Long-term Functional Outcomes of Sacrococcygeal Teratoma – A Systematic Review of Published Studies Exploring 'Real World' Outcomes

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Abstract

Sacrococcygeal teratoma (SCT) is a rare neoplasm affecting 1:35,000 newborns. Long-term follow-up from small observational studies report impaired bladder/bowel function. This current study comprehensively analyses all published studies to better define true long-term functional sequelae. Medline/Embase databases were searched with PRISMA guidelines. Final analysis yielded 37 studies involving 1,116 patients (854 female; 77%). Individual datasets were available in 14 studies (222 patients). According to Altman classification - 298/845 (35%) were Type I, 252/845 (30%) Type II, 133/845 (16%) Type III, and 128/845 (15%) Type IV tumours. Most neoplasms were benign (640/858; 75%), 77/858 (9%) immature and 141/858 (16%) malignant. Abnormal bladder function was reported in 7/39 (18%) Altman Type I, 23/61 (37.7%) Type II, 11/34 (32.4%) Type III, and 15/25 (60%) Type IV cases (p=0.007). Adverse urological outcomes were notably common in immature/malignant neoplasms vs benign tumours and in patients requiring reoperation(s); p=0.002 and p=0.01. Bowel dysfunction was evident in 19% index cases and constipation in 26% with no significant association(s) with tumour characteristics. Higher Altman stage, unfavourable tumour biology and reoperation are associated with poor functional outcome(s). Multidisciplinary management from primary diagnosis of SCT is crucially important for all patients to best optimise functional outcomes across surgical specialities.

Keywords: sacrococcygeal teratoma; Altman stage system; urinary incontinence; neuropathic bladder; bowel continence; functional outcome

Abbreviations: SCT – sacrococcygeal teratoma

Introduction

Sacrococcygeal teratoma (SCT) is a rare neoplasm affecting 1:35,000 newborns [1-4]. It is the most common germ-cell tumour encountered in newborns and infants. Presentation generally follows two distinct clinical scenarios either as a monstrously large fetal neoplasm detected on antenatal imaging or immediately evident index diagnosis at newborn delivery. Older infants or children may have occult SCT tumour phenotypes (Type IV) which tend to harbour malignancy [5].

The current classification system of SCT is historically based on Altman's survey of the Surgical Section of the American Academy of Pediatrics [6]. The majority of SCT neoplasms are benign and management herein consists of complete surgical resection (R0) including excision of the coccyx [7]. In a small number of index cases, however grossly large vascular tumours may lead to major complications including tumour rupture with haemorrhage or development of high output cardiac failure with fetal hydrops necessitating fetal intervention or emergent surgery at birth [8]. Primary surgical excision may not always be possible in SCTs, particularly those affecting older infants or children due to malignant tumours invading surrounding structures. In these cases, neoadjuvant chemotherapy followed by delayed resection is recommended [9].

Damage to abdomino-pelvic anatomy may result from the tumour mass effect during development or may occur as a resultant complication of extensive dissection at the time of tumour resection. This can lead to morbid long-term sequelae including faecal and urinary incontinence, constipation, neurogenic bladder, and sexual dysfunction [10]. Studies to date which have sought to report long-term outcomes following treatment of SCTs, are however limited to mainly observational series involving small number of index cases.

Against this background the current study sought to therefore comprehensively analyse all available published studies to seek to better define the ‘real world’ or true long-term functional sequelae.

Material and Methods

Identification and Selection of Studies

A comprehensive search in PubMed and Embase databases was performed according to PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) guidelines [11]. The following terms were used as keywords: ‘sacrococcygeal teratoma’ and ‘sacrococcyx teratoma’ in combination with terms ‘Altman’, ‘outcome’, ‘urinary continence’, ‘faecal continence’, ‘bowel continence’, ‘sexual function’, and ‘quality of life’. All articles published up to February 28, 2022, were included in the study review.

Inclusion and Exclusion Criteria

This study included all original articles reporting on long-term (minimum two year) outcomes of SCT. Non-English language papers, and case reports (<3 patients) were excluded with title and abstract screening. Also, duplicate data from the same centres or research groups were excluded. (Figure 1)

Data Extraction and Analysis

Identified papers were independently reviewed by two study authors (AS and AR) with final selection approved by the senior author (PDL). Data on Altman tumour stage, size, histology and reported long-term functional outcome(s) were then extracted from the original publications.

Abnormal bladder function was defined as urinary incontinence and/or neurogenic bladder on urodynamics and/or need for clean intermittent catheterization (CIC).

Bladder and bowel functional outcomes were reviewed from the included studies and abnormal bladder function was herein defined as involuntary urinary leakage / incontinence in children older than 3 years of age. Soiling or faecal incontinence was defined as the involuntary passing of stool content and constipation symptoms were only included when it required medical therapy intervention(s) either with laxatives or enemas. Various validated tools have been used to define bladder and bowel continence [12-14], however due to the variability of the scoring systems utilised in the various studies included we have not been able to fully combine the results.

Statistical Analysis

Chi-Square and Fisher's exact tests were used to analyse categorical variables, and Wilcoxon test for continuous variables. A significance level of $p \leq 0.05$ (two-tailed) was set. No attempt to replace missing values was made. Analyses were performed using JMP Pro, version 16.1.0 for Windows (SAS Institute Inc., Cary, NC, USA).

Results

The original search identified 801 articles. A total of 543 studies were then evaluated with screening of titles and abstracts after duplicates were excluded. Eighty-five papers met strict inclusion criteria in screening and were then selected for full text review. After full text review of 85 articles, 37 papers met the eligibility criteria and were selected for review. (Figure 1) The published studies covered the time period from 1990 to 2021.

Final analysis yielded 37 studies involving 1,116 SCT patients (854 female; 77%) [15-52]. Individual data were available in 14 studies (222 patients). SCT diagnosis was established antenatally (20%). Median tumour lesion size was 7 cm (range 2 – 18) – Table 1.

According to the Altman Surgical Section AAP classification, there were 298/845 (35%) Type I, 252/845 (30%) Type II, 133/845 (16%) Type III, and 128/845 (15%) Type IV tumours. Most neoplasms were benign (640/858; 75%), 77/858 (9%) immature, and 141/858 (16%) malignant. Malignant tumours were of interest significantly more common among the higher Altman grade stages ($p<0.0001$). The commonest co-associated congenital anomalies were anorectal malformations 42/90 (47%), spinal dysraphism 19/90 (21%), and renal tract/urogenital disorders 10/90 (11%).

Abnormal bladder function was reported in 7/39 (18%) Altman Type I, 23/61 (37.7%) Type II, 11/34 (32.4%) Type III, and 15/25 (60%) Type IV cases ($p=0.007$). Abnormal bladder function was also more commonly associated with malignant tumours vs benign or immature tumours ($p=0.002$). Patients who required more than one surgical resection episode were found also more likely to develop urinary complications ($p=0.01$) – (Figure 2).

Soiling was documented in 19% index cases and constipation in 26% of patients with no apparent significant associations detected here with the individual tumour characteristics.

Lower extremity weakness was found less commonly observed in patients with Altman Type I-II (2/14) lesions (14.3%) vs Type III-IV tumours (5/6, 83.3%), $p=0.003$. Two patients who developed weakness had original SCT tumours which involved the spinal cord [21, 26]. Two patients received surgery to lengthen Achilles tendons and are now able to walk without assistance but with a mild degree of limp [42]. One patient was reported to have a unilateral hip subluxation as a sequalae of displacement by huge tumour without neurological deficit [30].

Sexual function was documented in only three studies, all with healthy comparison controls [24, 36, 46]. Sexual function scores were comparable in the male patients and controls with no increase (%) in erectile dysfunction. Female patients however scored much lower than their

healthy controls. Additionally, 55 index patients reported unsatisfactory cosmesis from operations and surgical scar site revision was performed in some 13/55 cases (24%).

Discussion

Abnormal bladder function is recognized after excision of SCT [53-55]. This may be attributed to factors such as bladder outlet obstruction from gross intrapelvic tumour extension [42, 53, 56, 57], or resultant from R0 extensive surgical resection [43], or a combination of both. This is well supported by the current findings of this systematic review which clearly show that higher Altman stage and requirement for reoperation(s) were strongly associated with a higher incidence of abnormal bladder function – (Figure 2). Index patients presenting with neurological symptoms at first primary diagnosis of SCT were usually at an advanced tumour stage though surprisingly in some cases complete neurological improvement can be observed following combination treatment of chemotherapy and surgical resection [28].

The reported incidence (%) of bowel dysfunction following excision of SCT has allegedly varied widely from 6% – 54% [17, 24, 27, 34], with many troublesome symptoms apparently improving or completely resolving over time [39, 42]. This current systematic review identified soiling in some 19% index cases and constipation in up to 26% of patients at a median follow-up time of 39 months. In our study population herein reported up to 47% children with SCT also had co-associated anorectal malformations and 21% also had associated spinal dysraphism, both factors which can also significantly attribute to bowel disturbance(s).

As functional symptoms are thought to improve or even completely resolve over time [15, 24], it has been suggested that minimal surgical intervention and ‘watchful waiting’ aftercare follow-up involving multi-disciplinary care teams may benefit long-term functional outcomes of patients [15]. Given the incidence (%) of abnormal bladder function documented in later life,

we however strongly recommend early urology speciality referral so that these children can proactively receive best care with appropriate early assessments and treatment.

Lower extremity weakness / motor disorders were surprisingly sparsely documented in the published studies we analysed. Locomotor morbidity was more common in patients with Altman stage III-IV tumours. Although these SCT tumours are more often malignant (%), lower extremity impairment was not strongly associated with tumour histology. We therefore speculate that more extensive surgery which is often required in Altman III-IV tumours affects the functional outcome. Also, higher grade and stage SCT tumours are often diagnosed late and / or undergo operation in older age groups.

Sexual function surprisingly after SCT appears to be somewhat satisfactory for the majority of patients from analysed studies [24, 36, 46]. Female patients though reported lower scores in sexual function than their controls. However, most females scored above the threshold for sexual dysfunction. As Kremer et al. [36] speculated, diminished sexual function may be caused by worse self-perceived body image likely from operation and scarring. Unsatisfactory scars were here a commonly reported problem in the analysed studies with scar revision rates of some 24%. Aesthetic new surgical approaches for SCT resection to minimise unsightly scars – eg. midline vertical incisions vs Chevron are now increasingly favoured as evidenced from recently published studies [32, 47, 58, 59].

From the current systematic review abnormal bladder function was overall documented in 35% of SCT patients with faecal soiling and constipation in 19% and 26% index cases respectively. Although impaired bowel and bladder dysfunction appears to be common after SCT resection strikingly only two studies comprising a total of 39 patients had healthy age-matched controls (n=91) for valid comparison. Rintala et al. [46] from Helsinki, Finland reported deficient anorectal function to be undeniably more common in SCT patients. Cozzi et al. [24] from Rome, Italy by contrast however found no significant difference(s) between SCT patients and

healthy controls regarding bladder and bowel function. However, it may be debated from these comparative individual reports that the total number of SCT patients vs healthy controls was perhaps too small indicating a crucial need for larger scale prospective international collaborative network studies to better answer these questions.

Limitations

We herein acknowledge this current systematic review has certain inherent limitations related to the variations in methods of data reporting from all the eligible published studies. Individual patient data records were also not available in all eligible studies we scrutinized. Finally, all extracted variables were not wholly accessible in all published studies making multivariate analysis impossible.

Conclusion

In summary, higher Altman grade stage, unfavourable tumour histology and re-operation are all strongly associated with poor long-term functional outcomes. Multidisciplinary team management with key involvement across all specialties ie. surgical oncology, urology, colorectal services is best recommended from SCT index diagnosis to better optimise clinical outcomes.

Author Contributions

The study was conceived and designed by PDL. AS and AR conducted the literature review, data collection, analysis and interpretation under supervision by PDL. The manuscript was drafted by AS and AR. All authors fully participated in manuscript drafting and revisions and approved the final, submitted manuscript.

Declaration of Competing Interest

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Legends

Figure 1. PRISMA SCT study selection flow diagram.

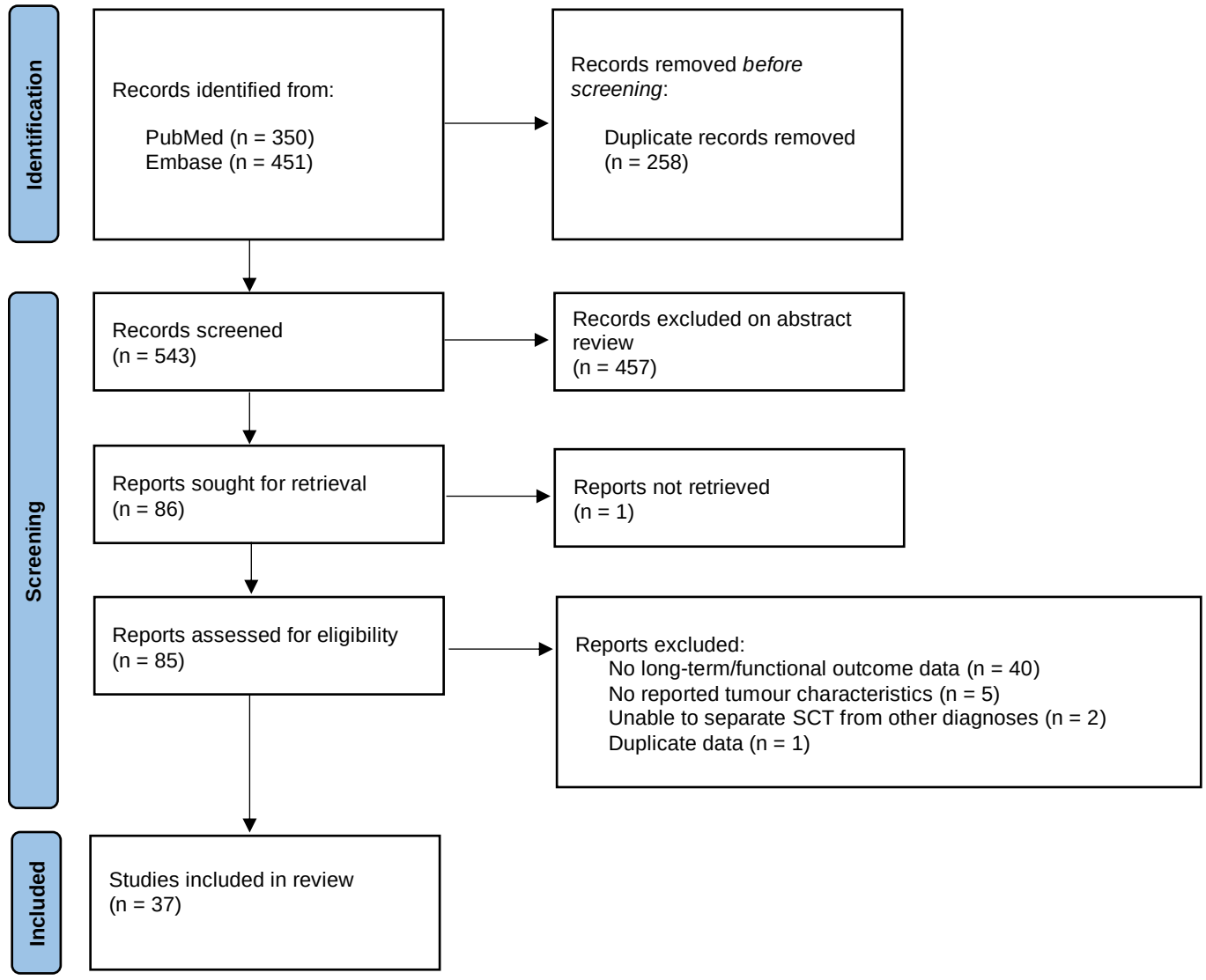

Figure 2. Long-term bladder function in patients with SCT.

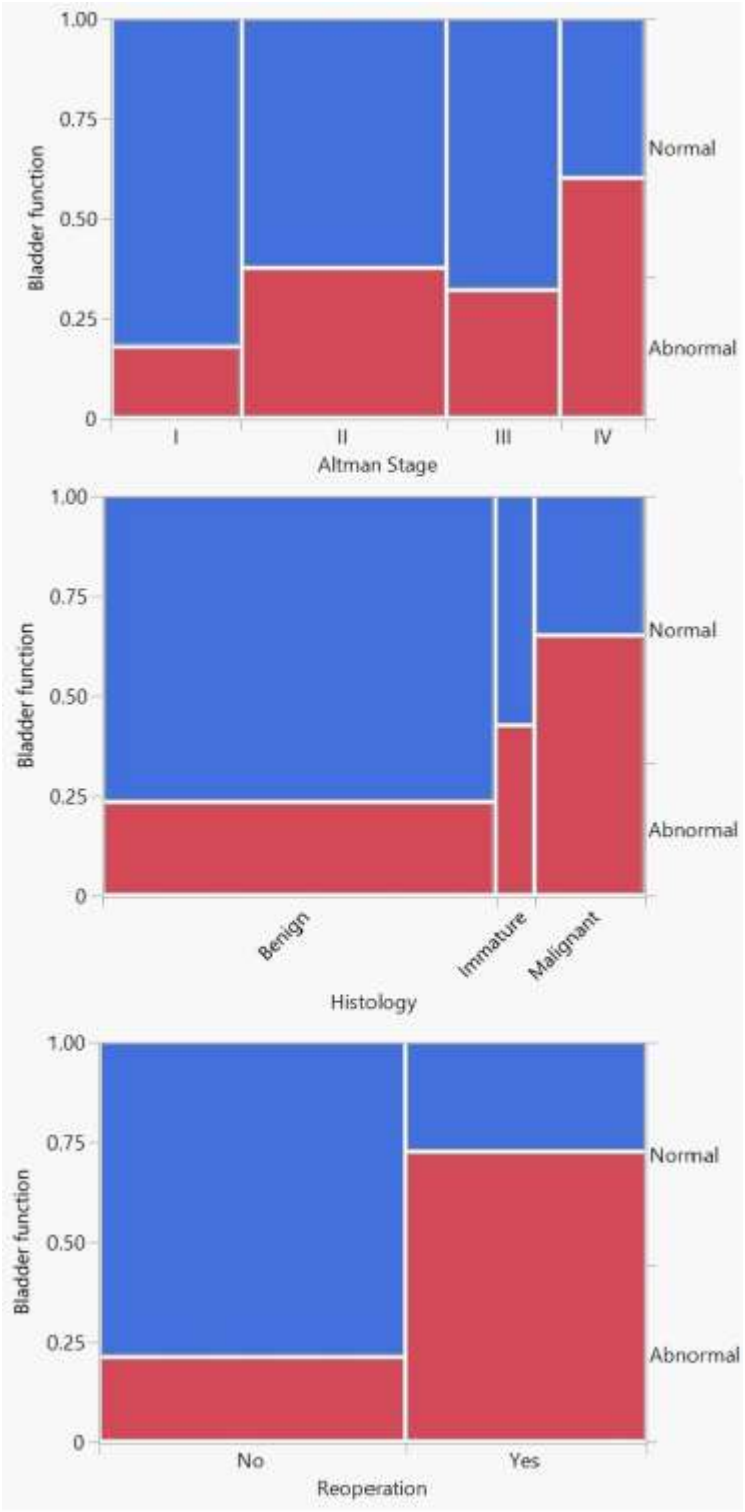

Table 1. Patient demographics. Values given as median and range. All columns include missing values.

	Female (%)	**Age at diagnosis (months)**	**Tumour size (cm)**	**Malignant tumour (%)**
Altman I (n=46)	27 (73.0%)	0 (-6 – 2)	14 (7 – 15)	2 (8.3%)
Altman II (n=68)	34 (77.3%)	0 (-5 – 3)	7 (3.5 – 15)	4 (13.3%)
Altman III (n=44)	27 (75.0%)	3.5 (-5 – 36)	7 (2 – 14)	5 (20.8%)
Altman IV (n=33)	24 (82.8%)	14.5 (-5 – 84)	4 (2.5 –15)	17 (65.4%)
Total (n=222)	117 (77.0%)	0 (-6 – 84)	7 (2 – 18)	30 (23.1%)