



ORIGINAL ARTICLE

Functional and aesthetic evaluation of sacrococcygeal teratomas. Not everything ends with surgery^{☆,☆☆}



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KEYWORDS

Teratoma;
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Abstract

Introduction: Sacrococcygeal teratoma is the most common solid neonatal tumour. The improvement in survival has meant that postoperative sequelae can be diagnosed and treated. The aim of this article is to evaluate the long-term outcomes of patients treated in our centre.

Material and methods: Records of patients treated for a sacrococcygeal teratoma in our hospital from 1977 to 2014 were retrospectively reviewed. Personal data was collected and a telephone questionnaire was used to assess long-term bowel and urinary habits, as well as an aesthetic and functional self-assessment.

Results: A total of 14 patients were treated during the study period, of whom 11 were females and 3 males, with a mean age at the time of the survey of 17 years (8 months to 37 years). Eight patients completed the questionnaire (57.1%). The mean age of the 8 patients was 23 years (4–37 years), of whom 37.5% were operated on due to a sacrococcygeal teratoma type I, 25% type II, 25% type III, and 12.5% type IV. Two of them (25%) had constipation, and one (12.5%) had faecal incontinence. Two (25%) patients suffered from recurrent urinary tract infections, and 3 (37.5%) patients had urinary incontinence. Five patients (62.5%) had a perception of being physically impaired, with limitation of their social life.

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PALABRAS CLAVE

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Conclusions: The incidence of constipation does not differ from that found in the literature. Faecal incontinence is slightly improved compared to what has been published. However, urinary tract infections and incontinence are more prevalent in our series. Five patients out of the eight that responded suffered from psychosocial problems, according to DAS-59 questionnaire. Patients with SCT require urological, bowel, and psychological counselling, until they have a complete functional and emotional development.

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Evaluación funcional y estética de los teratomas sacrococcígeos. No todo termina con la cirugía

Resumen

Introducción: El teratoma sacrococcígeo es el tumor neonatal sólido más frecuente. El aumento de la supervivencia de estos pacientes ha permitido diagnosticar y tratar secuelas posquirúrgicas. Evaluamos el resultado funcional y estético a largo plazo de los pacientes intervenidos en nuestro centro.

Material y método: Estudio retrospectivo de los teratomas sacrococcígeos intervenidos en nuestro hospital desde 1977 hasta 2014. Se recogieron datos personales de los pacientes y se realizó encuesta telefónica sobre hábito intestinal, urinario y autoevaluación estética y social.

Resultados: En el período de estudio se intervino a 14 pacientes, 11 mujeres y 3 varones, con una edad mediana en el momento de la encuesta de 17 años (8 meses-37 años). Se encuestó a 8 pacientes (57,1%). La edad mediana de esos 8 pacientes fue de 23 años (4-37 años). El 37,5% se intervino por un teratoma sacrococcígeo tipo I, 25% tipo II, 25% tipo III y 12,5% tipo IV. De los pacientes estudiados, 2 (25%) presentan estreñimiento y uno (12,5%) incontinencia fecal. Dos pacientes (25%) presentan infecciones urinarias recurrentes y 3 (37,5%) incontinencia urinaria. Cinco pacientes (62,5%) presentan alterada su percepción física con limitación en su vida social.

Conclusiones: En nuestra serie, se ha encontrado un porcentaje similar a la literatura en la incidencia de estreñimiento. La incontinencia fecal está levemente disminuida respecto a series publicadas. Sin embargo, la frecuencia de infecciones y de incontinencia urinarias es mayor en nuestra serie. Cinco pacientes presentan problemas psicosociales según el cuestionario DAS-59. Los pacientes con teratoma sacrococcígeo requieren asesoramiento urológico, digestivo y psicológico, para poder adquirir un completo desarrollo funcional y emocional.

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Introduction

The incidence of sacrococcygeal teratoma (SCT) is of 1 per 28 500–40 000 live births.^{1,2} When it comes to the long-term outcomes after tumour resection, there is a high prevalence of post-surgical sequelae resulting from local compression by the tumour or iatrogenic surgical trauma,³ while 24.3% of patients have associated congenital anomalies, such as musculoskeletal disorders, genitourinary abnormalities, neural defects, pulmonary disorders and anorectal malformations, among others.⁴

Based on the literature on patients that have undergone surgical resection of SCT, the most frequent sequelae are bowel dysfunction (42% of patients),⁵ bladder dysfunction (36% of patients),⁶ dissatisfaction with cosmetic outcomes (40% of patients)² and recurrence (6.8% to 18.8% of patients).^{7,8} The aim of our study was to assess the long-term bowel function, urinary function, cosmetic and psychosocial outcomes of the patients operated in our hospital over a

period of 37 years, and to compare our results with those of other published case series.

Materials and methods

We performed a retrospective review of surgical protocols and medical records of patients with SCT that underwent surgery in our department between 1977 and 2014. After obtaining approval from the hospital, we telephoned patients aged more than 4 years and administered a questionnaire on bowel habits based on the Krickenbeck International Classification⁹ (Table 1). We assessed urologic involvement by means of a previously validated questionnaire (Shalaby et al.)⁵ (Table 2) and, last of all, we used the DAS-59 scale¹⁰ to detect potential problems in self-perceived appearance. To conclude, we compared our results with those of previous studies.

We also collected data on the type of SCT based on the classification developed by Altman et al.,¹¹ the surgical

Table 1 International classification of Krickenbeck.

1. <i>Voluntary bowel movements</i>	Yes/no
Feeling of urge, capacity to verbalise Hold the bowel movement	
2. <i>Soiling</i>	Yes/no
Grade 1: occasionally (once or twice a week)	
Grade 2: everyday, no social problem	
Grade 3: constant, social problem	
3. <i>Constipation</i>	Yes/no
Grade 1: manageable by changes in diet	
Grade 2: Requires laxative	
Grade 3: Resistant to laxatives and diet	

Source: Holschneider et al.⁹

approach used (sacral and/or abdominal approach), tumour histology, the association with other developmental anomalies and the rate of recurrence during the follow-up.

Results

We identified 14 patients that underwent surgery for treatment of SCT in our hospital in the past 37 years, of who 11 were female (female: male ratio, 3.66:1). The age range of patients at the time of the study was 8 months to 37 years (median, 17 years).

The diagnosis was made antenatally in 8 patients (57.14%), at birth in 4 (28.57%) and delayed in the remaining 2 (at ages 6 months and 4 years). In the last 2, diagnosis resulted from a chance finding of ultrasound examination in the patient aged 6 months, and of computed tomography of the abdomen and pelvis for assessment of recurrent abdominal pain in the patient aged 4 years. Histological examination in the patient operated at age 6 months revealed an endodermal sinus tumour, while the patient operated at age 4 years had a mature cystic teratoma (Tables 3 and 4).

Magnetic resonance imaging in patients with suspected SCT allowed their classification based on the system proposed by Altman et al., with 7 of 14 patients with a predominantly external SCT (Altman type I, 50%), 2 with a type II SCT (14.3%), 4 with a type III SCT (28.6%) and 1 with a type IV SCT (7.1%). The mean tumour diameter was 8.6 cm (range, 2–14 cm). Tumours were cystic in 28.6% of cases, solid in 14.3% and mixed in 57.1%.

The median age at the time of surgery was 12 days (9 h to 4 years), and resection was performed during the neonatal period in 8 of the 14 patients. The surgical approach consisted of an inverted Y- or V-shaped incision in 12 patients and a sacral approach with median supra-infraumbilical laparotomy in the other 2 patients, with no intraoperative complications in any cases. Intraoperative rupture of the cystic component of the tumour with spillage of contents occurred in 7 patients, of who 2 experienced a recurrence. The histological examination at the time of diagnosis determined that the teratoma was mature in 8 patients, immature in 4, and malignant in only 1 patient who had a stage II (low-risk) endodermal sinus tumour. This patient received 14 cycles of chemotherapy and required second-look surgery for excision of the tumour remnant. We were unable to find

the results of the histological examination for one of the cases.

Three patients had associated anomalies: rib agenesis, dysplasia of the left kidney and polymalformative syndrome with thyroid agenesis, dysplasia of the right kidney and hypospadias. The early postoperative complications were wound dehiscence (3 patients) and bowel perforation (1 patient), and required reoperation. One of the patients presented with a urethrovaginal fistula during the long-term follow-up that was not associated with the performed surgery. Another complication detected during the long-term follow-up was a small eventration in a patient in who surgery was performed with a combined abdominosacral approach.

Alpha-fetoprotein (AFP) was abnormally elevated in 2 of the 14 patients (133 466 and 3 000 000 ng/mL) at 24 and 48 h from birth, respectively, and normalised after tumour resection. We defined abnormal elevation as AFP levels above the normal range in neonates (>100 000 ng/mL), as they are always high in this age group.¹²

Two tumours recurred: one mature and one immature. Both recurred as endodermal sinus tumours, so the patients were treated with a chemotherapy regimen based on cisplatin, bleomycin and etoposide.

We administered the questionnaire regarding continence and self-perceived function and appearance to 8 patients (57.1%) aged more than 4 years, excluding 3 patients because they were aged less than 1 year and another 3 because we were unable to reach them via telephone, and the median age was 23 years (4–37 years).

Of all patients, 37.5% underwent resection of a type I SCT, 25% of a type II, another 25% of a type III and 12.5% of a type IV.

Of all surveyed patients, 87.5% reported some form of problem (Table 5):

- Gastrointestinal involvement: applying the Krickenbeck classification, 2 patients had constipation (the 2 patients that required the abdominosacral approach), one of them with encopresis secondary to the SCT; a third patient had faecal incontinence with daily soiling. The rest of the patients (62.5%) did not report gastrointestinal involvement.
- Urologic involvement: 2 patients developed recurrent urinary tract infections (UTIs), 4 reported urinary urgency, urinating more than 8 times a day and getting up to urinate more than once each night. Three patients had urinary incontinence (including the patient with a urethrovaginal fistula), which resulted in an impaired sexual life in one.
- Psychosocial involvement: when we administered the DAS-59 questionnaire, only 3 respondents reported having an acceptable quality of life. A girl aged 4 years was in psychotherapy to treat social isolation and withdrawal and selective mutism. Another female patient aged 18 years had urine leaks during sexual intercourse, which had a psychosocial impact. Lastly, 3 patients aged 4, 29 and 31 perceived their social and cosmetic outcomes as unacceptable due to the appearance of the scar. The youngest of these patients has been referred to plastic surgery for reassessment of her scar.

Table 2 Urologic questionnaire used in the study.

1. *How difficult do you find it to pass urine?*
 - a) Easy
 - b) Sometimes difficult
 - c) Always difficult
2. *How many times do you typically urinate from waking in the morning until sleeping at night?*
 - a) 7 times or less
 - b) Between 8 and 14 times
 - c) 15 times or more
3. *How many times do you typically wake up to urinate from sleeping at night until waking in the morning?*
 - a) 0
 - b) 1
 - c) 2
 - d) 3 or more
4. *How often do you have a sudden urge to urinate that is difficult to defer?*
 - a) Never
 - b) Once a week
 - c) More than once a week
 - d) Once a day
 - e) 2–4 times a day
 - f) At least 5 times a day
5. *Do you have urine leaks?*
 - a) Never
 - b) Occasionally
 - c) Most of the time
 - d) Always
6. *How frequently do you have urine leaks?*
 - a) Never
 - b) Once a week
 - c) More than once a week
 - d) Once a day
 - e) 2–4 times a day
 - f) At least 5 times a day
7. *How often do you have urine leaks during physical activity?*
 - a) Never
 - b) Once a week
 - c) More than once a week
 - d) Once a day
 - e) 2–4 times a day
 - f) At least 5 times a day
8. *How often do you need to use a pad?*
 - a) Never
 - b) Occasionally
 - c) Mostly
 - d) Always
9. *Have you been treated for a urinary tract infection?*
 - a) Never
 - b) Occasionally
 - c) Frequently
 - d) Always
10. *Was the infection confirmed by taking a urine specimen?*
 - a) Yes
 - b) No
 - c) Never had infections

Source: Shalaby et al.⁵

Table 3 Summary of data of surveyed patients.

Patient	Age at diagnosis	Altman	Approach	Intraoperative tumour rupture	Histology	Recurrence	Age at time of survey
1	Antenatal	II	Abdominal-sacral	Yes	60% immature	No	4 years
2	Antenatal	IV	Sacral	Yes	Mature	No	13 years
3	Antenatal	III	Abdominal-sacral	Yes	Mature	No	18 years
4	At birth	II	Sacral	Yes	Immature	Yes	24 years
5	Antenatal	I	Sacral	No	Mature	No	28 years
6	At birth	III	Sacral	No	Immature	No	29 years
7	6 months	I	Sacral	Yes	Malignant: endodermal sinus tumour	No	31 years
8	At birth	I	Sacral	No	Unknown	No	37 years

Table 4 Summary of data of patients that were not surveyed.

Patient	Age at diagnosis	Altman	Approach	Intraoperative tumour rupture	Histology	Recurrence
1	At birth	III	Sacral	No	Mature	No
2	Antenatal	I	Sacral	No	Immature	No
3	Antenatal	I	Sacral	No	Mature	No
4	Antenatal	I	Sacral	No	Mature	No
5	Antenatal	I	Sacral	Yes	Mature	Yes
6	4 years	III	Sacral	Yes	Mature	No

Table 5 Long-term complications in surveyed patients.

Patients	Long-term complications	Survey
1	Eventration	Encopresis. Psychosocial problems
2		Recurrent UTIs
3	Urethrovaginal fistula	Constipation. Urinary urgency. Recurrent UTIs. Urinary incontinence
4	Recurrence. HE: endodermal sinus tumour	No associated problems
5		Urinary urgency
6		Urinary incontinence. Psychosocial problems
7		Urinary urgency. Psychosocial problems
8		Faecal incontinence. Urinary urgency. Urinary incontinence. Psychosocial problems

Discussion

Sacrococcygeal teratoma is the most common neonatal solid tumour.⁵ It is nevertheless a rare disease, as evinced by the low number of patients gathered over periods spanning several decades in most case series,¹ including this one.

Sacrococcygeal teratoma is more prevalent in the female sex, and in our study the male to female ratio was 3.6:1, similar to the ratios reported in other series^{13,14} (although there are also studies where the predominance of female patients seems to be lesser¹⁵).

The classification proposed by Altman et al.¹¹ refers to the anatomy and location of the tumour, but offers no prognostic information.¹⁶ In our series, the proportion of Altman type I teratomas (50%) slightly exceeded the range of

values reported in the literature (proportions between 18% and 46.7% in studies by other authors).^{17,18}

Sacrococcygeal teratomas are associated with other anomalies in up to 18% of cases, and the most frequent abnormalities are anorectal, genital and spinal.¹⁹ They may also be associated with renal anomalies, such as dysplasia or agenesis (found in 2 of our patients).²⁰

The most frequent complication in the immediate post-operative period is wound dehiscence, due to the proximity of the surgical site to the perianal region.¹⁹ Recently, our hospital introduced a new protocol that includes bowel preparation the day before surgery (antegrade gut lavage with polyethylene glycol delivered through a nasogastric tube, supplemented with cleansing enemas) based on the recent literature,²¹⁻²⁵ and placement of a rectal tube

Table 6 Long-term outcomes of sacrococcygeal teratomas. Bowel function. Summary of studies published between 1992 and 2014 and comparison with our data (last row).

Study authors	Patients (n)	Constipation	Faecal incontinence
Shalaby et al. (2014) ⁵	31	39%	19%
Partridge et al. (2014) ³⁴	45	28.89%	8.88%
Berger et al. (2011) ³⁰	24	14.30%	7.14%
Draper et al. (2009) ³	14	42.85%	7.14%
Khalil et al. (2009) ²⁹	12	25%	8.33%
Derikx et al. (2007) ²	79	17%	13%
Gabra et al. (2006) ¹⁷	33	25%	30%
Rintala et al. (1993) ³¹	26	34.60%	27%
Havránek et al. (1992) ³⁵	25	16%	40%
Villamil et al. (2017) ³⁷	8	25%	12.5%

Table 7 Long-term outcomes of sacrococcygeal teratomas. Urologic function. Summary of studies published between 1975 and 2014 and comparison with our data (last row).

Study authors	Patients (n)	Recurrent UTIs	Urinary incontinence
Shalaby et al. (2014) ⁵	31	29%	29%
Partridge et al. (2014) ³⁴	45		15.55%
Berger et al. (2011) ³⁰	24	10%	
Draper et al. (2009) ³	14		7.14%
Derikx et al. (2007) ²	79		31%
Gabra et al. (2006) ¹⁷	33		20%
Schmidt et al. (1999) ¹³	17	11.11%	11.11%
Rintala et al. (1993) ³¹	26		50%
Havránek et al. (1992) ³⁵	25		16%
Mahour et al. (1975) ³⁶	48	6.25%	
Villamil et al. (2017) ³⁷	8	25%	37.5%

during surgery to prevent faecal contamination. This tube also serves as an anatomical guide during the resection of the anterior surface of the tumour.

The chances of malignancy increase when the tumour size is more than 10 cm, in Altman type III and IV tumours (due to delayed diagnosis) and with the presence of solid areas.¹⁹ The prognosis seems to be better in patients that undergo surgery in the first two months of life, with the risk of malignant transformation increasing after this age.²⁶

The main factors responsible for tumour recurrence are incomplete resection,⁴ failure to excise coccyx, immature or malignant histology and intraoperative tumour rupture with spillage of solid components (as opposed to cyst fluid).²⁷

Sacrococcygeal teratomas are germ cell tumours^{3,8} mostly of a benign nature, although endodermal sinus tumours,⁸ which develop from germ cells, can also occur, as seen in 3 of our patients (1 at diagnosis and 2 in the context of recurrence). Extragonadal germ cell tumours amount to approximately two thirds of paediatric germ cell tumours, compared to only 5% to 10% of germ cell tumours in adults.²⁸

Two of the 14 operated patients in our series experienced recurrences. A factor the two patients had in common was that the tumour ruptured during their surgeries, although there was only spillage of fluid, and not solid, contents; furthermore, one of the patients had a tumour with immature histology.

Alpha-fetoprotein levels need to be monitored until it is certain that they have normalised. This marker is usually elevated during foetal life and should be normal by 9 months of age.²⁸ Recurrence may occur in the form of a benign or malignant tumour, and elevation of AFP may be the first indicator that it has occurred.¹⁹ The rate of recurrence is 10% for mature teratomas, 33% for immature teratomas, and 18% for malignant tumours, which is lower because chemotherapy is used in their management.²⁰ The mortality associated with malignant SCTs has decreased significantly in the past few decades with the introduction of regimens based on carboplatin, etoposide and bleomycin.²⁹

The risk of bladder and rectal dysfunction associated with SCT depends on its location, anatomical anomalies in the tumour region, and the complications of surgery.³⁰ In our series we found an incidence of constipation of 25% and an incidence of faecal incontinence of 12.5%. One possible explanation is damage to the afferent nerves during the skeletonisation of the rectum,³ so that utmost care should be exerted during surgery to minimise the risk of faecal incontinence. It is also possible that the incidence of constipation in these patients does not differ from that in the general paediatric population, in which constipation is extremely common.³ Table 6 presents the long-term outcomes as regards constipation and faecal incontinence reported in previous studies that used surveys and questionnaires administered in the clinic, as well as urodynamic

testing, manometry and imaging tests, which were similar to the outcomes found in our study. In a study that assessed anorectal function in 26 adults that underwent excision of an SCT during childhood, 38.54% of survey respondents reported restrictions in their social life due to impaired functioning of the anal sphincter and sexual problems.³¹

When it came to urologic changes, we found that 25% of patients had recurrent urinary tract infections and 37.5% urinary incontinence. The incidence of both problems was higher in our series compared to other studies (Table 7) that used the same methodology described for the assessment of rectal dysfunction. At this point, it may be worth noting that not everything that is published is accurate, as there is a risk of selection bias with underestimation of urologic complications. In this context, an early urologic consultation and multidisciplinary management of the patient should be strongly considered.¹⁴

We now elaborate on the case of a patient that developed an urethrovaginal fistula after tumour resection (a finding previously described in the literature, and secondary to either an associated urogenital sinus or to necrosis of the medial urethra resulting from its compression between the pubic symphysis and the tumour or a iatrogenic lesion).^{27,32} Currently, at age 18 years, the patient has significant urologic manifestations, so we contacted her to arrange for urodynamic testing, the results of which were normal; the patient then started anticholinergic therapy and was referred to the adult urology department.

We decided to include the questions of the DAS-59 scale in the survey to assess for psychosocial problems, as this instrument has been validated, is used in many hospitals and it is highly reproducible. Although the recent study by Kremer et al.³³ did not find statistically significant differences in quality of life between patients operated for SCT and a control group, poor cosmetic results in the gluteal region may lead to distortions in body image, especially in adolescents, and can cause psychological problems and even depression.¹⁹ In the case series published by Derikx et al.,² 40% of patients reported dissatisfaction with the functional and cosmetic outcomes of surgery, and considered the appearance of the resulting scar unacceptable.

To conclude, the prevalences of constipation and faecal incontinence in our series were similar to those reported in other studies (25% vs 26.8% and 12.5% vs 17.3%, respectively). As for the incidence of urologic problems, it was higher in our series, as one patient that was eventually lost to follow-up presented with an urethrovaginal fistula during the postoperative period. We think that she may have had an undiagnosed urogenital sinus that could have caused the symptoms. Another patient had undergone right nephrectomy in the neonatal period because she had congenital dysplasia of the right kidney associated with symptomatic ureteral atresia (fever and abdominal distension), which could be the cause of urinary urgency as opposed to the SCT. The psychological and cosmetic outcomes were somewhat poor. Of all surveyed patients, 62.5% reported psychosocial problems associated with the appearance of the scar, so we must not forget that while the priority of the surgical management of teratoma is to achieve full resection, the attention and care devoted to the wound should be exquisite, since cosmetic outcomes, as we have seen, have a significant impact in the social life of patients.

There are limitations to this study, some of which involve the intrinsic weaknesses of retrospective studies, the lack of a control group and the small sample size, which poses a challenge to the generalisation of its conclusions, as well as the limitations intrinsic to telephone surveys, which are not carried out face-to-face and may lead to biases, such as collection of inaccurate information.

Conflicts of interest

The authors have no conflicts of interest to declare.

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