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Functional outcome and health-related quality of life in patients with sacrococcygeal teratoma – a Swedish multicenter study

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ABSTRACT

Background/Purpose: The aims of this study were to evaluate bowel and urinary tract function, to identify predictors for dysfunctional outcome and to evaluate health-related quality-of life (QoL) in patients treated for sacrococcygeal teratomas (SCT).
Methods: Medical records of patients with SCT born between 1985 and 2015 treated at three Swedish pediatric surgical centers were reviewed. Questionnaires regarding urinary tract function, bowel function and QoL were sent to patients and parents. Different QoL instruments were used for the different age groups.
Results: **T**otally 85 patients were identified. Four patients died in the neonatal period. Forty-nine patients answered the questionnaires (60%). Median age at follow-up was 8.9 years (range 3.6–28.8). Bowel dysfunction was reported by 36% and urinary tract dysfunction by 46% of the patients. Univariate analysis revealed that urinary tract dysfunction correlated with gestational age ($p = 0.018$) and immature histology ($p = 0.008$), and bowel dysfunction correlated with gestational age ($p = 0.016$) and tumor size ($p = 0.042$). Low gestational age was an independent predictor for both urinary tract and bowel dysfunction. Good or very good QoL was reported by 56% of children aged 4–7 years, 90% of children aged 8–17 years and 67% of the adults.
Conclusion: Although a considerable proportion of bowel and urinary tract dysfunction was found, the reported QoL was good in a majority of the patients with SCT. Low gestational age was found to be a predictor for bowel- and urinary tract dysfunction.
Level of Evidence: Level III.

Sacrococcygeal teratomas (SCT) are rare congenital tumors with a birth prevalence of 1 in 14,000–28,000, and with a female predominance of 4:1 [1–5]. SCT are germ cell tumors and the majority of tumors are histologically mature [6,7]. The Altman classification describes the extension of the teratoma, ranging from predominantly external (Altman I), to entirely internal localization (Altman IV) [1]. Due to the internal localization, diagnosis of Altman IV tumors is often delayed, which leads to a higher risk of malignant transformation [8]. With increasing sonographic sensitivity, the prenatal detection rate of SCT has increased to more than 50% in several series [5,9]. This accentuates the need for knowledge of mortality and morbidity when counseling parents expecting a child with SCT.

Several authors have reported a good overall perinatal survival rate [3,5,7,9–17], but rapidly growing and highly vascularized SCT remain a challenge with mortality rates up to 60% [10,12,13,14–18]. Reports on long-term functional outcome, with respect to bowel and urinary tract function, are scanty, heterogeneous and often based on single-center experience [19–26]. In addition, only a few studies have evaluated Quality of Life (QoL) [21,27].

The aims of this study were to evaluate bowel and urinary tract function, to identify predictors for dysfunctional outcome and to evaluate QoL in a multicenter-based cohort of patients treated for SCT.

1. Patients and methods

Patients with SCT treated at three Swedish centers of pediatric surgery from January 1985 to December 2015 were identified in this cross-sectional observational study. In total 16 surgeons had performed the operations over the years. Patients with a presacral teratoma as part of the Currarino triad were excluded. All data were pseudonymized prior to analysis. Part of the children from one center were previously reported on in a follow-up at another point of time in another methodological setting [28].

Medical records were reviewed for the following information: gender, gestational age at birth, birth weight, age at diagnosis, Altman

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classification, age at surgery, tumor size, tumor histopathology, resection margins, perioperative and postoperative complications with respect to recurrence, age at recurrence, mortality, cause of death and age at follow-up.

Questionnaires were sent to patients and parents and written consent was obtained. Bowel function was evaluated using the Bowel Function Score (BFS) described by Rintala et al. [29]. Bowel dysfunction was defined as inability to withhold defecation, soiling, fecal accidents, and, or constipation.

Urinary tract function was evaluated according to the definitions made by the International Children Continence Society and a related questionnaire, which has been used in previous studies on urinary tract functional outcome [30]. Urinary tract dysfunction was defined as previous urinary tract infection (UTI), urgency, incontinence, and, or bladder emptying problems.

The DISABKIDS chronic generic instrument was used to assess the QoL of children aged 8–17 years and the DISABKIDS Smiley instrument was used in children aged 4 to 7 years [31,32]. Corresponding proxy versions were available for parents. The questionnaires cover mental, social and physical domains of QoL. The DISABKIDS Smiley instrument enables young children to answer questions by selecting 'smileys' to rate their QoL. SF-36 and Gastrointestinal Quality of Life Index (GIQLI) questionnaires validated for the Swedish population were used for patients aged 18 years or older [33,34]. Both questionnaires consist of 36 questions and the answers are categorized in the same QoL domains as the DISABKIDS instruments. The results from the QoL questionnaires were expressed as scores, where a higher score implies a better functioning level.

1.1. Ethics

The study protocol was approved by the Regional Ethics Review Board (registration number 2016/147 and 2016/289).

1.2. Statistics

In order to evaluate factors related to the risk of developing bowel or urinary tract dysfunction, the statistical analyses were performed in three steps (for each outcome). All pre-defined risk factors were tested univariately. The univariable analyses were primarily a screening procedure to reduce the number of variables included into the multivariable analysis. However, we did not want to be too strict and exclude variables that potentially could have an impact in the presence of other variables in the multivariable analysis. With that in mind we considered a p-value of 0.2 as a reasonable cut-off to include a variable in the multivariable analysis. Finally the optimal model was selected using stepwise selection process where the optimal model was determined as the one with the lowest AIC value (Akaike information criterion). p-Values <0.05 were considered statistically significant. The analyses were performed using R version 3.4.0 (R Core Team (2017). R: A language and environment for statistical computing. R Foundation for Statistical Computing, Vienna, Austria. URL (https://www.R-project.org/). Correlation analysis between gestational age and tumor size was performed using IBM SPSS Statistics, version 24.0, Armonk, NY; IBM Corp.

Table 1
Patient and tumor characteristics in 53 patients with sacrococcygeal teratoma.

Gestational age at birth (weeks)	38 [28–41]
Premature birth (<37 weeks)	16 (30%)
GA <30 weeks at birth	5 (9%)
Birth weight (g)	3550 [1200–3550]
Female:male-ratio	4.9:1
Age at surgery (days)	5 [0–1488]
>6 months at surgery	9 (17%)
Tumor size (cm)	7.5 [2–21]
Altman	
I	23 (43%)
II	11 (21%)
III	11 (21%)
IV	8 (15%)
Histology	
Mature	38 (72%)
Immature	14 (26%)
Malignant	1 (2%)
Radicality	
Yes	33 (62%)
No	13 (25%)
Unsure	7 (13%)
Mortality	4 (8%)
Recurrence/residual teratoma (n = 49)	8 (16%)

n = number (%) or median (range).

Table 2
Bowel function in patients with sacrococcygeal teratoma.

Age	8.9 [3.6–28]
Constipation treatment	9 (20%)
(respondents n = 46)	
Diet treatment	1 (2%)
Laxatives	7 (15%)
Enema treatment	2 (4%)
Ability to hold back defecation	
(respondents n = 46)	
Always	36 (78%)
Problems <1 time/week	6 (13%)
Problems >1 time/week	2 (4%)
Never	2 (4%)
Defecation frequency	
(respondents n = 47)	
Twice per day to every other day	35 (74%)
Less than every other day	8 (17%)
More than twice a day	4 (9%)
Soiling	
(respondents n = 47)	
Never	36 (77%)
< 1 time per week	8 (17%)
Frequently/daily, change of underwear	2 (4%)
Always, protective aids	1 (2%)
Fecal accidents	
(respondents n = 41)	
Never	37 (90%)
<1 time per week	3 (7%)
>1 time per week	0 (0%)
Daily, protective aids	1 (3%)
Social problems due to bowel issues	
(respondents n = 47)	
No	44 (94%)
Occasionally	3 (6%)
Problems limiting activities	0%
Serious social and psychiatric problems	0%

n = number (%).

2. Results

During the study period, a total of 85 children were treated for SCT, of whom 49 children and young adults agreed to participate in the study

Table 3.1
Univariate logistic regressions for bowel dysfunction.

	Odds ratio	95% CI	p-Value
Gestational age	0.76	(0.60–0.95)	0.016
Gender	0.50	(0.11–2.33)	0.377
Tumor size	1.15	(1.00–1.32)	0.042
Histopathology	0.46	(0.12–1.75)	0.253
Altman (III-IV vs I-II)	0.71	(0.21–2.44)	0.590
Age at surgery, 2-4d vs 0-1d	0.80	(0.18–3.46)	0.765
Age at surgery, 15-183d vs 0-1d	0.80	(0.13–4.74)	0.806
Age at surgery, >183d vs 0-1d	0.38	(0.06–2.53)	0.318
Recurrence	1.07	(0.22–5.17)	0.932
Age at follow-up	0.99	(0.92–1.08)	0.871

Table 3.2
Multiple logistic regression for bowel dysfunction including variables with an independent p-value <0.2.

	Coefficient	95% CI	p-Value
Gestational age	0.74	(0.55–0.99)	0.043
Tumor size	0.98	(0.81–1.19)	0.815

(response rate 60%). Patient and tumor characteristics are shown in Table 1.

All deaths (n = 4, 8%) were caused by perioperative bleeding in prematurely born infants (GA 28–30 weeks) with large Altman I tumors (15–21 cm in diameter). A significant correlation was found between gestational age and tumor size ($p < 0.001$).

2.1. Bowel symptoms

Questions regarding bowel function were completed by 47 patients. The reported bowel function is shown in Table 2. A total of 36% of the patients reported bowel dysfunction, and 19% reported weekly and, or daily problems. Constipation was reported by 20% of the patients, the majority of whom were treated with laxatives. Two patients needed enema for bowel emptying and 4% reported no voluntary control of defecation. Any degree of soiling was reported by 23% of the patients, the majority of whom had problems less than once a week. Daily or weekly fecal accidents were reported by 10%.

Risk factor analysis for bowel dysfunction is presented in Tables 3.1-3.3. Low gestational age was found to be an independent risk factor and a predictor for bowel dysfunction.

2.2. Urinary tract symptoms

Of 47 respondents, 53% reported a completely normal urinary tract function while 47% reported at least one urinary tract symptom, including previous UTI, urinary urgency, stress incontinence, leakage without stress and, or necessity to strain (Table 4). Weekly, and or daily problems were reported by 28% of the patients. Social problems due to urinary tract dysfunction or urinary tract infections was reported by 10%, and the most common cause of social problems was daytime leakage. Only two patients reported social issues due to nighttime leakage. Risk factor analysis for urinary tract dysfunction is presented in Tables 5.1–5.3. Low gestational age turned out to be an independent risk factor and predictor for urinary tract dysfunction.

2.3. QoL

The DISABKIDS Smiley questionnaire was completed by 16 children aged 4–7 years (Fig. 1). Good or very good QoL was reported by 56%. Sixty-nine percent of parents of the children reported good or very good QoL. Twenty children aged 8–17 years returned the DISABKIDS questionnaire (Fig. 2). A total of 90% reported a good or very good QoL and the vast majority of the children reported good or very good outcome in mental, social and physical domains. No child reported severely reduced QoL. Parents of these children and adolescents reported proxy results similar to their offspring.

The SF-36 and GIQLI questionnaires were returned by 7 of 9 adult patients (Fig. 3). One of the respondents had late effects after chemotherapeutic treatment of yolk sac tumor, and another had a mild cerebral paresis. All respondents reported good or very good physical outcome and good or very good outcome in the pain-scale, but a majority of the patients reported reduced scores concerning energy/fatigue, emotional well-being, social functioning and general health. In GIQLI the median score was 114 (range 91–133), and 33% scored <105, which corresponds to a reduced gastrointestinal-related QoL.

Table 3.3
Multiple stepwise logistic regression for bowel dysfunction.

	Coefficient	95% CI	p-Value
Gestational age	0.76	(0.60–0.95)	0.016

3. Discussion

In this multicenter study of SCT, bowel dysfunction was found in 36% and urinary tract dysfunction in 46% of the patients. Low gestational age was found to be a predictor for urinary tract- and bowel dysfunction. QoL-scores were high in school-aged children, whereas pre-school aged children reported reduced scores. Adult SCT-patients reported lower QoL with regard to emotional well-being, social functioning and general health.

The mortality rate of 8% in our study is comparable to a Dutch nation-wide study reporting a mortality rate of 8.8% in 79 patients with SCT treated between 1980 and 2003 [21]. All deaths in our study were caused by perioperative bleeding in prematurely born infants with large Altman type I tumors.

3.1. Bowel and urinary tract function

Comparison with results of other studies is complicated by methodological heterogeneity and great variation in patient numbers and age distribution between study populations. Other authors have reported urinary tract symptoms in 13–55% of patients with SCT [21,22,26,28,35], which is comparable to the 46% with symptoms in our study. In parallel, the reported rate of constipation in patients with SCT, spans a wide spectrum ranging from 8% to 47% [28,36,37]. The constipation rate of 20% in this study is in line with a study of 79 SCT-patients by Derikx et al. showing a constipation-rate of 16.7% [21]. In a study of a population of Swedish children aged 4–15 years that were

Table 4
Urinary function in 47 patients with sacrococcygeal teratoma.

Previous UTI	
Yes	14 (30%)
No	32 (70%)
Frequency	
1–3 times/day	7 (15%)
4–8 times/day	34 (74%)
>8 times/day	5 (11%)
Urinary urgency	
Never	30 (65%)
<1 time/week	9 (20%)
>1 time/week	5 (11%)
Always	2 (4%)
Stress incontinence	
Never	31 (66%)
<1 time/week	7 (15%)
>1 time/week	6 (13%)
Always	3 (6%)
Leakage, no stress	
Never	34 (72%)
<1 time/week	8 (17%)
>1 time/week	4 (9%)
Always	1 (2%)
Needs to strain to void	
Never	35 (76%)
<1 time/week	7 (15%)
>1 time/week	3 (7%)
Always	1 (2%)
Social problems	
None	32 (68%)
None, in spite of incontinence	8 (17%)
Yes, due to leakage day + night	1 (2%)
Yes, due to UTI	1 (2%)
Yes, due to night time leakage	1 (2%)
Yes, due to day time leakage	4 (4%)

UTI = Urinary tract infection. n (%).

Table 5.1
Univariate logistic regressions for urinary tract dysfunction.

	Odds ratio	95% CI	p-Value
Gestational age	0.74	(0.57–0.95)	0.018
Gender	0.86	(0.19–3.93)	0.843
Tumor size	1.11	(0.97–1.26)	0.126
Histopathology	0.10	(0.02–0.55)	0.008
Altman (III-IV vs I-II)	0.73	(0.22–2.35)	0.595
Age at surgery, 2-4d vs 0-1d	1.00	(0.24–4.20)	1.000
Age at surgery, 15-183d vs 0-1d	1.00	(0.18–5.68)	1.000
Age at surgery, >183d vs 0-1d	0.50	(0.09–2.84)	0.434
Recurrence	2.16	(0.45–10.32)	0.336
Age at follow-up	1.00	(0.93–1.08)	0.998

supposed to be healthy, 16% reported constipation [38], similar to the constipation-rate of SCT-patients in the present study. The 22% incidence of soiling in this study is comparable to the results reported by other authors [36,37]. Low gestational age and constipation could influence the urinary tract function, which must be considered also in SCT patients.

3.2. Predictors

A recent single-center study including 17 patients with SCT found that children with large tumors and immature histology were more likely to have constipation, uncontrolled voiding, difficulty in bladder emptying and pyelonephritis [28].

In the present study, univariate analysis revealed that urinary tract dysfunction correlated with gestational age and immature histology, and bowel dysfunction correlated with gestational age and tumor size. The multiple stepwise regression revealed gestational age as a predictive factor for urinary tract and bowel dysfunction. The predictive value did not increase when adding tumor size or immature histology to the multiple stepwise model, even though these variables were found to correlate with dysfunctional outcome in the univariate analysis. This result could be explained by the close relationship between gestational age, tumor size and immature histology. It is also reasonable to assume that other not identified factors are involved.

Surgical excision in very prematurely born infants might be more complicated because of the size and immaturity of anatomical structures. The premature birth rate was high among children with SCT and if taking previous literature on the subject into account [3,18,39], speculating that the premature birth may be caused by large tumor and growth pattern, polyhydramnion, fetal hydrops, premature rupture of membranes or maternal pre-eclampsia.

3.3. QoL

In the present study DISABKIDS chronic generic module was used as there was not any specific questionnaire available for children with SCT and DISABKIDS Smiley was used as it was available from 4 years of age. Our study showed that 44% of the youngest children reported a reduced QoL compared to 10% of children aged 8–17 years. The seven adults reported a good or very good outcome regarding physical function and pain, whereas a majority reported reduced scores in the energy/fatigue, emotional well-being, social functioning and general health subcategories. Two of the seven adult patients had other potential confounders, i.e. late-effects of chemotherapy and cerebral paresis. Thus, our results show a biphasic QoL-outcome in childhood with a decreased QoL in early childhood, followed by a phase without detectable reduction during school-age. The adult phase is difficult to determine due to the limited number of patients. It has previously been suggested that QoL in patients with SCT does not differ from a normal population neither in general QoL (SF36) nor in appearance related distress [27,35]. It has also been suggested that QoL in patients with SCT may be impaired due to urinary tract and bowel dysfunction [21,37] but such correlation was not analyzed in our study.

There could be several reasons to the age-dependent differences found in our study. The QoL-instruments differed between the age groups, which could explain the differences in scoring between children and adults. Also, QoL might reflect the patients´ self-efficacy - how they managed to cope with symptoms in different situations-, which the instruments used in this study did not allow to measure. QoL has been suggested to depend on physical outcome, which might change over time if symptoms change with age. Because of the limited size of the age groups, we did not analyze statistical correlation between physical function and QoL. The study covers a broad age-span, and patients might have been managed differently in childhood and as adults, affecting the outcome.

This study is strengthened by the multicenter design, which enables evaluation of a fairly large sample size considering the low birth prevalence of SCT. Interpretation of the study results are hampered by the response rate of 60%, which entails a risk of selection bias. Due to the lack of ethical approval, dropout analysis was not possible. The retrospective collection of data from charts might imply to recall bias, and the cross-sectional design means analysis of a broad age-span, here without age-specific analysis, although age might be of importance for outcome and could be relevant to study in larger cohorts. Further, the choice of QoL-instruments must be considered in the interpretation of results, because generic and diagnose specific instruments might reveal different outcomes. Measuring outcome with quantitative methods, such as questionnaires, limits the sensitivity, and future studies using qualitative analysis of functional outcome may further elucidate prevalence and important risk factors.

Table 5.2
Multiple logistic regression for urinary tract dysfunction including variables with an independent p-value <0.2.

	Coefficient	95% CI	p-Value
Gestational age	0.80	(0.57–1.13)	0.214
Tumor size	1.02	(0.84–1.25)	0.814
Histopathology	0.42	(0.05–3.45)	0.421

Table 5.3
Multiple stepwise logistic regression for urinary tract dysfunction.

	Coefficient	95% CI	p-Value
Gestational age	0.74	(0.57–0.95)	0.018

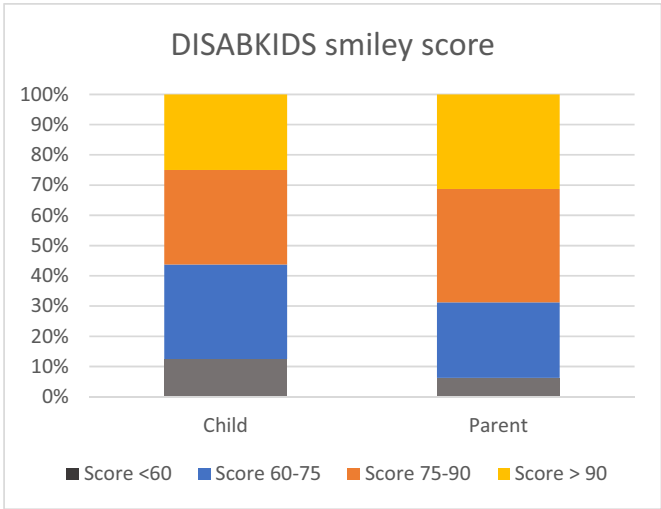

Fig. 1. DISABKIDS smiley scores of 16 children with sacrococcygeal teratoma aged 4–7 years.

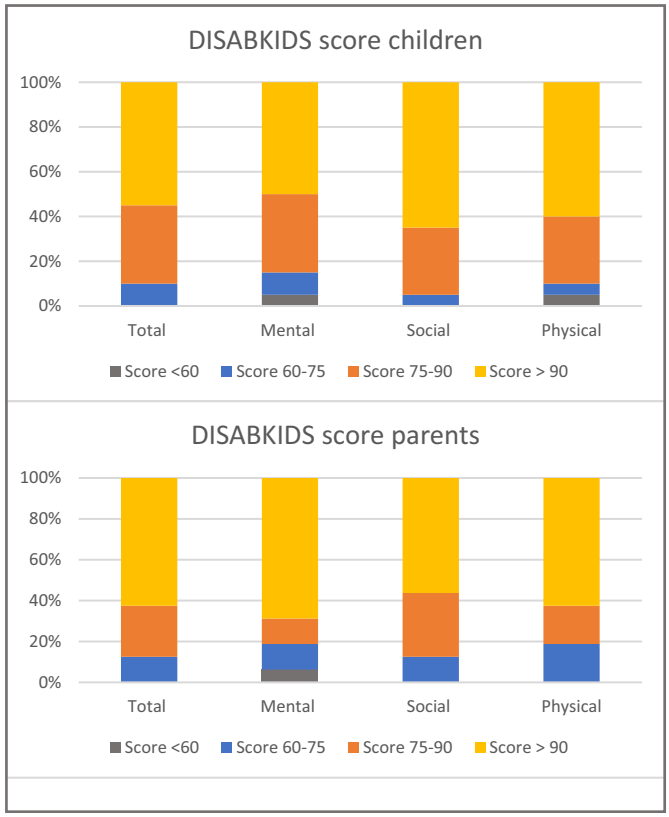

Fig. 2. DISABKIDS of 20 children and adolescents aged 8–17 years with sacrococcygeal teratoma and their parents (n = 16).

The results of the study suggest that patients with SCT need careful follow-up. Impaired bowel and urinary tract function should be appropriately addressed. Premature children need special attention. Prenatal counseling is suggested to include information about possible functional distress, and that treatment and support to good coping mechanisms might be needed.

4. Conclusions

Although a considerable proportion of bowel and urinary tract dysfunction was found in patients with SCT in this study, the reported QoL was good in a majority of the patients. Low gestational age was found to be a predictive factor for adverse urinary tract- and bowel function.

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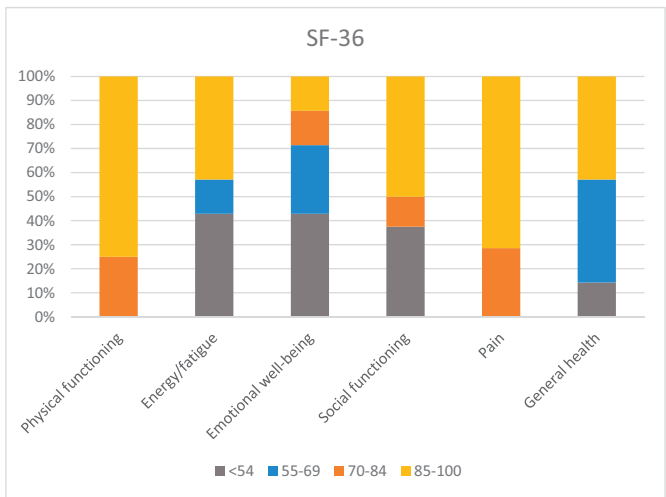

Fig. 3. SF-36 in seven adult patients with sacrococcygeal teratoma.

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