

# Postoperative Bowel and Urinary Dysfunction of Sacrococcygeal Teratoma

Salar Sabah Perdawd  (FIBMS)<sup>1</sup>

<sup>1</sup> Department of Surgery, College of Medicine, Hawler Medical University, Erbil, Iraq

## Abstract

**Background:** Teratomas are composed of multiple tissues foreign to the organ or site from which they arise. Although teratomas are sometimes defined as having three embryonic layers (endoderm, mesoderm, and ectoderm), and are generally divided into gonadal and extragonadal types. The most common extragonadal type being sacrococcygeal teratomas which constitute the most prevalent childhood germ-cell cancers

**Objective:** To assess postoperative bowel and urinary dysfunction in babies with sacrococcygeal teratomas

**Patients and Methods:** 23 patients having sacrococcygeal teratomas were received at the time between May 2014 to September 2022 at the department of pediatric surgery in Raparen Teaching Hospital in Erbil city. All recorded data from case note, operation notes together with demographic data and operative details obtained and recorded. The follow-up time span was between three months to seven years.

**Results:** The age range was from 1 day to 4 year, (median 3 days), Presenting features include posterior sacral mass in 17 cases (73.9%), pelviabdominal mass in 5 cases (21.7%), urinary retention in 4 cases (17.4%), palpable rectal mass in 2 cases 98.7%), intestinal obstruction in 1 case (4.3%), and constipation in 4 cases (17.4%). Eight cases (34.8%) were type I, 6 cases (26.1%) type II and III, and 3 cases (13%) were type IV. postoperative complications were wound infection in 4 patients (17.4%), constipation in 5 cases (21.7%), 1 (4.3%) ugly scar arranged for aesthetic operation, fistula 1(4.3%) case, recurrence in 1(4.3%).

**Conclusion:** Patients with sacrococcygeal teratoma were more likely to experience uncontrolled urination, difficulties emptying the bladder, pyelonephritis, and constipation than healthy children. Children with big and immature teratomas had a higher prevalence of dysfunctional outcomes.

**Keywords:** Teratoma, bowel dysfunction, urinary dysfunction.

## OPEN ACCESS

**Correspondence Address:** Salar Sabah Perdawd

Department of Surgery, College of Medicine, Hawler Medical University, Erbil, Iraq

**Email:** [salar.sabah@hmu.edu.krd](mailto:salar.sabah@hmu.edu.krd)

**Copyright:** ©Authors, 2023, College of Medicine, University of Diyala. This is an open access article under the CC BY 4.0 license

(<http://creativecommons.org/licenses/by/4.0/>)

**Website:**

<https://djm.uodiyala.edu.iq/index.php/djm>

**Received:** 30 January 2023

**Accepted:** 23 February 2023

**Published:** 30 October 2023

## Introduction

Teratomas are composed of multiple tissues foreign to the organ or site from which they arise. Although teratomas are sometimes defined as having three embryonic layers (endoderm, mesoderm, and ectoderm), and are generally divided into gonadal and

extragonadal types, the most common extragonadal type being sacrococcygeal teratomas (SCT) which are the most prevalent childhood germ cell cancers (40%) [1]. Sacrococcygeal teratomas divided into benign (mature) and malignant (immature),

The head and neck, brain, gonadal, chest, retroperitoneum, vagina, stomach, and pineal region are less frequent locations [2,3].

It is crucial that practitioners are aware of this condition, which would have prevented complications in this series. A excellent prognosis is linked to early diagnosis and full excision with removal of the coccyx. During excision, tumor leakage is a factor in recurrence. Constipation and fecal soiling are persistent lower gastrointestinal issues that are associated with Altman's tumor classification. [4]. Even though SCT is mostly benign, caregivers must deal with issues like recurrence, malignant changes in patients who present later, and long-term functional consequences. To describe this illness in underdeveloped nations and evaluate the long-term functional effects on survivors, a multi-center study may be required [5,6]. The extent of sacrococcygeal teratomas is classified by the American Academy of Pediatrics' surgical section as type I when it is primarily externally located with little presacral component, type II when it is externally present but has significant intrapelvic extension, type III when it is apparent externally but primarily a pelvic mass extending into the abdomen, and type IV when it is presacral with no external presentation [7]. Anorectal and genital anomalies are of particular concern among these congenital abnormalities, which are linked to between 5% and 26% of sacrococcygeal teratoma instances. The most common anorectal malformations being Currarino triad ,Urogenital anomalies including hypospadias, vasicoureteric reflux, patent urachus, undescended testicles and duplication of vagina or uterus [8,9]. The

preferred treatment for SCT is complete surgical excision performed early in the neonatal period. Complete tumor excision, coccyx removal, pelvic floor reconstruction, anorectal sphincter reconstruction, and cosmetic improvement are the objectives of surgery for an SCT. [10,11]. SCT frequently causes long-term issues, with 10% developing neuropathic bladders or minor bowel dysfunction (constipation or incontinence) and 40% possibly experiencing mild bowel dysfunction [12]. Aim of study is designed to assess postoperative bowel and urinary dysfunction in babies with SCT .

### **Patients and Methods**

Twenty three patients having SCT were received during the period from May 2014 to September 2022 at the department of pediatric surgery in Raparen Teaching Hospital in Arbil city . All recorded data were obtained from case notes, operation notes, discharge summary sheets, and the parents or the caregivers after agreement and obtained informed consent. A data chart was designed to collect demographic data including name of the patient, age, sex, , file number, maternal history, antenatal history, family history, Age at presentation, clinical features, associated anomalies, preoperative laboratory and radiological studies, operative details including abdominal, posterior sagittal, or combined approach, type and nature of teratoma, completeness of resection, tumor rupture, and bleeding, histopathology report, early and late complications , recurrence , cosmetic appearance and functional results. Postoperative bowel and urinary tract function together with sensory-motor deficit are assessed both clinically and

radiologically. The follow up period ranged between 3 months to 7 years. The patients were periodically followed by digital rectal examination, serum level of AFP and US, CT scan or MRI accordingly.

### Statistical Analysis

Data were analyzed using the statistical package for social science (SPSS version 19). Descriptive statistic was carried out to calculate the frequencies and percentages.

### Results

The age range was from 1 day to 4 year, (median age was 3 days), 16 cases (69.6%) of them were less than one month and the main presenting symptom was posterior sacral

mass and all of them were benign. Seven (30.4%) cases aged more than one month during presentation with mostly intrapelvic component and malignant histology, 20 cases were female (87%), and 3 cases were male (13%). The ratio of females to males was 6.7:1. six presenting features were observed in this study; posterior sacral mass in 17 cases (73.9%), pelviabdominal mass in 5 cases (21.7%), urinary retention in 4 cases (17.4%), palpable rectal mass in 2 cases (8.7%), intestinal obstruction in 1 case (4.3%), and constipation in 4 cases (17.4%) as shown in Table (1).

**Table(1):** age , sex and presenting feature

Demographic data		No.	%
Age	<1month	16	96.6
	>1month	7	30.4
Sex	Male	3	13
	Female	20	87
Presenting features	Posterior sacral mass	17	73.9
	Abdominopelvic mass	5	21.7
	Urinary retention	4	17.4
	Constipation	4	17.4
	Palpable rectal mass	2	8.7
	Intestinal obstruction	1	4.3

Seven cases (30.4%) had associated congenital anomalies Table (2).

**Table (2):** Associated anomalies with sacrococcygeal Teratoma

Associated anomalies	No.	%
<b>Present</b>	<b>7</b>	
Congenital Heart disease	2	30.4
Congenital Renal anomaly	2	
Mixed anomaly :		
ASD+absent radial bone	1	
ASD+bilateral club feet+bilateral DDH	1	
VSD+polydactyly	1	

According to Altman classification of SCT, 8 cases (34.8%) were type I, 6 cases (26.1%) type II, 6 cases (26.1%) type III, and 3 cases (13%) were type IV. Histopathological

examination revealed mature teratoma in 18 cases (78.3%), immature in 2 cases (8.7%), and malignant in 3 cases (13%), Table (3).

**Table (3): Tumor characteristics**

Tumor characteristics		No.	%
<b>Altman classification</b>	Type I	8	34.8
	Type II	6	26.1
	Type III	6	26.1
	Type IV	3	13.0
<b>Microscopic features</b>	Mature	18	78.3
	Immature	2	8.7
	Malignant	3	13.0
<b>Total</b>		23	100

Sacral approach was accessed in 18 patients (78.3%), abdominal in 2 patients (8.7%), and abdominosacral in 3 patients (13%). Complete resection and coccygectomy were

achieved in all cases. Intraoperative bleeding occurred in 3 cases (13%), tumor rupture and spillage in 6 cases (26.1%), injury to adjacent pelvic organ in 1 case (4.3) Table (4).

**Table (4): Intraoperative complications**

Intraoperative Complications	No.	%
Bleeding	3	13.0
Tumor rupture & spillage	6	26.1
Injury to adjacent organ	1	4.3

Table (5) shows occurrence of postoperative complications which were; wound infection in 4 patients (17.4%), constipation in 5 cases (21.7%), 1 (4.3%) ugly scar arranged for aesthetic operation, fistula 1(4.3%) case.

Recurrence occurred in 1(4.3%) case which was malignant at the first histopathological examination, occurred after 1.3 years after first operation, received radiotherapy, chemotherapy died at 3.6yr.

**Table (5): Postoperative outcome**

Complications	No.	%
Wound infection	4	17.39
Constipation	5	21.73
Urinary fistula	1	4.34
Recurrence	1	4.34
Ugly scar	1	4.34
<b>Total</b>	12	52.17

## Discussion

Management of SCT is mainly surgical. Not only is the prognosis better the sooner the diagnosis, but also, the prognosis is

improved by surgical intervention when it performed early [2,5]. In our study early resection was performed in all cases as soon as the diagnosis confirmed. Coccygectomy

and a thorough surgical removal of the tumor was accomplished in all cases to avoid recurrence as recommended in previous studies [1,12].

The frequency of different congenital abnormalities linked to sacrococcygeal teratomas varies from (5% - 26%) [3,5,13]. In our series, congenital anomalies including cardiac, renal and skeletal were recorded in 7 cases (30.4%).

Many reports highlighted the potential postoperative complications after SCT resection. In this study incidence of constipation was 21.73% while 47% in M Hambraeus *et al*[6] were low gestational age, and was a reliable indicator of both bowel and urinary tract dysfunction, 70% in study done by T Sakurai<sup>9</sup> and 36% in another study done also by M. Hambraeus *et al* in 2019[11]. The difference is due to small number of cases in the current study and small number of patients who presented beyond the age of toilet training at the end of the study. Functional impairment's cause has not been determined with certainty. Tumors with a substantial and adherent intrapelvic component may cause surgical trauma, direct pressure on pelvic nerves and plexuses, and traction caused by compression of surrounding tissues [14].

As symptoms vary in severity and particularly as a function of age, rarely manifesting before toilet training, and take a long time to develop, they are frequently not understood to be related to an operation in the neonatal period by either parents or the patient, and they are also rarely disclosed on routine follow-up because of the fear of stigmatization. Anorectal dysfunction typically only comes into play when it is

contributing to psychological issues. These can be identified by a thorough history that takes into account the patient's age and by a straightforward physical exam that includes digital rectal palpation [14].

Although in this study there was no any case of urinary dysfunction and none of them send for urodynamic study , there is debate about whether asymptomatic patients should undergo technically challenging examinations such rectomanometry and video urodynamic investigations [7,10,12,14]. The possibility of upper urinary tract injury necessitates a particular approach to urologic functional impairment. It seems appropriate to incorporate video urodynamic studies into standard US and uroflometry. In this study, there wasn't any case of urologic dysfunction and this might be due to the same reason mentioned above , while Manal *et al* found 40% incidence of bowel and bladder dysfunction in their study [12] and in another study by K. Masahata *et al* found anorectal dysfunction and urologic dysfunction in (20.7%) and (13.8%), respectively [7]. In the study by M. Hambraeus *et al* in 2018 they found that sacrococcygeal teratoma patients were more likely to report having uncontrolled urination (12%), trouble emptying the bladder (24%), and pyelonephritis (18%).

In the current study, one case female (4.3%) developed recurrence 15 months after primary resection of malignant tumor, presented as a case of urinary retention, elevated serum AFP level. In a study by R. Niramis *et al*, 3 out of 41 individuals with teratomas developed disease recurrence (7.3%)[15] and in ji .Phi *et al* study, tumor recurrence affects about 10 to 15 of patients

[10].Mortality rate was (4.3%), similar in a cohort of patients in Manal et al study , 95.6% of people survived in total. The two fatalities in our series were brought on by a massive lung metastasis and an enhanced disease load [12].

### Conclusions

Patients with sacrococcygeal teratoma were more likely to experience uncontrolled urination, difficulties emptying the bladder, pyelonephritis, and constipation than healthy children. Children with big and immature teratomas had a higher prevalence of dysfunctional outcomes.

### Recommendations

High index of suspicion and early referral to specialized pediatric surgery is important to ensure early proper management and close long term follow up also is mandatory to deal with potential functional post-operative sequelae.

**Source of funding:** The current study was funded by our charges with no any other funding sources elsewhere.

**Ethical clearance:** The local College of Medicine Ethics Committee gave its approval to the study protocol at Hawler Medical University (HMU). Informed consent in writing was obtained from each participating patient.

**Conflict of interest:** Nil

### References

[1]Jean-Martin Laberge, MD , Pramod S. Puligandla, MD , Kenneth Shaw, MD. Teratomas, Dermoids, and other Soft tissue tumors. *Aschcraft's principles and practice of Pediatric surgery* 2020;67:1066-90.  
[2]Adeline Salima, Arimatias Raitiob, Paul D.Losty. Long-term functional outcomes of

sacrococcygeal teratoma – A systematic review of published studies exploring ‘real world’ outcomes. *European Journal of Surgical Oncology* ;Volume 49, Issue 1, January 2023, Pages 16-20.

[3]Helene Flageole, Sacrococcygeal teratoma, *Fundamentals of Pediatric Surgery*2011;95:735-40.

[4]Hussam S. Hassan and Akram M. Elbatarny: Sacrococcygeal teratoma: management and outcomes, *Annals of Pediatric Surgery* 2014, 10:72–77.

[5]Lohfa B.Chirdan, Aba F. Uba, Sunday D.Pam. Stephen T. EEdino, Barnabas M. Mandong, Oluwabunmi O. Chirdan, Sacrococcygeal teratoma: Clinical characteristics and long-term outcome in Nigerian children, *Annals of African Medicine*, Vol. 8, No. 2; 2009:105-109.

[6]Mette Hambraeus, Lars Hagander, Pernilla Stenström,Einar Arnbjörnsson, Anna Börjesson, Long-Term Outcome of Sacrococcygeal Teratoma: A Controlled Cohort Study of Urinary Tract and Bowel Dysfunction and Predictors of Poor outcome. Published:April 12, 2018. *jpeds*.2018.02.031.

[7]Kazunori Masahata, Chihiro Ichikawa, Katsutoshi Makino, Takatoshi Abe, Kiyokazu Kim, Taku Yamamichi, Ai Tayama, Hideki Soh, Noriaki Usui. Long-term functional outcome of sacrococcygeal teratoma after resection in neonates and infants: a single-center experience. *Pediatr Surg Int* .2020 Nov;36(11):1327-1332.

[8]Sarah Braungart, Eleanor Ca James, Mark Powis, Hany Gabra, CCLG Surgeons Collaborators, Paul D. Losty. Sacrococcygeal teratoma: Long-term outcomes. A UK CCLG Surgeons Group Nationwide Study. *Wiley Online Library*; published: 13 October 2022.



<https://doi.org/10.1002/pbc.29994>.

[9]Tsuoyoshi Sakurai, Masatoshi Hashimoto, Megumi Nakamura & Naobumi Endo. Differences in postoperative complications and prognosis of sacrococcygeal teratoma and presacral tumors in Currarino syndrome. *Annals of Pediatric Surgery* volume 18, Article number: 19 ,March 2022.

[10]Ji Hoon Phi. Sacrococcygeal Teratoma : A Tumor at the Center of Embryogenesis. *J Korean Neurosurg Soc* 64 (3) : 406-413, 2021

<https://doi.org/10.3340/jkns.2021.0015>

[11]Mette Hembraeus, Ammar Al-Mashhadi, Tomas Wester, Pär Johan Svensson, Pernilla Stenström, Helene Engstrand Lilja. Functional outcome and health-related quality of life in patients with sacrococcygeal teratoma . *Journal of Pediatric Surgery* 2019; Volume 54, Issue 8, Pages 1638-1643.

[12]Manal Dhaiban, Kate Cross, Stefano Giuliani. 109 Sacrococcygeal teratoma: long-term outcome in paediatric population. *BMJ Journals*, December 2021 - Volume 106 - Supplement 3.

<http://dx.doi.org/10.1136/archdischild-2021-gosh.109>.

[13]Hira Ahmad, MD; Devin R. Halleran, MD; Jon Vardanyan, n/a; Youn Ju Shin, n/a; Alessandra C. Gasior, MD; Ihab Halaweish, MD; Mark Ranalli, MD; Marc A. Levitt, MD; Richard J. Wood, MBChB, FCPS(SA); Jennifer H. Aldrink, MD. Functional Fecal and Urinary Outcomes after Sacrococcygeal Tumor Resection in Pediatric Patients. *Pediatrics* (2021) 147 (3\_MeetingAbstract): 907–908.

<https://doi.org/10.1542/peds.147.3MA9.907>.

[14] Vanessa do nascimento santos, Simone de oliveira coelho tcbc-rj, Alan araujo Vieira. Sacrococcygeal teratoma: evaluation of its approach, treatment and follow-up in two reference children cancer centers in Brazil / Rio de Janeiro. *Rev. Col. Bras. Cir.* 49 • 2022  
<https://doi.org/10.1590/0100-6991e-20223341-en>.

[15]Rangsan Niramis, Maitree Anuntkosol, Veera Buranakitjaroen, Achariya Tongsin, Varaporn Mahatharadol, Wannisa Poocharoen, Suranetr La-orwong, and Kulsiri Tiansri. Long-Term Outcomes of Sacrococcygeal Germ Cell Tumors in Infancy and Childhood. *Surgery Research and Practice*/ 4 Oct 2015.

## الخلل المعوي والخلل البولي بعد العملية الجراحية من الورم المسخي العجزي

### العصصي

سالار صباح بيردادود<sup>1</sup>

### الملخص

**خلفية الدراسة:** تتكون التراتومة من أنسجة متعددة غريبة عن العضو أو الموقع الذي نشأت منه. على الرغم من أن التراتومة تُعرّف أحياناً بأنها تحتوي على ثلاث طبقات جنينية (الأديم الباطن ، والأديم المتوسط ، والأديم الظاهر) ، وتنقسم عموماً إلى أنواع غدد تناسلية وخارجية. النوع الأكثر شيوعاً خارج الأطوار هو الأورام المسخية العجزي العصصية التي تشكل أكثر سرطانات الخلايا الجرثومية انتشاراً في مرحلة الطفولة.

**اهداف الدراسة:** لتقييم خلل الأمعاء بعد العملية الجراحية والخلل البولي عند الأطفال المصابين بالورم المسخي العجزي العصصي.

**المرضى والطرائق:** تم استقبال 23 مريضاً يعانون من ورم مسخي في العجز العصصي في ذلك الوقت بين مايو 2014 إلى سبتمبر 2022 في قسم جراحة الأطفال في مستشفى رابارين التعليمي في مدينة أربيل. تم الحصول على جميع البيانات المسجلة من مذكرة الحالة ، وملاحظات العمليات ، إلى جانب البيانات الديموغرافية وتفاصيل المنطوق ، وتسجيلها. تراوحت فترة المتابعة بين ثلاثة أشهر وسبع سنوات.

**النتائج:** تراوحت الفئة العمرية من يوم واحد إلى 4 سنوات (متوسط 3 أيام) ، وتشمل ميزات العرض الكتلة العجزية الخلفية في 17 حالة (73.9٪) ، وكتلة الحوض في 5 حالات (21.7٪) ، واحتباس البول في 4 حالات (17.4٪) ، كتلة مستقيمة محسوسة في حالتين 98.7٪ ، انسداد معوي في حالة واحدة (4.3٪) ، إمساك في 4 حالات (17.4٪). ثمان حالات (34.8٪) كانت من النوع الأول ، و 6 حالات (26.1٪) من النوع الثاني والثالث ، و 3 حالات (13٪) كانت من النوع الرابع. مضاعفات ما بعد الجراحة هي إصابة الجرح في 4 مرضى (17.4٪) ، الإمساك في 5 حالات (21.7٪) ، 1 (4.3٪) ندبة قبيحة مرتبة لعملية تجميل ، ناسور 1 (4.3٪) حالة ، تكرار في حالة واحدة (4.3٪).

**الاستنتاجات:** كان المرضى الذين يعانون من الورم المسخي العجزي العصصي أكثر عرضة للتبول غير المنضبط ، وصعوبات إفراغ المثانة ، والتهاب الحويضة والكلية ، والإمساك من الأطفال الأصحاء. كان لدى الأطفال المصابين بالورم المسخي الكبير وغير الناضج معدل انتشار أعلى للنتائج المختلفة.

**الكلمات المفتاحية:** ورم مسخي ، ضعف الأمعاء ، ضعف المسالك البولية

البريد الإلكتروني: [salar.sabah@hmu.edu.krd](mailto:salar.sabah@hmu.edu.krd)

تاريخ استلام البحث: 30 كانون الثاني 2023

تاريخ قبول البحث: 23 شباط 2023

<sup>1</sup> كلية الطب - جامعة هولير الطبية - أربيل - العراق