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Review Article

Impact of Altman Classification on Fecal continence following Sacrococcygeal Teratoma Resection

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Abstract

Background: Sacrococcygeal teratoma (SCT) is a type of tumor that develops at the base of the coccyx through indefinite etiologies suggested by many embryological theories. The aim of this work was to evaluate outcomes of fecal continence after excision of SCT.

Methods: This retrospective study was carried out on 20 patients, aged more than 3 years old, males and females, operated for SCT in the last 10 years in Sohag University Hospitals. Fecal continence is assessed using Rintala's continence score.

Results: Pathological types were mature benign in 80% patients, immature teratoma in 15% patients, malignant teratoma in 5% patients. Wound infection occurred in 10% patients, bleeding in 10% patients, reoperation in 5% patients and neurogenic bladder in 5% patients. The median of Rintala continence score was 18(16 -19).

Conclusions: There are favorable outcomes of fecal continence following the excision of SCTs. Most patients achieved high continence scores. Rintala's continence score was significantly lower in type III and type IV than in type I and II (P value=0.012).

Keywords: Fecal Continence, Sacrococcygeal Teratoma, Altman's Classification

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Introduction:

Sacrococcygeal teratoma (SCT) is one of the most common neoplasms presenting in the neonatal period. The incidence is about 1:35000 live births. Females are four times more affected than males [1].

It is an extra-gonadal germ cell tumor that contains all 3 embryonic tissue components. According to Altman's classification there are 4 types of the mass depending on the external, pelvic or intra-abdominal extension .[2]

In many cases it is diagnosed by routine prenatal ultrasound examination which necessitates cesarean delivery in a specialized center with qualified pediatric surgeons. The definitive treatment relies on complete excision of the mass together with the coccyx. This can be achieved through external approach via classic chevron or posterior sagittal incisions. An abdomino-sacro-perineal approach however is necessary in masses with significant intra-abdominal extension .[3]

Histopathology is benign in most of the lesions whether mature or immature teratomas, however a small proportion of cases may demonstrate embryonic malignant component.

Complications may arise during the prenatal period due to the increasing size of the tumor including rupture of the tumor, compression on the umbilical vein or high cardiac output heart failure with hydrops fetalis. Complications during surgery may arise due to close proximity of the tumor to the rectum and urinary tract or bleeding from a large feeding vessel arising from median sacral artery. Postoperative complications range from infection causing gapped wound or complete dehiscence requiring repair or deranged healing causing unsightly scar up to long term functional problems such as fecal or urinary incontinence. (1–3) . Salim et al. in a study that reviewed the results of 37 papers published about the subject documented that 19% of patients experienced soiling and 26% constipation.[4]

The aim of this work was to evaluate outcomes of fecal continence after excision of SCT.

Patients and Methods:

This retrospective study was carried out on 20 patients, aged more than 3 years old, both sexes, operated for SCT in the last 10 years in Sohag University Hospitals. The study was done after approval from the Ethical Committee Sohag University Hospitals, Sohag, Egypt. A Consent was obtained from the relatives of the patients.

Exclusion criteria were patients lost follow up before 6 months after surgery and the cases with other associated comorbidities such as Currarino triad.

All patients were subjected to complete history taking and examination.

Many scoring systems for fecal incontinence were developed. The Rintala scoring system.[5] was the most extensive, incorporating both the questions related to "social problems" and the contents of the other classifications such as Kricknebeck .[6] and Pena. [7]. Because it somewhat represents the psychosocial issues, it is currently the most extensively utilized.

Bowel function was evaluated using the Bowel Function Score (BFS) described by Rintala which consists of 7 questions about the ability to hold back defecation, the feel of urge to defecate, the frequency of defecation, soiling, accidents, constipation and social problems.

All questions are rated 0-3 except the frequency of defecation 0-2, and the final score ranges from (0-20).

Statistical analysis

Statistical analysis was conducted using SPSS version 26 (IBM Inc., Chicago, IL, USA). Quantitative parametric data were expressed as mean and standard deviation (SD). Quantitative non-parametric data were reported as median and interquartile range (IQR). Qualitative factors were expressed as frequency and percentage (%).

Results:

Demographic data, associated anomalies and Altman classification were enumerated in this table. **Table 1**

Table 1: Demographic data, associated anomalies and Altman classification of the studied patients

		N=20
Age (years)		6.4 ± 2.18
Sex	**Male**	5 (25%)
	Female	15 (75%)
Age at diagnosis	**Antenatal**	9 (45%)
	At birth	10 (50%)
	Late presentation	1 (5%)
Associated anomalies	**Congenital heart diseases**	3 (15%)
	Renal anomalies	1 (5%)
Altman classification	**I**	9 (45%)
	II	8 (40%)
	III	2 (10%)
	IV	1 (5%)

Data are presented as mean ± SD or frequency (%).

Surgical procedure, type of perineal incision, preoperative imaging and pathological types of the studied patients were enumerated in this table. Table 2

Table 2: Surgical procedure, type of perineal incision, preoperative imaging and pathological types of the studied patients

		N=21
Surgical procedure	**Perineal approach**	17 (85%)
	Abdominoperineal approach	3 (15%)
Type of perineal incision	**Vertical elliptical with midline closure**	11 (55%)
	Classic chevron with transverse closure	9 (45%)
Pathological types	**Mature benign**	16 (80%)
	Immature teratoma	3 (15%)
	Malignant teratoma	1(5%)

Data are presented as frequency (%). MRI: Magnetic resonance imaging, CT: computed tomography.

Postoperative complications, Rintala's continence score and quality of life of the studied patients were enumerated in this table. **Table 3**

Table 3: Postoperative complications, Rintala's continence score and quality of life of the studied patients

		N = 21
Postoperative complications	**Wound infection**	**2 (10%)**
	Bleeding	2 (10%)
	Reoperation	1 (5%)
	Urinary incontinence	1 (5%)
	Rintala's continence score	18(16-19)

Data are presented as median (IQR) or frequency (%).

Significant wound infection and gapping occurred in 2 (10%) patients which improved by administration of systemic and local antibiotics, one of those cases is an Altman type 3 operated by combined abdomino-perineal approach, Rintala's score is 12 and constipation improved by diet modification, The second case is an Altman type 2 male operated through vertical incision, however, follow up revealed excellent continence (Rintala's score 18).

Significant intra-operative bleeding necessitating blood transfusion occurred in 2 (10%) patients, one case was a female with Altman type 1, the mass had increased vascularity but bleeding was controlled after ligation of the median sacral artery, histopathology revealed immature teratoma, (Rintala's score 20), the other case is also a female with type 1 but the pathology was mature teratoma, favorable outcomes has been reached in relation to continence (Rintala's score 18) Reoperation in 1 (5%) patient was inevitable due to local recurrence, the male child presented with Altman type 1 SCT which was removed through vertical perineal approach, the histopathology of the mass revealed a mature cystic teratoma, recurrence of the tumor was discovered by routine ultrasound and AFP screening, The second operation was done through the same approach but the pathology revealed a yolk sac tumor (Rintala's score 20).

Urge incontinence in 1 (5%) patient due to over-active bladder was confirmed by detrusor muscle overactivity and reduced maximum cystometric capacity in urodynamic studies and was treated by anticholinergic drugs, the female child had an Altman type 4 SCT which was removed by a combined abdomino-perineal approach, good continence was found in follow up (Rinata's score was 16).

Regarding follow- up, 21 (100%) patients followed-up regularly after surgery. The duration of follow up ranged from 6 to 12 months with a mean value (± SD) of 8.9 (±2.21) months.

Table 4 Results of Rintala's score of the studied cases in relation to their Altman type, sex and surgical approach

	Number of cases	Range of Rinatala's score	Mean of Rintala's score	Standard deviation
Type 1	9	3(17-20)	19	0.94
Type 2	8	4(14-18)	16.63	1.32
Type 3	2	3(12-15)	13.5	1.5
Type 4	1			Rintala score 16
Type 1 and 2	17	6 (14-20)	17.29	1.89
Type 3 and 4	3	4	14.33	2.05
Male	5	8 (12-20)	17	2.83
Female	15	6 (14-20)	17	1.97
Perineal	17	6(14-20)	17.29	1.89
Abdomino-perineal	3	4	14.33	2.05

Table 5: Relation between Rintala's continence score and Altman classification

		Type I and II (n=17)	Type III and IV (n=3)	P value
Rintala's continence score	**Median**	18	15	**0.012***
	IQR	17 – 19	13.5 - 15.5	

*: Significant as P value≤0.05.U: Mann-Whitney test.

Rintala's continence score was significantly lower type III and type IV than in type I and II (P value=0.012).

Table 6: Relation between Rintala's continence score and sex

		Male (n=5)	Female (n=15)	P value
Rintala's continence Score	**Median**	18	18	1
	IQR	16 – 19	16 - 19	

Rintala's continence score was insignificantly different between both groups.

Discussion:

In the present study, the age ranged from 3 to 11 years with a mean value (± SD) of 6.4 (±2.18) years. The diagnosis was antenatal in 45% patients, at birth in 50% patients and late presentation in 5% patient. There were 25% males and 75% females. Pathological types were mature benign in 80% patients, immature teratoma in 15% patients, malignant teratoma in 5% patient.

The present study indicated that the median of Rintala continence score was 18 with IQR 16 -19. This aligns with Elgendy et al. [8], who demonstrated that soiling occurred in just 12 patients (12.6%). Of the 12 patients, seven were treated with oral laxatives. Two received supplementary rectal irrigation in addition to laxatives. The remaining three received additional surgery to address faecal incontinence using an antegrade continence enema (ACE). In total, four individuals had both urine and bowel dysfunction, all classified as Altman III/IV neoplasms.

In contrast, Derikx et al. [9] demonstrated that among patients treated for SCT, 46% reported

impaired bowel function and/or urinary incontinence, with 9% experiencing involuntary bowel movements, 13% reporting soiling, 17% suffering from constipation, and 31% experiencing urinary incontinence.

Limitations of the study included that the sample size was relatively small. The study was in a single center. Lack of comparison with a control group or alternative surgical techniques restricts the ability to assess relative effectiveness.

Conclusion:

There are favorable outcomes of fecal continence following the excision of SCTs. Most patients achieved high continence scores. Rintala's continence score was significantly lower type III and type IV than in type I and II (P value=0.012).

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