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Enhancing safety of laparoscopic vascular control for neonatal sacrococcygeal teratoma

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Abstract Life-threatening bleeding is a hazard of major tumor excision in children. However, fatalities from inadvertent arterial ligation should not be overlooked. Sacrococcygeal teratoma is the commonest neonatal tumor. Laparotomy to ligate the median sacral artery has been used to preempt potentially fatal resectional bleeding. Use of laparoscopy to achieve the same is an evolving technique, with only 7 neonatal cases described. As such, the Idea, Development, Exploration, Assessment, Long-term study (IDEAL) guidelines on surgical innovation recommend case reports addressing proof of concept, technical factors and safety tips. Fortunately, mistaken arterial division is so far unreported during laparoscopic median sacral artery ligation. However, as uptake widens, anatomical distortion by tumor and surgeon disorientation at endosurgery are risk factors for even such inconceivable complications. We report a successful case of laparoscopic vascular control for neonatal sacrococcygeal teratoma and demonstrate an observation that serves as a useful safety check for this procedure (as well as the open alternative).

Life-threatening bleeding is a risk of major tumor excision in children [1]. Less publicized are potentially fatal outcomes from inadvertent ligation of major vessels such as the celiac, superior mesenteric, or iliofemoral arteries [2]. However, the frequency of iatrogenic arterial injuries may be rising in children and adults [3]. Sacrococcygeal teratoma (SCT) is the commonest neonatal tumor [4]. Its vascularity may cause cardiac failure, coagulopathy, and fatal bleeding during resection [5,6]. Laparotomy to control the aorta and/or ligate the median sacral artery has therefore been used to preempt excisional bleeding [1,7]. Use of laparoscopy to achieve the same, while sparing the need for laparotomy, has been described in 7 neonatal cases [8-12]. Hence, the Idea, Development, Exploration, Assessment, Long-term study (IDEAL) guidelines on surgical innovation categorize laparoscopic median sacral artery ligation as stage 1 (Idea) or perhaps 2a (Development): with few surgeons and patients currently engaged, case reports are recommended to address proof of concept, technical factors, and safety tips [13]. At this early stage, mistaken vessel ligation during laparoscopic median sacral artery ligation is thankfully unreported. However, as uptake widens, anatomical distortion by tumor and surgeon disorientation by endosurgery remain risk factors for apparently inconceivable complications. We report a successful laparoscopic ligation of the median sacral

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artery before excision of a sizeable ulcerated SCT in a neonate, avoiding both laparotomy and transfusion. We present an observation that can be readily adopted as part of a "checklist" to enhance the safety of laparoscopic median sacral artery ligation for SCT (and can be used for the open alternative, too).

1. The case

Admitted on the day of birth, our patient was born at 37 weeks of gestation and weighed 2230 g including a 780-g sacrococcygeal mass. Distally, the skin over the mass was ulcerated (Fig. 1A). High-output cardiac failure, coagulopathy, and other anomalies were not present. Given the ulcerated tumor and its attendant infection risk, we pushed forward with a magnetic resonance scan, without anesthesia, on the evening of birth.

Fig. 1 A, Fixed mottling of the spontaneously ulcerated SCT supervened after median sacral artery ligation. B, A laparoscopic view of the median sacral artery (outlined by dotted lines) running along the white sacrum (arrowed).

This demonstrated both the heterogenous external mass comprising both solid and cystic components as well as abnormally increased signal reported to extend along the presacral space and suggestive of a pelvic component. Serum α-fetoprotein and β-human chorionic gonadotrophin levels were 85,000 IU/mL and 19 mIU/mL, respectively.

To examine for pelvic disease, laparoscopy was undertaken on the first postnatal day using a 5-mm umbilical optical port, 5-mm 30° telescope, and two 3-mm lateral lower quadrant instrument ports. We avoided stay sutures described for the uterus and simply used a dilator placed transanally to draw the rectosigmoid over to the patient's left [9].

Using 3-mm hook diathermy to open the mesorectum from the right-hand side, we located the enlarged median sacral artery running along the sacrum with no sign of any pelvic disease (Fig. 1B). The median sacral artery was ligated with bipolar diathermy (PlasmaKinetic, Gyrus Medical Ltd., Cardiff, UK) before desufflation and port closure. The baby was then positioned prone for excision of the posterior mass. We noted the marked change in the mass soon after division of the median sacral artery: the overlying skin appearances altered from mild hyperemia to fixed mottling (Fig. 1A). The posterior excision was completed with coccygectomy but without need for transfusion because blood loss was negligible. No other arterial inflow was encountered, but large veins draining toward the left side of the presacral area were ligated. Histology confirmed a benign SCT. Postoperative progress has been uneventful to more than a year of age, with unremarkable follow-up ultrasound and tumor markers.

2. Discussion

New surgical techniques evolve, and the importance of this process has been recognized in the IDEAL recommendations [13]. Laparoscopic approaches to neonatal SCT are a rational minimally invasive alternative to laparotomy for treatment of abdominopelvic disease and/or arterial inflow control [8-12]. At these early stages, with only 7 neonatal cases described, the IDEAL guidelines emphasize the need for case reports such as this to focus on proof of concept, technical considerations, and safety tips [13].

As an emerging procedure, the benefits of early vascular control in SCT need to be weighed against any hazards of the laparoscopic approach. It is therefore important to consider each step that can be made to enhance safety as more cases are undertaken. Aside from the hazard of trocar injury, the laparoscopic approach shares the risks of inadvertent vessel ligation seen in open tumor excisions [2]. Such devastating injuries can be fatal, particularly when patient size hampers timely revascularization. Although in our case the median sacral artery was readily identified from its midline presacral

position and visible pulsations, it is conceivable that greater abdominopelvic disease can distort the vascular anatomy as seen in other tumors [14,15]. Furthermore, introducing laparoscopic alternatives (eg, cholecystectomy) can be associated with risks of misidentification and mistaken division of key structures (eg, common bile duct) that appear highest as the technique is first adopted more widely [16]. Therefore, in using the laparoscopic approach to SCT and vascular control in particular, we need to focus on minimizing hitherto unreported but predictable hazards.

Our observation that the tumor developed fixed mottling soon after median sacral ligation leads us to a safety modification for this procedure. Firstly, in preparing the patient for surgery, we suggest that the lower limbs and sacrococcygeal mass be covered with a transparent sterile drape or encompassed within a sterile bowel bag. Secondly, we propose that once the median sacral artery is located at laparoscopy, it is temporarily occluded with a laparoscopic instrument. At this point, the surgical team can examine the lower limbs and the mass, via the transparent drape, for the fixed mottling pictured. Hence, before definitive ligation, lower limb vascularity can be ensured; and the completeness of tumor vascular control can be judged. If fixed mottling does not appear on test clamping, the surgeon can carefully check the vascular anatomy and lower limb perfusion, proceeding to arterial ligation only if completely unambiguous in their assessment; absence of fixed mottling can also alert the surgeon to possible additional arterial supplies that may be encountered during tumor excision.

In summary, we present a newborn with an ulcerated SCT who therefore underwent surgery on the first postnatal day. Laparoscopy to evaluate for pelvic disease allowed median sacral artery ligation to facilitate tumor excision without transfusion. During this, we observed fixed mottling of the tumor and suggest steps to use this observation as a "checklist" step for reducing risks of erroneous vessel ligation and judging completeness of vascular control. Sharing such modifications forms a key part of the IDEAL process for developing surgical innovations and can help promote safe surgery in children [17].

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