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Urological and intestinal functional outcomes of sacrococcygeal teratoma following surgery

Unal Bicakci[a*], Dilek Demirel[b], Nilufer Bicakci[c], Yasar Issi[a], Beytullah Yagiz[a], Mithat Gunaydin[a], Burak Tander[a], Ender Ariturk[a], Ferit Bernay[a]

[a] *Department of Pediatric Surgery and Divison of Pediatric Urology, Faculty of Medicine, Ondokuz Mayis University, Samsun, Turkey*
[b] *Pediatric Surgery, Maternity And Childrens Hospital, Samsun, Turkey*
[c] *Department of Nuclear Medicine, Educational and Research Hospital, Samsun, Turkey*

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*** Correspondence to:**
Unal Bicakci
Department of Pediatric Surgery and
Divison of Pediatric Urology,
Faculty of Medicine,
Ondokuz Mayis University, Samsun,
Turkey
e-mail: drbicakci@gmail.com

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ABSTRACT

Surgery for sacrococcygeal teratoma (SCT) in neonatal period may result in functional problems of micturation and defecation caused by pelvic nerve damage. We aimed to evaluate long term outcome of patients with SCT in terms of bladder dynamics. To evaluate the postoperative urological outcome related with SCT excision, urodynamic study, ultrasonographic and scintigraphic examinations were performed. The families are asked about the micturition and defecation paterns. Twenty-one patients (eight male, thirteen female) underwent SCT excision between 2006 and 2013 and seventeen (six male, eleven female) are regarded eligible for long term evaluation. Incidence of constipation was 9.4%, encopresis 9.4% and urinary incontinence 4.7%. Two patients had a history of urinary tract infection without reflux on voiding cystourethrography or renal scarring on renal cortical scintigraphy. Three patients had increased postvoiding residual urine of 25 to 35 mL while four had increased bladder capacity. Meticulous surgical technique for excision of SCT in newborns may avoid significant urinary and defecation problems subsequently.

1. Introduction

The most common tumor of the newborn is sacrococcygeal teratoma (SCT) when the stillborns are included. Although more than 90% is benign, they have the potential for malignant transformation. The primary choice of treatment is excision of the tumor together with the coccyx (Havranek et al., 1992; Cozzi et al., 2008). After the complete and successful excision of the tumor; temporary enuresis, encopresis, depletion of lower extremity motor functions can occur. Also, long-term functional sequelae including fecal and urinary incontinence, recurrent urinary tract infections, chronic constipation, probably due to surgical injury to pelvic nerves or compression effect of the large tumor are also remarkable problems (Gabra et al., 2006).

In this paper, we aimed to evaluate the long-term urinary and gastrointestinal outcome of patients after SCT excision.

2. Patients and methods

The data of patients with SCT within an eight-year period were collected retrospectively and also evaluated prospectively. Twenty-one patients (nine male, twelve female) underwent SCT excision between 2006 and 2013. Seventeen (six male, eleven female) are regarded eligible for evaluation of long term functional outcomes. Age of the patients ranged from 20 months to eight years. Median follow up was four years. Type of the SCT were type I (n=2), II (n=6) or III (n=9). There was no type IV SCM. All of the patients were operated at the neonatal period. Urological evaluation constitute urodynamic study, ultrasonography and renal cortical scintigraphy (if necessary). The families were asked whether their children have postoperative functional elimination problems such as involuntary bowel movements, soiling, urinary incontinence and constipation.

Statistically, descriptive analysis was performed.

3. Results

Urodynamic examination revealed enlarged bladder capacity in four patients and increased residual urine in three (20, 25 and 35 mL). Detrussor overactivity is encountered in two patients and one of them had also enuresis nocturna. On ultrasound, diameter of the bladder wall was increased in two patients (1-2 cm) and a pelvicaliceal fullness was present in four. Two patients had hydronephrosis with AP diameter over 1 cm without an underlying condition such as vesicoureteral reflux (VUR), etc. Two patients had a history of urinary tract infection but none of them had VUR or nephropathy. We performed renal cortical scintigraphy (DMSA) in eight patients and none had renal scarring. There were two patients with unilateral hypoplastic kidney without renal scarring. Only two patients had constipation and another two patients had encopresis. All four were temporary and resolved with proper management. The position and appearence of anus was normal in all instances. Only one patient needed a revision surgery for the cosmesis.

4. Discussion

Patients with SCT may have early or long term functional impairments after operation. Temporary enuresis and encopresis, depletion of lower extremity motor functions are considered as short term morbidities while fecal or urinary incontinence, neuropathic bladder, recurrent urinary tract infections and chronic constipation are long-term functional morbidities.

Malone et al. (1990) were the the first investigators reporting the functional impairment following SCT excision and showed that tumors with large intrapelvic extensions are associated with higher incidence of fecal and/or urinary incontinence. Another study showed functional abnormalities in five of seventeen patients which consisted of fecal soiling, nocturnal enuresis, perineal anesthesia and recurrent urinary tract infections but poor functional outcome was independent of the presence of the intrapelvic component (Schmidt et al., 1999). Havranek et al. (1992) reported 16% urinary incontinence and 40% soiling rates. They found no difference in functional outcome between patients with intrapelvic or extrapelvic tumor location. Gabra et al. (2006) reported that approximately one-third of patients developed some degree of long-term functional impairment, including soiling, neuropathic bladder, VUR, and recurrent urinary tract infections. Draper et al. (2009) showed that only one of the ten patients who participated the study had problems about fecal or urinary continence, or lower extremity weakness while constipation was seen in five. In our series, incidence of constipation was 9.4%, encopresis 9.4% and urinary incontinence 4.7%. Two patients had a history of urinary tract infection without reflux on voiding cystourethrography or renal scarring on DMSA scintigraphy. Three patients had increased postvoiding residual urine of 25 to 35 mL while four had increased bladder capacity.

Le et al. (2011) investigated the incidence and severity of urologic complications in patients with SCT and encountered neurogenic bladder in nine of the twenty patients which was attributed to the mass effect of the tumors. Among these nine patients, five also had renal injury. These five patients had higher grade lesions. The authors suggested that type 3 and type 4 lesions should alert the surgeon for the potential urologic sequelae (Le et al., 2011). In our series, although there was no type IV SCT (presacral teratoma), there were six patients with type III. We have not demonstrated any relation between SCT type and sequelae development.

Tailor et al. (2009) had reported major urologic complications (3 in nine patients) and they suggested that routine ultrasonography should be performed in the first postoperative year.

In many other studies, soiling was reported in 24%-27%, constipation in 8%-35%, urinary complications in 16%-28% (Rintala et al., 1993; Gabra et al., 2006; Ozkan et al., 2006), after SCT resection while in the larger series reported by Derikx et al. (2007) constipation was present in 16%, urinary incontinence in more than 30%. In our series, incidence of constipation was 9.4%, soiling 9.4% and urinary incontinence 4.7%.

Our data has more optimistic results than the previous studies in terms of urinary and bowel dysfunction. We did not find any serious urological or intestinal sequleae. We encountered moderate problems that resolved with simple medical interventions. We suggest that meticulous surgical technique for excision of SCT in newborns may avoid any subsequent significant urinary and defecation problems. Larger part of the teratoma involved in presacral area and pressure onto

sacral nerves may be the primary factor on subsequent functional outcome of surgery. Excision of this type of SCT is more challenging than others.

In conclusion, the clinicians and families should be aware of the potential long-term sequela after SCT excision. A standart follow-up protocol questionnaire needs to be created to assess long-term sequelae and follow-up strategy. Ultrasonography in the first postoperative year may help to identify any urological problems. Patients with urinary problems including urinary tract infection, urinary or fecal incontinance should be seeked.

REFERENCES

Cozzi, F., Schiavetti, A., Zani, A., Spagnol, L., Totonelli, G., Cozzi, D.A., 2008. The functional sequelae of sacrococcygeal teratoma: A longitudinal and cross-sectional follow-up study. J. Pediatr. Surg. 43, 658-661. doi: 10.1016/j.jpedsurg.2007.10.066.

Derikx, J.P., De Backer, A., van de Schoot, L., Aronson, D.C., de Langen, Z.J., van den Hoonaard, T.L., Bax, N.M., van der Staak, F., van Heurn, L.W., 2007. Long-term functional sequelae of sacrococcygeal teratoma: A national study in the Netherlands. J. Pediatr. Surg. 42, 1122-1126. doi:http://dx.doi.org/10.1016/j.jpedsurg.2007.01.050.

Draper, H., Chitayat, D., Ein, S.H., Langer, J.C., 2009. Long-term functional results following resection of neonatal sacrococcygeal teratoma. Pediatr. Surg. Int. 25, 243–246. doi: 10.1007/s00383-009-2322-1.

Gabra, H.O., Jesudason, E.C., McDowell, H.P., Pizer, B.L., Losty, P.D., 2006. Sacrococcygeal teratoma: A 25-year experience in a UK regional center. J. Pediatr Surg. 41, 1513-1516. doi: http://dx.doi.org/10.1016/j.jpedsurg.2007.01.050.

Havranek, P., Hedlund, H., Rubenson, A., Güth, D., Husberg, M., Frykberg, T., Larsson, L.T., 1992. Sacrococcygeal teratoma in Sweden between 1978 and 1989: Long-term functional results. J. Pediatr. Surg. 27, 916-918. doi: http://dx.doi.org/10.1016/0022-3468(92)90398-q.

Le, L.D., Alam, S., Lim, F.Y., Keswani, S.G., Crombleholme, T.M., 2011. Prenatal and postnatal urologic complications of sacrococcygeal teratomas. J. Pediatr Surg. 46, 1186-1190. doi: http://dx.doi.org/10.1016/j.jpedsurg.2011.03.045.

Malone, P.S., Spitz, L., Kiely, E.M., Brereton, R.J., Duffy, P.G., Ransley, P.G., 1990. The functional sequelae of sacrococcygeal teratoma. J. Pediatr Surg. 25, 679-680. doi: http://dx.doi.org/10.1097/00006534-199110000-00078.

Ozkan, K.U., Bauer, S.B., Khoshbin, S., Borer, J.G., 2006. Neurogenic bladder dysfunction after sacrococcygeal teratoma resection. J. Urol. 175, 292-296. doi: http://dx.doi.org/10.1016/s0022-5347(05)00012-1.

Rintala, R., Lahdenne, P., Lindahl, H., Siimes, M., Heikinheimo, M., 1993. Anorectal function in adults operated for a benign sacrococcygeal teratoma. J. Pediatr Surg. 28, 1165-1167. doi: http://dx.doi.org/10.1016/0022-3468(93)90156-f.

Schmidt, B., Haberlik, A., Uray, E., 1999. Sacrococcygeal teratoma: Clinical course and prognosis with a special view to long-term functional results. Pediatr Surg. Int. 15, 573-576. doi: http://dx.doi.org/10.1007/s003830050675.

Tailor, J., Roy, P.G., Hitchcock, R., Grant, H., Johnson, P., Joseph, V.T., Lakhoo, K., 2009. Long-term functional outcome of sacrococcygeal teratoma in a UK Regional Center (1993 to 2006). J. Pediatr. Hematol/Oncol. 31, 183-186. doi: http://dx.doi.org/10.1097/mph.0b013e318199389d.