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Vertical wound closure following sacrococcygeal teratoma excision: an approachable aesthetic solution

Amr Abdelhamid AbouZeid*, Ahmed Bassiouny Radwan, Mostafa Mohamed Elghandour, Nader Nassef Guirguis and Mohamed Ali Bersy

Abstract

Background: Sacrococcygeal teratoma is a rare tumour, usually presenting in the neonatal period. The benign nature of most tumours and the high survival rates would emphasise on the importance of both cosmetic and functional outcomes.

We report on our extended experience with more cases concentrating on the aesthetic outcome of vertical wound closure following excision of large irregular sacrococcygeal tumours.

The study included primary cases of sacrococcygeal teratoma who were referred to our surgical team for excision. Cases of presacral tumours associated with anorectal anomalies and sacral bony defects (Currarino triad) were excluded. In all cases, we planned for a vertical midline wound closure after tumour excision. The aesthetic outcomes are evaluated concerning the vertical midline scar, buttock's contour, and position of the anus.

Results: In addition to twelve previously reported cases (during the period 2011 through 2016), we included another ten new consecutive cases operated during the period 2017 through 2021. Collectively, the study included 22 cases of sacrococcygeal teratoma that underwent vertical perineal wound closure after excision of the tumour. In 13 cases (those with relatively small or medium-sized tumours), the perineal wound was perfectly closed in the midline (well-hidden vertical scar in the natal cleft). For the rest of the cases (9 cases with large and/or irregular sacrococcygeal tumours), some modification was applied on the vertical linear mid-line skin closure to accommodate for skin redundancy and irregularity at the lower end of the wound, usually ending with an 'inverted-Y' skin closure

Conclusion: Vertical wound closure was always feasible after excision of sacrococcygeal teratomas. Even with large and irregular tumours, the vertical scar was perfectly or partially hidden within the natal cleft. Usually, there was adequate buttock development with minimal disturbance to the normal anal location within the perineum.

Keywords: Teratoma, Scar, Cosmesis, Hip dysplasia, Buttock, Anus

Background

Sacrococcygeal teratoma is a rare tumour with a reported incidence of about 1:35,000 live births [1]. It is the most common tumour in the neonatal period [2], although

some cases may present later during infancy [1, 3]. Antenatal diagnosis allows to arrange for delivery at a specialised centre with paediatric surgical facilities that can manage the condition postnatally, while very selected cases may benefit from foetal intervention [4, 5].

The majority of sacrococcygeal teratomas are mature (benign) with favourable outcome [6]. Reported complications include life-threatening intraoperative bleeding from a large feeding vessel (median sacral artery) [7], risk

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of recurrence/malignant transformation [2, 3], functional problems related to bowel and urinary control [8], and lastly cosmetic issues related to unsatisfactory scar [9, 10]. Vertical midline skin closure has an aesthetic advantage over the traditional chevron incision by providing a well-hidden scar in the natal cleft [11-13]. Although the chevron incision is still popular, vertical closure may be preferred especially with small-sized tumours [10]. Jan and colleagues reported on their successful experience with the vertical posterior sagittal incision even with large sacrococcygeal teratomas [11]. In a previous report, we applied the principles of vertical wound closure following sacrococcygeal teratoma excision in 12 consecutive cases; a modified 'inverted-Y' skin closure was used to manage skin redundancy associated with large tumours [12]. Here, we report on our extended experience with more cases concentrating on the aesthetic outcome of vertical wound closure following excision of large irregular sacrococcygeal tumours.

Methods

The study included primary cases of sacrococcygeal teratoma who were referred to our surgical team for excision. Cases of presacral tumours associated with anorectal anomalies and sacral bony defects (Currarino triad) were excluded [14]. Preoperative cross-sectional imaging (MRI or CT) was needed to detect the level of deep tumour extension [15, 16]. Tumours with significant intrabdominal components (Altmann type III) would require a combined abdominoperineal approach (Fig. 1); otherwise, sacrococcygeal tumours are completely excised via a perineal approach (Fig. 2). In all cases, we planned for a vertical midline wound closure after tumour excision.

Operative technique

With the patient placed in the prone position, a vertical elliptical incision is made over the tumour to include involved unhealthy skin that would be excised 'en-bloc' with the tumour mass (Fig. 2). Here, we shall not go through the details of tumour excision that were discussed in previous reports [12]. After excision of the tumour, the perineal wound is closed vertically by reapproximating the pelvic floor muscles in the midline behind the rectum, starting from below upwards (Fig. 3). The skin is also closed vertically in the midline (Fig. 2). For large protruding tumours, some modification during closure of the lower part of the skin incision is needed to accommodate for skin redundancy [12]. Usually, this will end by a sort of an 'inverted-Y' skin

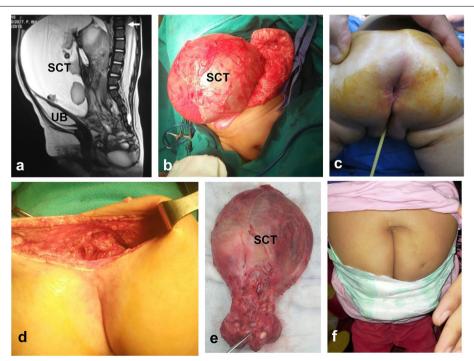


Fig. 1 Six-month-old girl presenting with huge pelviabdominal sacrococcygeal teratoma (Altmann III). **a** Mid-sagittal MRI showing major abdominal component reaching up to level T12 (arrow). **b** Abdominal incision to dissect the abdominal component. **c**, **d** The patient is then turned to the prone position to approach the pelvic component of the tumour. **e** The mass after excision. **f** Nine months later at follow-up, the vertical perineal scar (well hidden within the natal cleft). SCT, sacrococcygeal teratoma; UB, urinary bladder

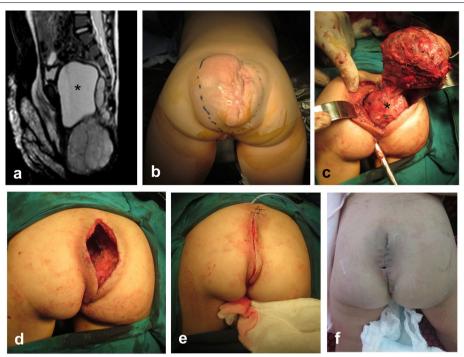


Fig. 2 Nine-month-old girl presenting with sacrococcygeal teratoma (Altmann II). **a** Mid-sagittal MRI showing a solid and a deep cystic component (*) of the tumour. The pathology of excised specimen showed a malignant component. **b** Marking for the skin incision (vertical elliptical) with the patient in the prone position. **c**, **d** Dissection and excision of the tumour through a perineal approach. **e** Vertical midline wound closure. **f** The vertical wound at three weeks follow-up

closure as illustrated (Fig. 4). A tube drain is kept in the tumour bed for few days postoperatively.

Follow-up

All cases were instructed to keep on follow-up every 3-4 months for at least 3 years after tumour resection. Although most tumours are benign, recurrence represents a concern at follow-up. Perineal examination and checking serum levels of tumour markers (alphafetoprotein) were helpful; Perineal examination was done for all cases at follow-up, while alpha-fetoprotein was ordered in high-risk patients (immature/malignant pathology). Imaging was ordered upon suspicion of recurrence. Routine asking about bowel habits and urinary symptoms is important to screen for possible functional complications. The aesthetic outcomes were evaluated concerning the vertical midline scar, buttock's contour, and position of the anus (Fig. 5). The vertical scar was considered perfectly hidden within the natal cleft when only seen upon retraction of the buttocks (Fig. 5h). The buttock's contour was either welldeveloped or distorted. Lastly, the anus was either in normal position or displaced.

Results

In addition to twelve previously reported cases (during the period 2011 through 2016) [12], we included another ten new consecutive cases operated during the period 2017 through 2021. Collectively, the study included 22 cases of sacrococcygeal teratoma that underwent vertical perineal wound closure after excision of the tumour. Their age at operation ranged from 3 days to 26 months (median: 24 days, mean: 3.7 months). There was a characteristic female predominance with a male-to-female ratio of 1 to 10. In all cases, complete excision of the tumour could be achieved. Two cases were associated with considerable intra-abdominal tumour extension (Altmann type III) reaching up to the level of the twelfth thoracic vertebra, which required a combined abdominoperineal approach to resect the tumour (Fig. 1). Otherwise, the rest of the sacrococcygeal tumours (20 cases) could be excised completely through a perineal approach. In 13 cases (those with relatively small or medium-sized tumours), the perineal wound was perfectly closed in the midline (well-hidden vertical scar in the natal cleft). For the rest of the cases (9 cases with large and/or irregular sacrococcygeal tumours), some modification was applied on the vertical linear mid-line skin closure to

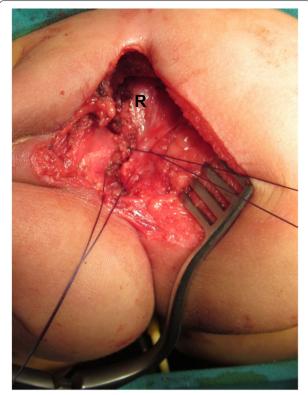


Fig. 3 After sacrococcygeal teratoma resection, the pelvic floor muscles are reapproximated in the midline behind the rectum (R), starting from below upwards

accommodate for skin redundancy and irregularity at the lower end of the wound (Fig. 6), usually ending with an 'inverted-Y' skin closure (Fig. 7). Postoperative major perineal wound disruptions were detected in three cases that were successfully managed conservatively to heal by secondary intention. We had a single case of postoperative mortality that occurred in a case that required an additional laparotomy incision to complete the dissection of the abdominal component of the tumour. 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 [12]. The pathology of resected tumours revealed mature cystic teratoma in 17 cases, immature teratoma in 4 cases, and malignant component (embryonal carcinoma) in one. Postoperative chemotherapy was established for the case with malignant component, in addition to another two cases that developed local tumour recurrence during follow-up. Local recurrence showed complete regression with chemotherapy; however, the tumour bed was surgically re-explored after chemotherapy in one of the two suspecting a small residual tumour mass that proved to be fibrous tissue. All



Fig. 4 Three-day-old boy presenting with large protruding sacrococcygeal teratoma. **a, b** With the patient in the prone position, the site of incision is marked on the skin to remove involved unhealthy skin with the mass. **c** Modified vertical skin closure (inverted-Y) to accommodate for skin redundancy at the lower end of the wound. **d** The skin wound three weeks postoperatively

three cases who received postoperative chemotherapy had tumour-free survival (3–5 years). From the functional point, there were no complaints regarding urinary and bowel control in this case series. Eight cases were old enough (beyond 3 years of age) to ask about continence; all of them have successfully achieved voluntary bowel control as well as daytime urinary continence.

The cosmetic outcome would be specifically addressed in this report. In 17 cases, the scar was perfectly hidden within the natal cleft at follow-up, which often required retraction of the buttocks to be seen (Fig. 5). Recently, we modified the site of the tube drain exit through the skin to be less apparent (Fig. 2e). In five cases (who presented with large irregular perineal tumours), the scar was not completely hidden with apparent branching at its lower end (Figs. 6g and 7g). However, in four out of these five cases, the scar was still satisfactory with minimal disturbance to the buttock's contour. In a single case (Fig. 8), the scar was less satisfactory at follow-up, significantly affecting the buttock contour on one side (left side). Besides the large and irregular tumour at presentation (Fig. 8a), this particular case had also neglected congenital hip dysplasia on the same 'left' side. Unfortunately, the diagnosis of congenital hip dysplasia was missed in



Fig. 5 The aesthetic outcome of vertical wound closure after sacrococcygeal teratoma excision in five different cases. The upper row (**a**, **b**, **c**, **d**, and **e**): different sizes of the tumour at presentation before operation. The lower row (**f**, **g**, **h**, **i**, and **j**): the cosmetic outcome several months after operation for the above cases, respectively. Note the relatively small size of the tumour in **a** and **b** with a perfect midline hidden scar; while a modified 'inverted-Y' skin closure was used for the rest of cases due to major skin involvement. Also, note the scar mark of the tube drain; recently, we have modified this site to be nearer to the upper end of the skin incision to avoid disturbing the buttock contour

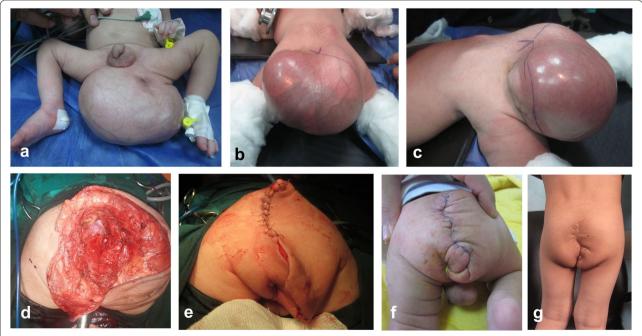


Fig. 6 Nine-day-old boy presenting with large protruding sacrococcygeal teratoma. **a** Patient in the supine position demonstrating the displacement of the anus. **b**, **c** The patient is placed in the prone position at operation. Note the marking on the skin for the site of coccyx and incision. **d** Tumour bed after excision of the tumour. **e** Modified vertical skin closure to accommodate for skin redundancy. **f** The scar in early postoperative follow-up. **g** The scar after 2 years



Fig. 7 Two-month-old girl presenting with large irregular protruding sacrococcygeal teratoma. **a** Patient in the supine position demonstrating the displacement of the anus. **b**, **c** The patient is placed in the prone position at operation. Note the marking on the skin for the site of coccyx and incision. **d** Tumour bed after excision of the tumour. **e**, **f** Skin refashioning and modified inverted-Y skin closure to accommodate for skin redundancy at the lower end of the wound. **g** The scar at 6-month follow-up

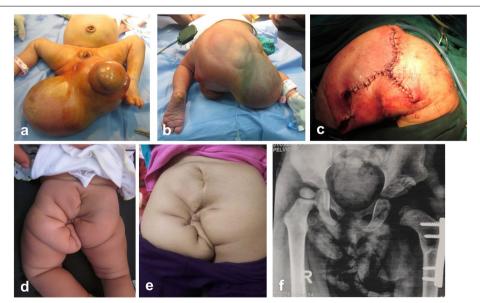


Fig. 8 Five-day-old girl presenting with large irregular protruding sacrococcygeal teratoma. **a** Patient in the supine position demonstrating the displacement of the anus. **b** The patient is placed in the prone position at operation. **c** Modified 'inverted-Y' skin closure to accommodate for skin redundancy at the lower end of the wound. **d**, **e** The scar at follow-up 6 months and 2.5 years, respectively. Note the less satisfactory cosmesis in this case mainly on the left side. **f** Plain x-ray of the pelvis demonstrating orthopaedic procedure to manage neglected left congenital hip dysplasia

the neonatal period to be discovered later by the parents when they noticed persistent limping of their child. Delayed orthopaedic management for this child was performed at the age of 2.5 years (Fig. 8f). In all cases, the anus was well located in the perineum without backward migration.

Discussion

Sacrococcygeal teratoma is a rare tumour, usually presenting in the neonatal period [1, 2]. Early excision is indicated for fear of malignant transformation [6]. Regular follow-up every 3 months is recommended for at least 3 years after tumour resection for early detection of possible recurrence [17]. Overall, the prognosis is very good, and a high survival rate can be expected [3, 6]. The benign nature of most tumours and the high survival rates would emphasise on the importance of both cosmetic and functional outcomes.

Few (if any) modifications have been made on the original description by Gross et al. for excision of sacrococcygeal teratoma [18]. Beside the 'less satisfactory' transverse scar of the chevron incision [13], we have a major concern on the transverse closure of the pelvic floor muscles after excision of the tumour [12]. Classically, the levator sling around the anorectum is sutured to the perichondrium of the anterior of the sacrum; this would distort the normal parasagittal arrangement of pelvic muscles and affect the process of normal defecation [12]. In addition, abnormal fixation of the anus in that manner may be responsible for a severely distressing complication when the anus migrates up to the back of the patient [19, 20].

In this report, we present our experience with shifting to vertical wound closure after sacrococcygeal teratoma excision over the past ten years. Obviously, the vertical midline scar has an aesthetic advantage over the transverse scar of the chevron incision [11-13]. Not only for its hidden site at the natal cleft, but also because it offers less disturbance to the normal development of the buttocks. Even in extreme cases with huge irregular projecting tumours, vertical wound closure was feasible by some modification at the lower end of the wound to accommodate for skin redundancy [12, 13]. The initial skin incision should be modified according to the size and protrusion of the tumour. For small and medium-sized tumours, a vertical midline or elliptical incision is used. For large and bizarre tumours, the initial incision may vary to accommodate for irregular tumour extension with skin involvement; however, we stress on the advantage of closing the wound vertically (Fig. 2b). In either case, we did not find extra limitations at surgical exposure or dissection that would affect the oncologic principles of tumour resection. One expected limitation for midline skin closure is poor healing, which was manifested by wound complications in about 14% of cases. However, even when healing occurred by secondary intention, still the scar was perfectly hidden in the natal cleft (Fig. 5i).

Although the study is lacking long-term follow-up, the intermediate and short-term outcomes were quite satisfactory in almost all cases. Exceptionally, the scar was less satisfactory in a single case that might have been aggravated by associated comorbidity (congenital hip dysplasia). In a previous report [21], congenital hip dysplasia may associate with sacrococcygeal teratoma in about 4%. In our case, the diagnosis of the skeletal anomaly was missed initially with delayed management at the age of 2 years, this might have affected the normal development of the buttocks on that side with less satisfactory cosmesis.

Conclusion

Vertical wound closure was always feasible after excision of sacrococcygeal teratomas. Even with large and irregular tumours, the vertical scar was perfectly or partially hidden within the natal cleft. Usually, there was adequate buttock development with minimal disturbance to the normal anal location within the perineum.

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None

Authors' contributions

Data acquisition, analysis, and interpretation were performed by all authors. AAA made the drafting of the manuscript. All authors have read and approved the final manuscript.

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Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

A written parental consent was taken before operation in all cases. Owing to the retrospective nature of the study, an IRB number was not required, and the study was approved through expedited review by the scientific/ethical committee of the Pediatric Surgery Department (Faculty of Medicine, Ain-Shams University).

Consent for publication

Patient identity did not appear in any part of the manuscript; therefore, consent for publication was not required.

Competing interests

The authors declare that they have no competing interests.

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