J Anesth (2010) 24:951–954
DOI 10.1007/s00540-010-1027-x

CLINICAL REPORT

Complications with massive sacrococcygeal tumor resection on a premature neonate

Edwin Abraham · Tariq Parray · Abid Ghafoor

Received: 15 March 2010 / Accepted: 15 September 2010 / Published online: 6 November 2010

Abstract Resection of large sacrococcygeal teratomas (SCT) in neonates can pose many anesthetic challenges. The pathophysiology of the SCT determines the varying management. We present a case report of a 34-week newborn with a massive Altman type 3 SCT. The surgery was delayed 2 days because of hyperkalemia; however, as a result of continued tumor lysis the patient's condition had worsened with little improvement of the potassium level. During the surgery, the patient had issues of bleeding needing massive transfusion. Ventilation was also difficult at times because of the massive tumor resting on the chest, resulting in respiratory acidosis. We also had difficulty in maintaining the temperature. This patient did well after the surgery and was discharged home. We address here the anesthetic issues involved in the perioperative care management of a premature infant with a massive SCT.

Keywords Sacrococcygeal teratoma · Tumor lysis · Hyperkalemia

Introduction

Sacrococcygeal teratomas (SCT) are the most common congenital neoplasm, occurring in 1 in 40,000 infants, with 95% of these occurring in females [1]. Surgical treatment involves immediate complete resection of both the tumor and coccyx [2]. Open fetal surgery, endoscopic laser ablation, and radiofrequency ablation are also being used to excise these tumors. Predictors of poor outcome include diagnosis before 20 weeks gestation, delivery before 30 weeks, development of hydrops, low birth weight, Apgar score of less than 7, malignant histotypes, polyhydramnios, and placentomegaly [2, 3].

Anesthetic management can be challenging depending on the pathophysiology of the disease. Intraoperative management of open surgery for SCT resection can be complicated by several factors such as difficulty positioning the patient in supine position to facilitate endotracheal intubation, difficulty ventilating the lungs in prone position, difficulty in thermoregulation, cardiovascular instability, associated coagulopathy, massive blood loss requiring transfusion, tumor lysis, etc. [4]. Infants can have high output heart failure, fetal anemia, and hypovolemia from rupture of the SCT. Survival rates range from 77 to 94% [1, 5].

Case report

We present a case of a 34-week premature baby with a large SCT born to a 18-year-old primigravida who had good prenatal care. SCT was seen at the first ultrasonography at 16 weeks of gestation and followed closely in a high-risk obstetrics center. Fetal magnetic resonance imaging at 33 weeks showed an Altman type 3 SCT $16 \times 15 \times 19$ cm extending from the fetal buttock and lower back muscles without evidence of hydrops fetalis. Appropriate team communication and coordination was present between the anesthesia, obstetric, neonatology, and pediatric surgery teams. Because of premature rupture of membranes, the mother underwent elective cesarean section at 34 weeks gestation. Apgar scores were 5 and 6 at 1

E. Abraham (✉) · T. Parray · A. Ghafoor
Department of Pediatric Anesthesiology and Pain Medicine, Arkansas Children's Hospital, University of Arkansas for Medical Sciences, 1 Children's Way, Slot 203, Little Rock, AR 72202, USA
e-mail: abrahamedwinj@uams.edu

and 5 min, respectively. The infant was intubated secondary to poor respiratory effort. Peripheral and umbilical venous access was obtained in the resuscitation are, and an umbilical arterial catheter was placed. The tumor sac was intact with a small area of color change suggesting intratumoral hemorrhage. The patient was stabilized for transfer to a pediatric hospital located 2 miles away.

Initial assessment and workup included an echocardiogram and chest X-ray, which were normal. The tumor was oozing, which was stopped by thrombin spray and surgical application. The patient weighed approximately 2 kg without the tumor. Initial laboratory values were remarkable for acidosis, severe hyperkalemia, and anemia requiring multiple blood product transfusions. Total preoperative transfusions of 43 ml packed red blood cells (PRBC), 24 ml platelets, and 23 ml fresh-frozen plasma (FFP) were given.

Surgery was scheduled for day 1 after birth. Laboratory results on the morning of surgery showed a white blood count of 12,000 K/μl, hemoglobin 9.2 g/dl, hematocrit (Hct) 30.5%, and platelets 89,000 after the aforementioned transfusions. Basic metabolic profile showed a sodium level of 127 mmol/l and a potassium of 7.7 mmol/l; other electrolytes were within normal range. After rechecking potassium, which was above 7.0 mmol/l, surgery was delayed for correction. The patient was clinically managed by the neonatal intensive care unit (NICU) team and scheduled to return to the operating room (OR) on day 3 for elective resection of SCT.

By day 3 the patient had deteriorated from the continuous tumor lysis. She was on dopamine at 10 μg/kg/min, received more transfusions with PRBC, platelets, and FFP; hemoglobin was 8.9 gm/dl, Hct 30%, platelets 50,000, sodium 129, potassium 6.6, and creatinine 1.4. Coagulation studies showed an international normalized ratio (INR) of 1.8, prothrombin (PT) of 17.2, and partial thromboplastin time (PTT) of 42.5. Arterial blood gas (ABG) showed metabolic acidosis with pH of 7.21, CO_2 of 48 mmHg, O_2 of 88 mmHg, and bicarbonate of 19 mEq/l with a base excess of –5 mEq/l. The decision was made to proceed with surgery because of the deteriorating condition of the patient.

The patient was transferred from the neonatal intensive care unit (NICU), intubated, ventilated, with monitoring lines [consisting of an umbilical arterial and venous catheter, infusions and one peripheral intravenous (PIV) line in her left hand]. Vital signs on arrival to the OR were within the normal range with pressors, and her oxygen saturation was 100% on 40% FiO_2. During transfer to the OR, the cystic component of the SCT ruptured; however, the patient was hemodynamically stable. The patient's endotracheal tube was connected to an Aestiva anesthesia machine, and the NICU ventilator settings were continued with pressure control of 18 mm Hg, tidal volume of 25 ml, and respiratory rate of 40. Standard American Society of Anesthesiologists monitors were placed, and monitoring lines continued to be transduced in the OR. A fentanyl dose of 1 μg/kg and rocuronium 2 mg were given initially, and two more 22 gauge PIV were placed in the bilateral saphenous veins. Anesthesia was maintained with isoflurane at 0.4% and fentanyl bolus. Total peripheral nutrition (TPN) was continued throughout surgery at 2 ml/h, and morphine infusion of 30 μg/kg/ml was stopped. Intraoperatively, dopamine was maintained at a continuous infusion of 10 μg/kg/min. The patient was then positioned prone for the procedure. A force warming device, hot towels, and an intravenous fluid warmer were used to keep the patient warm.

Normal saline was infused with a careful watch on serum potassium levels. We used hyperventilation and sodium bicarbonate, which brought the potassium level to 5.0 mmol/l. ABGs were measured every 30 min–1 h of the 3-h procedure. The solid component measured 18 cm by 20 cm (Fig. 1). The patient was having bouts of difficult ventilation because of increased peak pressures. There were frequent periods where she had to be bag-ventilated because the large tumor was resting on her chest during the resection. Intraoperative ABGs showed a pH of 6.87, CO_2 of 120 mm Hg, O_2 of 83 mmHg, bicarbonate of 23 mEq/l, and base excess of –11.2 mEq/l. Hemoglobin dropped to 4.3 and Hct to 13.6%. All the electrolytes were within the normal range except calcium, which fell to 0.60 mmol/l after the blood transfusion. The patient was given a total of 15 mEq sodium bicarbonate and 50 mg calcium gluconate during the surgery.

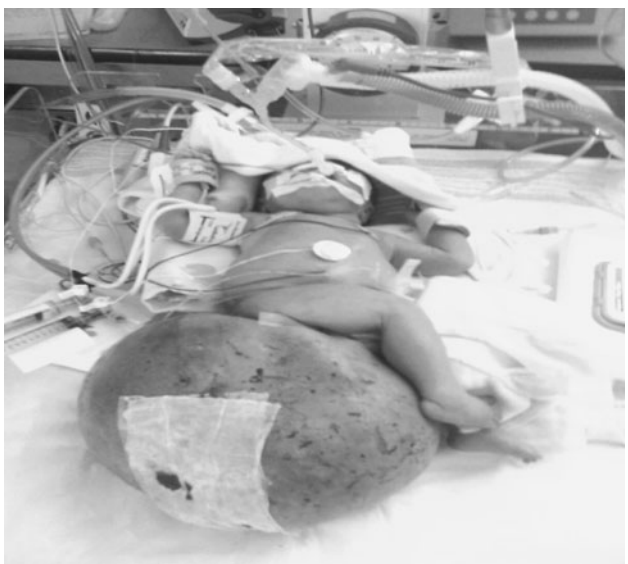

Fig. 1 Premature neonate with a large sacrococcygeal teratoma

Systolic blood pressure was maintained between 50 and 80 mmHg without increasing dopamine. After the SCT was resected, there was a period of diffuse oozing that took some time to resolve. Total blood loss was almost one blood volume, estimated to be between 150 and 200 ml. The patient received a total of 200 ml normal saline, 150 ml PRBCs, 60 ml FFP, and 40 ml platelets. The final ABG after surgery was completed showed a pH of 7.07, CO_2 of 77 mmHg, O_2 of 196 mmHg, bicarbonate of 19 mEq/l, and base excess of –8.9 mEq/l. The postoperative hemoglobin was 7.8, and Hct was 24%. All the electrolytes were within the normal range. Even with our measures to keep the patient warm, her temperature dropped to 34°C.

The patient was transferred to the NICU in stable condition. She was extubated on postoperative day (POD) 19, weaned to room air on POD 24, and discharged home in good condition on POD 40.

Discussion

This case shows the importance of early surgical treatment for SCT. After the tumor was resected, the patient improved drastically. The main causes of death during resection of large SCTs include hemorrhage, hypothermia, coagulopathy, extensive transfusion with blood products causing electrolyte abnormalities, and inability to provide enough cardiopulmonary support during the intraoperative manipulation of the tumor [3].

The mainstay treatment for SCT is surgical resection. Prenatal diagnostic studies include ultrasound scan, magnetic resonance imaging, and echocardiography The majority of SCT are not malignant and have good prognosis after resection. The fetus is monitored by serial ultrasound, and if there are no signs of fetal hydrops or high output cardiac failure, elective delivery by cesarean section is planned when the fetus is mature. If the SCT is small, vaginal delivery may also be possible. Resection of the SCT is done soon after the neonate is stabilized and evaluated [6].

If fetal hydrops is present with increased high output cardiac failure, the risk of survival diminishes significantly. If the fetus is mature, emergency cesarean is performed with possible resection of the SCT. Other potentially lifesaving interventions include aminoreduction, cyst aspiration, surgical debulking, and fetal surgery for the immature fetus [6].

Preoperative evaluation is critical because infants with SCT have a 5–25% chance of having other associated anomalies, which include pulmonary hypoplasia, meconium peritonitis, rectal atresia, renal complications, and meningocele [6]. This patient was initially stable but by day 3 was requiring dopamine infusion. Even though the hyperkalemia was dangerous, she had no EKG changes, and in retrospect surgery could have proceeded with an intention to treat clinically significant hyperkalemia; however, cardiac arrest secondary to hyperkalemia has been reported in SCT resection [7]. The 2 days of management in the NICU to control the hyperkalemia produced little change in the potassium level but worsened her hemodynamic status due to the continuous hemorrhaging of the tumor.

Frequent blood gas monitoring alerted us to give adequate blood products during hemorrhage. Bleeding was controlled with fresh-frozen plasma (FFP) and platelets in our patient. Having two saphenous IV lines allowed us to give large fluid boluses quickly during the periods of hemorrhage. These patients are more likely to develop coagulopathy. Disseminated intravascular coagulation (DIC) or dilutional coagulopathy and thrombocytopenia from massive transfusion could occur [1]. In a study published in the *European Journal of Pediatrics*, a report recommended factor VIIa for management of refractory hemostasis [8, 9]. One case report describes using initial devascularization with staged resection in a 26-week premature infant to manage blood loss [10].

Even with forced warming devices, wrapping the patient with warm towels, and an intravenous fluid warmer to warm the infusions, it was extremely difficult to keep the patient warm because of the large surface area of the SCT that was exposed. Hypothermia could also worsen coagulopathy, increase metabolic consumption if not paralyzed, and prolong the effects of the anesthetics.

There were periods of time intraoperatively when ventilation was difficult because of the tumor resting on the chest. The ventilator was not able to generate enough pressure to fill the lungs. We had to hand-ventilate with pressures of >30 mm Hg. The surgeons would lift the tumor during these periods to help with the ventilation. The ventilation improved after the tumor was resected. However, by the end of the surgery the patient had developed a significant respiratory acidosis, with the CO_2 rising to 77 mmHg.

Tumor lysis leading to extreme hyperkalemia can be a lethal complication. Although rare for solid tumors, there was a case report in the *Journal of Perinatology* of lethal hyperkalemia in a neonate due to progressing tumor necrosis [11]. After attempting to manage the hyperkalemia, emergency resection of the tumor was needed; however, that patient died of cardiac arrest. From our observation in our case, it seems that delaying the surgery to manage the hyperkalemia only made the patient's condition worse. We suggest managing the hyperkalemia (calcium gluconate, $NaHCO_3$, albumin, insulin/glucose, or Kayexelate) in the operating room during resection.

This case highlights the importance of understanding neonatal physiology and being able to clinically manage cardiovascular instability, massive blood transfusion, hypothermia, and coagulation dysfunction in the operating room. We are happy to report that the patient recovered and was discharged home.

Conflict of interest None.

References

1. Tran K, Flake A, Kalawadia N, Maxwell L, Rehman M. Emergent excision of a prenatally diagnosed sacrococcygeal teratoma. Paediatr Anaesth. 2008;18:431–4.
2. Perrelli L, D'Urzo C, Manzoni C, Pintus C, Santis M, Masini L, Noia G. Sacrococcygeal teratoma outcome and management. An analysis of 17 cases. J Perinat Med. 2002;30:178–84.
3. Hase T, Kodama M, Kishida A, Shimadera S, Aotani H, Shimada M, Yamamoto Y, Noda Y, Okabe H. Techniques available for the management of massive sacrococcygeal teratomas. Pediatr Surg Int. 2001;17:232–4.
4. Galinkin J, Schwarz U, Motoyama E. Anesthesia for fetal surgery. In: Smith RM, editor. Anesthesia for infants and children. 7th ed. St. Louis: Mosby; 2006. p. 555–6.
5. Rescorla F, Sawin R, Coran A, Dillon P, Azizkhan R. Long-term outcome for infants and children with sacrococcygeal teratoma: a report from the Childrens Cancer Group. J Pediatr Surg. 1998;33:171–6.
6. Hedrick H, Flake A, Crombleholme T, Howell L, Johnson M, Wilson R, Adzick N. Sacrococcygeal teratoma: prenatal assessment, fetal intervention, and outcome. J Pediatr Surg. 2004;39:430–8.
7. Reinoso-Barbero F, Sepulveda I, Perez-Ferrer A, De Andres A. Cardiac arrest secondary to hyperkalemia during surgery for a neonatal giant sacrococcygeal teratoma. Paediatr Anaesth. 2009;19:712–4.
8. Fadler K, Askin D. Sacrococcygeal teratoma in the newborn: a case study of prenatal management and clinical intervention. Neonatal Netw. 2008;27:185–91.
9. Girisch M, Rauch R, Carbon R, Habash T, Hofbeck M. Refractory bleeding following major surgery of a giant sacrococcygeal teratoma in a premature infant: successful use of recombinant factor VIIa. Eur J Pediatr. 2004;163:118–9.
10. Robertson F, Crombleholme T, Frantz I, Shephard B, Bianchi D, D'Alton M. Devascularization and staged resection of giant sacrococcygeal teratoma in the premature infant. J Pediatr Surg. 1995;30:309–11.
11. Jona J. Progressive tumor necrosis and lethal hyperkalemia in a neonate with a sacrococcygeal teratoma. J Perinat Med. 1999;19:538–40.