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Infantile Sacrococcygeal Teratoma - intraoperative Cardiac Arrest due to Electrolyte Imbalance

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Introduction

Sacrococcygeal teratoma (SCT) is the most common congenital neoplasm of newborn with a reported incidence of 1 in 35000 live births.[1-3] The first large series of infants and children with SCT was reported by Gross et al.[4] They recommended complete surgical excision of tumour enbloc with the coccyx, which remains the treatment of choice today.

Clinical reviews of SCT include several mortalities due to exsanguinating hemorrhage during surgery.[5-9] No study has focused on the complications occurring due to electrolyte imbalance in patients with bleeding problems, associated with these neoplasms.

We are reporting a case of neonate who was operated for excision of SCT, had preoperative coagulopathy and bleeding into the tumour. This neonate had intraoperative prolonged Q-T interval on ECG followed by cardiac arrest, and was resuscitated successfully.

Case Report

Fifteen hours old, 2.7 Kg, full term, normally delivered male neonate who was otherwise active and moving all four limbs, was presented in emergency, with a single huge mass approximately 14 x 15 centimeters extending on to both buttocks (Figures 1 and 2). Tumour was generally cystic in consistency but firm at places with breaching epithelium underlying hemorrhage and echymosis. Preoperative investigations showed Haemoglobin 16.0 gm/dl, Haematocrit (Hct) 47.6, Platelets 96 x 109/lit, prothrombin time 16.3 seconds (control 12 Seconds), activated partial thromboplastin time 62 seconds (control 30 Seconds) with INR 1.6, magnesium 1.5 meq.lit (1-2 meq.lit), Calcium, 8.2 mg/dl (10mg/dl) Phosphate 9.8+ mg.dl (4.4 - 6.6 mg.dl), Sodium 132 mmol.lit, potassium 4.8 mmol.lit and Lactic Dehyodrogenase (LDH) 2735 IU.lit (253 - 548 IU.lit). In next 12 hours patient dropped his Hemoglobin from 16 gm. dl to 12.3 gm.dl and Hct 37.0, which was replaced with packed cells and coagulation was corrected prior to surgery by Fresh Frozen Plasma (FFP). Surgery was performed 27 hours after delivery. Inhalational induction was done with Sevoflurane, Atracurium was used as muscle relaxant and Fentanyl as analgesic. Intraoperative monitoring included ECG, heart rate, noninvasive blood pressure, End-tidal CO_2, Pulse oximetry , temperature, and oesophageal stethescope. Surgery lasted for about two hours in prone position. Intraoperative blood loss was approximately 135 ml. which was replaced by 120 ml.of packed cells, 50 ml. of FFP and 50 ml. of crystalloid. Near the end of surgery, the patient developed prolonged Q-T interval on ECG followed by ventricular tachycardia. The situation was managed immediately by turning the neonate in supine position and external cardiac massage was started. During CPR, he received adrenaline, bolus of normal saline and calcium gluconate. Normal sinus rhythm with good cardiac output was reestablished within 5 minutes. In the immediate post resuscitation period, he developed seizures activity for which he received valium and was loaded with phenytoin sodium. He was electively ventilated in Neonatal Intensive Care Unit (NICU) for 24 hours and then extubated successfully. Post extubation the patient remained stable with no obvious neurological deficit and was discharged on the eighth postoperative day.

Blood sample which was taken in the immediate post resuscitation period and sent for laboratory investigation showed hypocalcemia (Table) despite of calcium gluconate administration during CPR.

Figure 1. A 2.7 kg male, neonate with 14 x 15 centimeters sacrococcygeal teratoma.

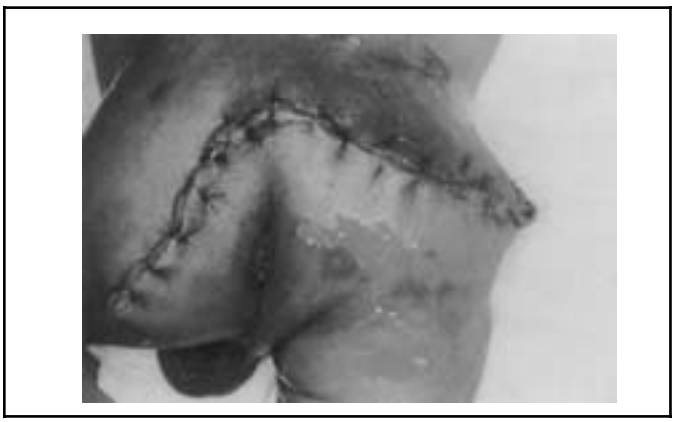

Figure 2. Neonate after operative removal of tumor as in Figure 1.

Discussion

Despite extensive literature on SCTs, little attention has been focused on the associated electrolyte and hemorrhagic complications. Most reviews include mortalities secondary to exsanguinating hemorrhage. In the American Academy of Pediatrics Surgical Section Survey 1973 (AAP), Altman et al.[5] reported a 6% mortality amongst 247 patients presenting in the first month of life. Several mechanisms have been postulated to explain this high mortality including hemorrhage into the tumor[10], obstruction of umbilical vessel flow[11] and high output heart tfailure.[11-12] Non-neoplastic causes of cardiac failure such as fetal cardiac rhythm disturbances [13,14], valvular disease[15] or electrolyte imbalance may produce similar physiological derangement. In Grosfeld et al's review of 41 neonates with SCTs [8], there were 4 deaths due to intraoperative bleeding. Noseworthy et al[9] reported 4 mortalities secondary to massive hemorrhage in a group of 78 patients with mature SCT and excessive transfusion requirements in neonates undergoing resection of SCTs especially those with immature histology. Dewan et al[7] reported one death due to overwhelming hemorrhage in their review of 21 neonates with SCTs. Smith et al[16] noted that blood loss in the large, even benign tumors can be substantial, reaching the patients blood volume. Daniel et al[17] showed that patients were extremely unstable intraoperatively which was explained due to high output cardiac overload. In our case, patient dropped his Haemoglobin from 16 gm/dl to 12.3 gm/dl within 12 hours even after replacement of packed cells and FFP, due to spontaneous bleeding and oozing from the tumor. He also had high phosphate levels and low normal calcium prior to surgery which may be due to tumor lysis. Intra-operative blood loss was about 50 ml/kg (135 ml) for which he received packed cell and FFP throughout the surgery. Near the end of surgery patient showed prolongation of QT interval on ECG followed by ventricular tachycardia. The event of cardiac arrest which was preceded by prolonged QT interval may be due to the rapid transfusion of blood products resulting in elevated plasma levels of Potassium, citrate and low level of ionized calcium[18] which was subsequently confirmed by laboratory Test (Table).

Table. Laboratory investigations.

Laboratory (Normal Values)	Pre-operative	Post-operative
Haemoglobin 16.5 ± gm.dl	12.3	11.6
Haematocrit 48 ± 3	36.4	35.7
Platelets 150-400 ($x10^9$ lit)	96	74
$Ca+^2$ 10 mg/dl	8.2	6.0
$K+^2$ 3.5-5.5 (meq.L)	4.8	4.4
Po^4 4.4-6.6 (mg/dl)	9.0	3.7*

*48 hours postoperatively.

Apart from electrolyte imbalance related to blood transfusion, Tumor lysis and rhabdomyolysis can also cause hypocalcemia and hyperkalemia by increasing the plasma concentration of anions that chelate free calcium.[19] The phosphate released from cells into the circulation in these condition complexes with plasma ionized calcium and lowers the ionized calcium concentration. Citrate used in storage of blood products prevents blood coagulation by chelating free calcium. The rapid transfusion of blood products can result in elevated levels of potassium[20,21] and citrate can cause ionized hypocalcemia. The severity of hyperkalemia and ionized hypocalcaemia varies depending upon the transfusion rate, but the duration of hypocalcemia is usually transient, (less than 10-15 minutes), or the time it takes for the body to metabolize the citrate load.[22,23] Tumor lysis is known among patients undergoing induction therapy and Lymphocytic malignancies. The phenomenon of tumor lysis in infancy has been described only once, in association with surgical manipulation of hepatoblastoma. Jona reported a case of newborn with SCT who experienced spontaneous tumor lysis induced hyperkalemia.[24]

Clinical manifestation of hypocalcaemia and hyperkalemia is the depression of cardiovascular function, which can be clinically important in patients under anesthesia, in intensive care or with underlying heart disease. Prolongation of QT interval on the ECG correlates well with progressive ionized hypocalcaemia. Ionized hypocalcaemia and hyperkalemia can manifest as hypotension, cardiac failure and arrhythmias that do not respond to vasoactive agents or volume expansion.[18]

The neonates with sacrococcygeal teratoma who present with hemorrhagic complication and coagulopathy may require large blood transfusion. Frequent intraoperative assessment and timely correction of potassium and calcium abnormalities avoid this fatal complication.

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