



The functional sequelae of sacrococcygeal teratoma: a longitudinal and cross-sectional follow-up study

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Abstract

Aim: The purpose of the study was to determine clinical manifestations, prevalence, and natural history of functional sequelae in patients operated on for sacrococcygeal teratoma (SCT) during infancy.

Methods: The medical records of 18 infants operated on for SCT were reviewed. Data recorded during admission and during routine outpatient appointments were analyzed. Moreover, 13 surviving patients (mean age, 25 years) and 65 age- and sex-matched controls were evaluated at a special outpatient clinic.

Results: Seven adult patients and 25 controls presented with one or more functional disorders including abdominal pain, constipation, diarrhea, soiling, enuresis, urinary frequency, urinary stress incontinence, and urinary straining. In 7 of 8 symptomatic children, one or more functional disorders disappeared during adult life. In comparison with controls, adult patients had a nonsignificant higher prevalence of each individual dysfunction.

Conclusions: Sequelae of SCT tend to improve with time, and the disorders reported are relatively common complaints in the general population.

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The first reports on the late sequelae of sacrococcygeal teratoma (SCT) appeared in the literature in the last 2 decades. These studies pointed out that, at follow-up, a good percentage of children operated on for SCT present with functional bowel dysfunction and bladder-voiding dysfunction [1–8].

With the passage of time, bowel functional sequelae such as chronic constipation and fecal soiling showed a tendency to improve [3,5,6]. Nevertheless, the prevalence of chronic

constipation and fecal soiling was found to be high in a study of young adults, which, to our knowledge, is the only case-control study on the late functional bowel sequelae of excision of SCT [9]. Actually, none of the adult patients had been longitudinally followed up [9].

The aim of this investigation was to study the clinical manifestations, the natural history of complications, and their prevalence in a cohort of patients operated on in infancy for SCT. For this purpose, we reviewed the records of our own series of patients with SCT; in addition, we made a case-control study of late sequelae. These data can be most valuable in counseling families.

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1. Materials and methods

Eighteen children were operated on for SCT by the senior surgeon (FC) at the Pediatric Surgical Unit of the University of Rome "La Sapienza" (Rome, Italy) between 1968 and 1988. The medical records of these patients were reviewed. Recorded data on patient demographics, preoperative complications, postoperative complications during admission, and late sequelae during regular outpatient follow-up were analyzed. Four of the 5 patients with malignant teratoma died of disease progression. One newborn with benign teratoma died of neonatal sepsis.

The tumors were classified according to the criteria proposed by the Surgical Section of the American Academy of Pediatrics [10]. Ten patients had type I (predominantly external tumor), 2 type II (intrapelvic component), 2 type III (tumor reached into the abdomen), and 4 type IV (intrapelvic tumor with no external presentation). In 11 patients, the tumor was diagnosed at birth, in 6 during the first 12 months of life, and in 1 at 36 months of life. The mean age at operation was 6 months (range, 1 day to 40 months). Twelve of the 18 patients were female. Four patients had a malignant local recurrence, and 2 patients had a benign local recurrence. Four of the 10 patients with type I had an intrapelvic recurrence.

The late functional sequelae of the 13 surviving patients were evaluated at a special follow-up outpatient clinic at a mean age of 25 years (range, between 17 and 38 years). A detailed history was obtained including all potential complications related to SCT. A physical examination was performed in each patient, including digital rectal examination with estimation of anal sphincter tone and assessment of the sensorimotor function of the lower extremities. Sixty-five age- and sex-matched controls, without a history of pelvic surgery (5 for each surviving patient), were randomly recruited and interviewed in person.

Constipation was defined as the passage of less than 3 stools per week and/or hard or lumpy stool. *Diarrhea* was defined as the passage of more than 3 stools per day and/or loose or

watery stool. *Urinary straining* was defined as the need of an effort to void the bladder. *Urinary stress incontinence* was defined as a leak of urine with coughing or laughing with a full-filled bladder. *Enuresis* was defined as nocturnal wetting beyond the age of 4 years. *Urinary frequency* was defined as needing to void at least 7 times/24 h.

All patients and controls gave informed consent. Statistical analysis was performed using Fisher's Exact test. All data analysis was conducted via SPSS 12.0 (SPSS, Chicago, Ill). Significance was set at the 5% level. All values calculated were one tailed.

2. Results

2.1. Complications during childhood

Eight of 13 surviving children presented with one or more bowel disturbances including abdominal pain, chronic constipation, recurrent episodes of diarrhea, soiling, and flatulence. According to the hospital notes, bowel function was satisfactory during the first few years of life. On average, bowel disturbances started at school age. Three children occasionally had minor soiling, usually during episodes of diarrhea, whereas 2 additional children required protective aids or frequent change of underwear. One child had difficulties controlling flatus at school age.

Of the 8 children with bowel disturbances, 5 had also minor bladder-voiding disturbances including straining, frequency, and stress incontinence, which, according to the hospital notes, all started at school age. Three of the 5 patients with bladder-voiding disturbances had nocturnal enuresis up to the age of 5, 15, and 20 years, respectively.

2.2. Complications during adulthood

Seven adult patients and 25 controls presented one or more functional genitourinary and/or bowel dysfunction (54% vs 38%; $P =$ not significant). Abdominal pain persisted in 3 patients, resolved in 2, and presented for the first time in 2. Constipation persisted in 5 patients and resolved in 1. Recurrent episodes of diarrhea persisted in 1 patient, resolved in 1, and presented for the first time in 4. Soiling and flatulence disappeared with time; only 1 adult patient had minor soiling after evacuation (Table 1).

Four adult patients with bowel disturbances also had one or more bladder-voiding dysfunctions. Straining on micturition completely resolved in 2 patients and improved in 1. Stress incontinence disappeared in 3 patients and first appeared in 1. Frequency disappeared in 1 patient and presented for the first time in 2.

Three adult patients with bowel disturbances also had sexual problems: 2 had leaking of small amount of urine during intercourse and one complained of dyspareunia. In 7 children, 1 or more functional dysfunctions disappeared

Table 1 Prevalence of bowel and bladder functional disturbances: comparison between 13 adult patients operated on for SCT and 65 controls

	Patients (%)	Controls (%)	<i>P</i>
Bowel dysfunction	54	37	.2
Pain	38	28	.32
Constipation	38	20	.14
Diarrhea	38	15	.07
Soiling	8	5	.52
Bladder dysfunction	38	17	.09
Enuresis	23	6	.09
Urinary frequency	15	15	.7
Stress incontinence	15	5	.19
Straining	8	-	.18
Sexual problems	23	8	.12

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