Sacrococcygeal teratoma excision: a vertical rather than transverse wound closure

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Background The chevron incision has been the standard approach for sacrococcygeal teratoma (SCT) excision. Here, we are reporting our experience of shifting to the vertical posterior sagittal approach.

Patients and methods During the period 2011 through 2016, we operated on 17 (16 female and one male) cases of SCT. Their age at presentation ranged from day 1 to 26 months (mean = 4.8 months, median = 2 months). The chevron incision was used in five, whereas the vertical posterior sagittal approach was used in 12 patients.

Results In this series, we had one case of perioperative mortality, in addition to another case of perineal wound disruption (in the group of vertical wound closure), which was managed conservatively (to heal by secondary intention) with a very satisfactory hidden scar at 6-month follow-up. Overall, we did not find the vertical approach to add any extra limitations to the surgical exposure or

Introduction

Sacrococcygeal teratoma (SCT) is the most common tumor seen in the neonatal period [1]. Some cases may present later in infancy or occasionally during childhood [2,3]. Most reports have shown female predominance [2,4]. Gross *et al.* [5] described a standardized surgical approach to excise the intact mass together with the coccyx through the chevron incision, with the infant in the prone (face down) position. Gross emphasized on the benefit and feasibility of tumor excision regardless 'of the large size of the mass or the small size of the patient' [5]. Successive reports have confirmed the favorable surgical outcome for most cases of SCT [3,6,7]; tumor recurrence (usually within the first 3 years) and delayed functional disability (regarding urinary and bowel control) are major postsurgical concerns [3,8,9].

The chevron (inverted V) incision has been the standard approach used by most pediatric surgeons [10]. However, there have been reports on the unsatisfactory cosmetic results, and that 'a better surgical procedure for closing the buttock region' should be discussed [11,12]. A vertical incision may be preferred for smaller teratomas leaving a normal-looking median raphe [10]. Jan *et al.* [13] successfully applied the vertical posterior sagittal approach for large tumors as well.

Over the last years, we have been using the classic transverse 'chevron' incision for excision of SCT, which has resulted in a cosmetically suboptimal outcome, in addition to the occasionally encountered 'distressing' complication of having the anus pulled up ending in the back of the patient (Fig. 1). Here, we are reporting our experience of shifting to vertical wound closure after dissection; meanwhile, it provided a well-recognized cosmetic advantage.

Conclusion The vertical posterior sagittal approach for excision of SCT is both feasible and advantageous in terms of the cosmetic outcome. It provides a well-hidden scar in the natal cleft and preserves normal contouring of the buttocks. *Ann Pediatr Surg* 13:207–212 © 2017 Annals of Pediatric Surgery.

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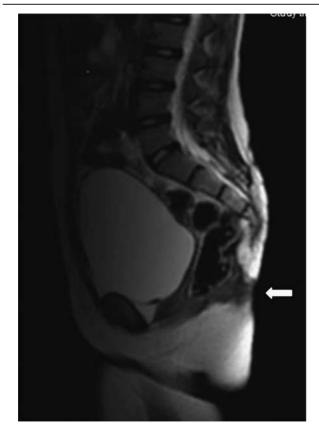
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Fig. 1



Midsagittal MRI (T2WI) of a 15-year-old female patient with a history of excision of a huge sacrococcygeal mass in the neonatal period, now presenting with fecal incontinence and abnormal backward location of her anus (white arrow).

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Fig. 2



5-month-old female patient with a relatively small sacrococcygeal teratoma. (a) Pelvic MRI (axial T2WI) showing the mass displacing the anorectum (black arrow). (b) The patient placed in the prone (face down) position. (c) Dissection of the mass off the rectum. (d) After excision of the mass, the pelvic floor muscles are closed vertically in the midline from below upwards. (e) Vertical midline skin closure. (f) Follow-up at 3 weeks postoperatively.

SCT excision. The scientific committee approved reporting our findings without review.

Patients and methods

The study included patients operated for SCT during the period 2011 through 2016. At the beginning, the chevron incision was routinely used for all cases. The resultant suboptimal scar stimulated us to shift to the posterior sagittal midline incision with smaller tumors (Fig. 2), which we have found to provide comparable excellent surgical exposure and superior cosmetic outcome. Lately, we used a vertical elliptical skin incision to excise large SCT as well, which is our main concern in this report.

Operative procedure

The patient is placed in the prone position. A vertical elliptical skin incision is made over the tumor, extending from the coccyx down to near the anal orifice (Fig. 3b). The involved unhealthy skin over the tumor should be included within the skin ellipse to be excised 'en-bloc' with the tumor mass (Fig. 4). Dissection progresses laterally on both sides, separating the mass from the healthy skin and then from the gluteal muscles. Incision through the sacrococcygeal junction will allow separation and excision of the coccyx together with the tumor mass and will provide exposure to the median sacral artery. The latter may be of considerable size, especially with large solid tumors, where it should be ligated and severed to allow safe delivery of the upper part of the

mass. SCT with considerable intra-abdominal tumor extension (Altman type III, Fig. 5) usually will require additional lower midline laparotomy incision (after turning the patient supine) to control the median sacral artery (high-up opposite the sacral promontory) and complete dissection of the abdominal component of the tumor.

The dissection continues to free the tumor mass on both sides and from above. The last step is to release the attachments between the mass and the back of the rectum. Almost always one can find a plane of dissection between the mass and the compressed rectum; however, occasionally, an injury to the rectum may necessitate a covering colostomy.

After excision of the tumor, the perineal wound is closed vertically by reapproximating the pelvic floor muscles in the midline behind the rectum, starting from below upwards (Fig. 2d). At the site of the excised coccyx, the muscles are too widely separated to be reapproximated in the midline, and so they are just left apart leaving a triangular gap below the sacrum (Fig. 2d). A drain is left in the tumor bed getting out from the edge of the wound, or through a separate lateral skin incision. The skin is also closed vertically in the midline. Care should be given during planning the skin incision from the start, in order to leave sufficient skin to be closed in the midline without tension. Excess tension on the suture line is sometimes inevitable due to massive involvement of the skin by the underlying tumor mass (Fig. 4). With large protruding tumors, a slight modification is made to the

Fig. 3



Two-day-old female with cystic type of sacrococcygeal teratoma. (a) Midsagittal MRI showing the cystic nature of the tumor and the intra-abdominal extension (type III). Note the intact vertebral bodies to differentiate it from anterior meningeocoele. (b) The patient placed in the prone (face down) position; marking the skin for the location of the coccyx and skin incision. (c) Modified vertical midline skin closure into inverted Y configuration. (d, e) The appearance of the scar at 2-week follow-up. (f) A well-hidden scar at 6-month follow-up.

lower end of the vertical skin closure into an inverted Y configuration (Figs 3c and 4e). We have found this modification helpful in managing redundant skin at this area, and in correcting forward anal tilting caused by the mass effect of the tumor.

Results

During the period 2011 through 2016, we operated on 17 (16 female and one male) cases of SCT. Cases of presacral dermoid cysts (mature cystic teratomas) associated with anorectal anomalies and sacral dysplasia as a part of Currarino syndrome were not included in this report. All cases except one presented either in the neonatal period or during the first year of life. Their age at presentation ranged from day 1 to 26 months (mean = 4.8 months, median = 2 months). According to Altman classification [4], five cases were of type I, 10 were of type II, and two cases were of type III. Five cases were operated using the classic 'chevron' incision, whereas the vertical posterior sagittal approach was used in the remaining 12 cases.

Among the two cases of SCT type III (having intraabdominal extension), one was purely cystic and it could be aspirated intraoperatively and excised completely through the perineal wound (Fig. 3). The other required an additional midline laparotomy incision to complete the dissection from the abdomen, and to perform a covering colostomy for a repaired rectal injury. The latter patient was referred to our hospital 25 days after birth with a huge pelviabdominal mass, severe abdominal distention, and bilateral hydroureteronephrosis (Fig. 5). The rectum was so stretched and thinned out over the pelvic component of the mass, predisposing to bowel injury during dissection; otherwise, the surgical dissection (both the abdominal and pelvic parts) went smoothly as usual. The laparotomy incision was complicated by burst abdomen on the third postoperative day requiring reoperation to close the abdomen. Unfortunately, the patient died on the 10th postoperative day from septic complications and metabolic derangements, representing the only perioperative mortality in this series. We had another case of perineal wound disruption (in the group of vertical wound closure), which was managed conservatively (to heal by





Three-day-old female patient with sacrococcygeal teratoma; (a) the prone (face down) position; (b, c) marking the skin incision; (d) the tumor bed after excision. (e) Modified vertical midline skin closure into inverted Y configuration; note the presence of some tension on suture line; (f) 10 days postoperatively showing wound disruption; (g) healing by secondary intention resulting in a satisfactory hidden scar at 6-month follow-up.

secondary intention) with a very satisfactory hidden scar at 6-month follow-up (Fig. 4).

Overall, we did not find the vertical approach to add any extra limitations to the surgical exposure or dissection; meanwhile, it provided obvious cosmetic advantage (hidden scar with good contouring of the buttocks; Figs 3 and 4) well recognized by both the parents and medical staff.

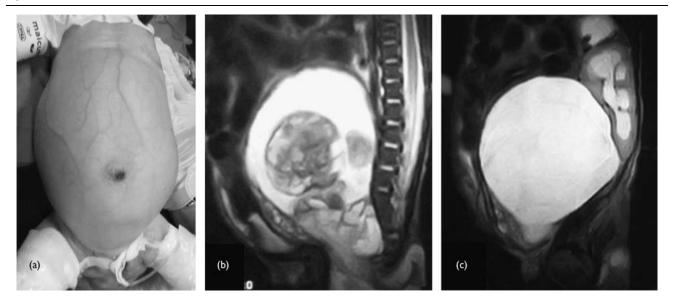
Discussion

The term 'teratos' means 'monster' [10], which reflects the terror experienced by the parents on having a neonate born with such a large bizarre sacrococcygeal mass that may weigh more than the weight of the whole baby itself. However, contrary to what the term implies, most cases of SCT were shown to have a favorable surgical outcome [3,6,7]. One exception is the presence of malignant foci within the tumor and another related to large antenatally diagnosed tumors that may cause severe circulatory compromise associated with life-threatening complications (hydrops fetalis, misconception). Recently, several reports appeared in the literature discussing antenatal diagnosis, risk stratification, and trials for fetal intervention [7,14-17]. The risk for hemorrhage is another point of concern, which has raised a role for laparoscopy in the era of minimally invasive surgery for

controling the median sacral artery to stabilize the baby before tumor excision [18].

As regards the surgical excision of SCT, few (if any) modifications have been made to the original technique described by Gross et al. [5]. The main principles are to excise the whole mass together with the coccyx, and to avoid tumor spillage during the operation [19]. In this report, we are concerned with the method of reconstruction of the muscles and closing the perineal wound after SCT excision. Looking at the pelvic anatomy, one can find most of the muscles to be arranged in a parasagittal manner, expecting them to be displaced laterally by the expanding 'median' sacrococcygeal tumor. Therefore, after excision of the tumor, it makes more sense to reapproximate the pelvic floor muscles vertically in the midline rather than performing a transverse closure. Classically, the central portion of the levator sling around the anorectum is sutured to the perichondrium of the anterior of the sacrum, which we believe to be a nonanatomical reconstruction that may predispose to the distressing complication of having the anus pulled up ending in the back of the patient [12].

As regards the skin closure, the vertical midline scar is a well-hidden scar in the natal cleft, preserving the normal contour of the buttocks and providing a well-recognized cosmetic advantage over the classic chevron incision. However, there might be major concerns when dealing Fig. 5



A 25-day-old female patient with a huge pelviabdominal sacrococcygeal teratoma type III. (a) Marked abdominal distention; note the dilated veins denoting inferior vena cava compression by the mass. (b) Midsagittal MRI (T2WI) showing combined solid and cystic components of the mass with intact vertebral bodies excluding intraspinal communications. (c) Parasagittal cuts showing hydronephrosis.

with large SCT, due to the expected limited exposure compared with the 'classic' chevron incision. With large tumors, the vertical incision actually turns to be elliptical giving a very wide exposure (almost similar to that of the chevron incision, Fig. 4). In our experience, planning for a vertical midline closure did not add any further restrictions to surgical exposure or dissection. Even when complicated by wound disruption (due to excess tension on the midline skin closure), healing by secondary intention resulted in a perfectly hidden scar at 6-month follow-up (Fig. 4).

With large protruding SCT, the anus is seen tilted forwards by the expanding posterior tumor. Unlike cases of Currarino triad [20], the anal canal is just pushed by the mass effect of the tumor and does not represent a sort of anorectal malformation. After removal of the mass, the anus will settle back to normal position. This can be further assisted by tailoring the lower end of the vertical skin closure into an inverted Y configuration, which we have found to be beneficial in dealing with any skin redundancy in this area.

We had one perioperative mortality in this case series, from which we have learned some lessons. The huge and complex appearance of the tumor in relation to the small size of the neonate may result in some hesitancy as regards the decision of surgery, hence leading to unnecessary delay. The delay in our case was 25 days before referral to our hospital, which most probably had exaggerated (to some extent) the adverse effects on the general condition of the neonate. Sometimes the delay is related to diagnostic confusion associating the cystic types of SCT. A midline cystic mass intimately in front of the vertebral column may cause confusion with anterior meningocoele [21]. In cases of SCT, the presence of intact vertebral bodies on MRI or CT scans would be quiet reassuring about the diagnosis and for the absence of any intraspinal communication [22]. Another lesson that we have learned was during closing the laparotomy incision after excising the abdominal component of SCT type III. Although closing the abdomen seemed to be an easy job after the successful removal of such a huge abdominal mass, the stretched (thinned out) abdominal wall muscles would require careful attention during closure to avoid failure (burst abdomen). Definitely, an extra unnecessary operation in this situation would have added to the adverse prognostic factors for such a vulnerable neonate.

The small number of cases is an expected limitation with a single center study on such a rare disease. Moreover, we did not address some important oncological issues (tumor pathology and rate of recurrence), which are beyond our scope in this report. Our main concern in this report was early postoperative complications and how to improve the cosmetic outcome, which represents one of the most important postsurgical sequels following excision of SCT [11].

Conclusion

The vertical posterior sagittal approach for excision of SCT is both feasible and advantageous in terms of the cosmetic outcome. It provides a well-hidden scar in the natal cleft and preserves normal contouring of the buttocks.

Conflicts of interest

There are no conflicts of interest.

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