n=132)	(n=26)
US	105 (57.7)	83 (62.9)	10 (38.5)
CT	16 (8.8)	9 (6.8)	5 (19.2)
MRI	159 (87.4)	112 (84.8)	24 (92.3)
Preoperative biopsy	4 (2.2)	0	4 (15.4)
Other	3 (1.6)	2 (1.5)	1 (3.8)
Pre-op AFP (ng/mL)	(n=138)	(n=111)	(n=14)
Mean±SD		87,658±77,076	20,287±42,396
Median (range)		64,745 (0.8–600,000)	2.1 (0.8–150,730)
Preoperative treatment	(n=189)	(n=137)	(n=27)
Preoperative chemotherapy	6		6
Preoperative embolization	1	1	
Preoperative chemotherapy and ASCT[b]	1		1
Preoperative complications	(n=189)	(n=137)	(n=27)
Yes	30 (15.9)	26 (19.0)	1 (3.7)
No	159 (84.1)	111 (81.0)	26 (96.3)

Values are presented as number (%).
US, ultrasound; MRI, magnetic resonance imaging; RFA, radiofrequency ablation; CT, computed tomography; AFP, α-fetoprotein; SD, standard deviation; ASCT, autologous stem cell transplantation.
[a]Including multiple selection; [b]Autologous stem cell transplantation.

4. Tumor characteristics

The maximal diameters of the tumors in the neonate group were significantly larger than those in the old-age group (7.9±5.1 vs. 4.2±4.8 cm, p<0.001). Using the Altman classification, the most common tumor type was type I in the neonatal group, but type IV was the most common type in the old-age group (p<0.001). Mature and cystic or predominantly cystic mixed type were the main characteristics of the tumors. In this study, we found that the pathologic reports of 12 patients did not belong to teratoma or germ cell tumors, but we included them in this study because their clinical features were more compatible with sacrococcygeal teratoma. A total of 11 patients required postoperative chemotherapy (**Table 5**).

5. Postoperative treatment and follow-up

Postoperative follow-up was available at a median age of 41 months (**Table 6**). Most of the patients underwent follow-ups at less than a one-year interval. Ultrasonography and MRI were the most common evaluation methods for follow-up. During the follow-up period, 39 patients had tumor recurrence. The pathological characteristics of the recurrent tumors are described in **Table 7**. Thirty-three patients underwent excision or excision with chemotherapy. Long-term complications were found in 44 patients, and most of them correlated with the function of defecation.

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Table 4. Operative treatment

Characteristic	Total (n=189)	Neonate (n=137)	Old-age (n=27)
Age at time of surgery (day)			
Mean	305.9±957.1	6.9±9.2	1,985.3±1,784.9
Median	6 (0–7,217)	4 (0–63)	1,426 (397–7,217)
No. of operations			
1	145 (76.7)	106 (77.4)	21 (77.8)
2	37 (19.6)	27 (19.7)	3 (11.1)
3	6 (3.2)	4 (2.9)	2 (7.4)
5	1 (0.5)	0	1 (3.7)
Body weight at time of surgery (kg)			
Mean±SD	6.2±8.8	3.1±0.6	22.0±15.9
Median (range)	3 (2–69)	3 (2–5)	16 (8–69)
Operation time			
Mean±SD	148.0±89.6	150.3±82.8	155.7±129.4
Median (range)	125 (20–590)	132 (20–410)	110 (34–590)
Mode of surgery			
Perineal approach	177 (93.7)	131 (95.6)	23 (85.2)
Perineal+laparotomy/laparoscopic	9	5	2
Others	3	1	2
Results of operation			
Complete excision	159 (84.1)	116 (84.7)	23 (85.2)
Complete excision with spillage	12 (6.3)	6 (4.4)	2 (7.4)
Incomplete excision	18 (9.5)	15 (10.9)	2 (7.4)
Intraoperative complications	21/189 (11.1)	15/137 (10.9)	3/27 (11.1)
Bleeding	8 (38.1)	6 (40.0)	2 (66.7)
CSF leakage	4 (19.0)	1 (6.7)	1 (33.3)
CPCR	4 (19.0)	4 (26.7)	0
Other complications	6 (28.6)	5 (33.3)	0
Postoperative complications[a)]	30/189	24/137	3/27
Bleeding	3 (10.0)	3 (12.5)	0 (0)
Wound problem	17 (56.7)	14 (58.3)	1 (33.3)
Intestinal obstruction	1 (3.3)	1 (4.2)	0 (0)
DIC	5 (16.7)	5 (20.8)	0 (0)
Other	7 (23.3)	4 (16.7)	2 (66.7)
Reoperation during hospitalization	9/189 (4.8)	7/137 (5.1)	1/27 (3.7)
Postoperative chemotherapy	11 (5.8)	5 (3.6)	4 (14.8)

Values are presented as number (%).
SD, standard deviation; CSF, cerebrospinal fluid; CPCR, cerebral resuscitation; DIC, disseminated intravascular coagulation.
[a)]Including multiple selection.

Currently, among the 189 patients, 1 death had occurred in a case of incomplete excision, 136 patients are living without tumors, 19 are living with tumors, and 33 patients have been lost to follow-up (**Fig. 2**).

6. Questionnaire for sacrococcygeal teratoma

The questionnaire for sacrococcygeal teratoma consisted of 7 questions, and 22 regular members of KAPS answered them. The following are the questions and the number of answers for each item (**Table 8**).

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Table 5. Tumor characteristics

Characteristic	Total (n=189)	Neonate (n=137)	Old-age (n=27)
Pathology of tumor			
Mature teratoma	138 (73.0)	103 (75.2)	16 (59.3)
Immature teratoma	33 (17.5)	30 (21.9)	2 (7.4)
Grade 1	4 (13.8)	4 (14.8)	0 (0)
Grade 2	10 (34.5)	8 (29.6)	1 (100)
Grade 3	15 (51.7)	15 (55.6)	0 (0)
Grade unknown	4 (13.8)	3	1
Mixed	4 (2.1)	2 (1.5)	0
Malignant (yolk sac)	2 (1.1)	1 (0.7)	1 (3.7)
Other[a)]	12 (6.3)	1 (0.7)	8 (29.6)
Largest tumor length (cm)	6.8±4.8	7.9±5.1	4.2±4.8
Type of tumor component			
Cystic type	63 (34.1)	43 (31.6)	9 (37.5)
Predominantly cystic mixed type	61 (33.0)	48 (35.3)	6 (25.0)
Predominantly solid mixed type	32 (17.3)	29 (21.3)	1 (4.2)
Solid type	29 (15.7)	16 (11.8)	8 (33.3)
Unknown	4	1	2
Altman classification			
I	68 (36.0)	53 (38.7)	8 (29.6)
II	51 (27.0)	47 (34.3)	2 (7.4)
III	25 (13.2)	21 (15.3)	1 (3.7)
IV	45 (23.8)	16 (11.7)	16 (59.3)

Values are presented as number (%) or mean±standard deviation.
[a)]Epidermal cyst (2), lipoma (2), lipoblastoma (2), duplication cyst (1), solitary fibrous tumor (1), infantile fibrosarcoma (1), lipomyelomeningocele (1), lymphangioma (1), epithelioid hemangioendothelioma (1).

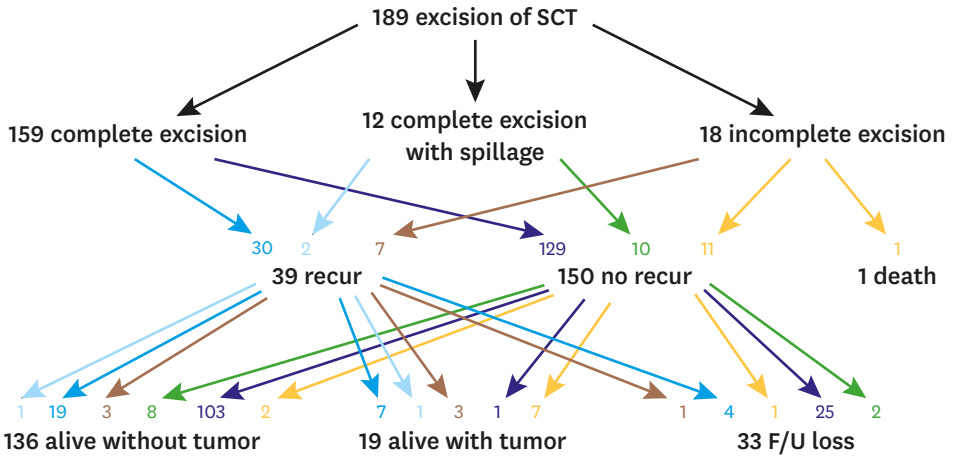

Fig. 2. Summary of treatments and prognoses.
SCT, sacrococcygeal teratoma; F/U, follow-up.

DISCUSSION

Studies on the clinical characteristics of and strategies used to treat sacrococcygeal teratoma are not rare, but its low incidence usually makes it challenging for pediatric surgeons to accumulate experience in this disease. Previous studies about this disease in Korea have been limited, and only a few studies have been published [1-4]. The significance of this study is that it is the first nationwide survey on sacrococcygeal teratoma in Korea and includes a significant number of patients.

Our study showed excellent results after surgical treatment. There was only 1 reported case of death after surgery, and the number of long-term functional complications was not high.

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Table 6. Postoperative treatment and follow-up

Characteristic	Total (n=189)	Neonate (n=137)	Old-age (n=27)
Postoperative follow-up (mo)			
Age at last follow-up	47.9±38.9 (median 41, range 0–243)		
Postoperative follow-up	37.9±27.6 (median 34, range 0–112)		
Follow-up method[a]			
US	89 (47.1)	66 (48.2)	13 (48.1)
CT	23 (12.2)	17 (12.4)	4 (14.8)
MRI	98 (51.9)	77 (56.2)	10 (37.3)
PET	5 (2.6)	3 (2.2)	2 (7.4)
Other	4 (2.1)	3 (2.2)	0 (0)
Follow-up interval (mo)			
1–3	25		
4–6	30		
7–12	37		
>12	14		
Tumor recurrence	39 (20.6)	28	5
Detection of tumor recurrence			
Physical examination	1 (2.6)	1 (3.6)	0 (0)
Elevated tumor marker	3 (7.7)	3 (10.7)	0 (0)
MRI	27 (69.2)	20 (71.4)	5 (100)
U/S	7 (17.9)	4 (14.3)	0 (0)
Other	1 (2.6)	0 (0)	0 (0)
Treatment of tumor recurrence			
Excision	21 (53.8)	14 (50.0)	3 (60.0)
Excision+CTx	12 (30.8)	9 (32.1)	1 (20.0)
CTx only	2 (5.1)	1 (3.6)	1 (20.0)
Observation	2 (5.1)	2 (7.1)	0 (0)
Other	2 (5.1)	2 (7.1)	0 (0)
Long-term functional complications[a]	44		
Constipation	21		
Soiling	14		
Urinary incontinence	7		
Lower extremity weakness	5		
Other	7		

Values are presented as mean±standard deviation or number (%).
US, ultrasound; CT, computed tomography; MRI, magnetic resonance imaging; PET, positron emission tomography; CTx, chemotherapy.
[a]Including multiple selection.

Table 7. Recurrent tumor pathology

Characteristic	Total (n=33)	Neonate (n=23)	Old-age (n=4)
Mature	19 (57.6)	15 (78.9)	1 (25.0)
Original pathology		Mature (10), immature (8), mixed (1)	
Immature	2 (6.1)	2 (8.7)	0 (0)
Original pathology		Mature (1), immature (1)	
Malignant (yolk sac)	6 (18.2)	3 (13.0)	1 (25.0)
Original pathology		Mature (3), immature (1), yolk sac (2)	
Mixed	1 (3.0)	1 (4.3)	0 (0)
Original pathology	Mature (1)		
Other[a]	5 (15.2)	2 (8.7)	2 (50.0)
Original pathology	Mature (3), immature (1), epithelioid hemangioendothelioma (1)		

[a]Epithelioid hemangioendothelioma (1), inflamed granulation tissue (1), lipomeningomyelocele (1), lipoma (1), no evidence of residual teratoma but r/o recurred tumor on follow-up magnetic resonance imaging (1).

Although not all the patients were followed-up for a long time, it is likely that the mortality of those who were lost to follow-up was not affected because the pediatric surgeons would probably have treated them if they had experienced any problems. These results are similar to or better than those of previously published studies [5-10].

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Table 8. Questionnaire for sacrococcygeal teratoma

Questionnaire	No.
1. Which of the following tests are the most important for the preoperative diagnosis of sacrococcygeal teratoma (excluding physical findings and multiple selections available)?	
① AFP	12
② US	4
③ CT	6
④ MRI	16
⑤ Biopsy	0
2. Have you needed to do a coccyx resection for patients with sacrococcygeal teratoma? please describe the method and extent of resection.	
① No resection	0
② Resection	22
Method: electrical cautery and en bloc resection	
3. Who do patients follow-up with after surgery?	
① Pediatric surgeon	14
② Pediatrician	4
③ Other—both pediatric surgeons and pediatricians (especially in cases of immature or malignant pathologies)	4
4. What tests are performed after surgery? (Multiple options are possible.)	
① AFP	21
② US	11
③ CT	4
④ MRI	12
⑤ Other—rectal exam	1
5. What is the timing (interval) of the postoperative follow-up?	
① Every 6 months after surgery	9
② Every year after surgery	7
③ Every 2 years after surgery	0
④ Less than every 6 months after surgery	4
6. How long after surgery do you follow-up? (n=20)	
① Until 1 year after surgery	0
② Until 2 years after surgery	1
③ Until 3 years after surgery	3
④ Until 4 years after surgery	0
⑤ Until 5 years after surgery	13
⑥ Mature-3 years, immature-5 years	3
7. Do you have any experience with minimally invasive surgery for sacrococcygeal teratoma?	
① Yes	5
② No	17

AFP, α-fetoprotein; US, ultrasound; CT, computed tomography; MRI, magnetic resonance imaging.

This study revealed that this incidence of this disease can be divided into 2 age groups. In the one group, the lesions were detected prenatally, and in the other, the disease was late-onset. Although the incidence of the old-age group was not high, and the prognosis of this group after surgical treatment was also excellent, pediatric surgeons must consider the possibility of this disease in old age.

One of the limitations of this study was that we evaluated only the patients who underwent surgical treatment, and therefore not all patients with sacrococcygeal teratoma were included. The prognosis of sacrococcygeal teratoma would have deteriorated in patients who could not undergo surgical treatment because of their poor general condition or the inoperability of the tumor.

Through the questionnaire, we were able to learn the current status of KAPS members' clinical practices with regard to sacrococcygeal teratoma. Most of the members take care of their patients themselves postoperatively, and they prefer a long-term follow-up of about

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5 years. In rare cases, a few members tried to treat patients using laparoscopic surgery or robotic surgery.

We believe that this study could be utilized as a guideline for the treatment of sacrococcygeal teratoma to diminish pediatric surgeons' difficulties in treating this disease and thus lead to better outcomes.

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