
Case report

**ANAESTHETIC CONFRONTS IN MANAGEMENT OF PEDIATRIC TERATOMAS:
AN UNIQUE CASE SERIES***Renuka R¹, Venkatachalapathy TS²*¹Department of Anesthesiology, Sapthagiri Institute of Medical Sciences and Research Centre, Chikkasandra, Bangalore, India²Department of General surgery, Sapthagiri Institute of Medical Sciences and Research Centre, Chikkasandra, Bangalore, India

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Financial Disclosures: None**Conflicts of interest :** None**RUNNING TITLE:** Anesthetic management of pediatric teratomas**Authors contributions :****Dr. Renuka R has helped** in designing , conducting , drafting the work and article .**Dr. Venkatachalapathy T S has helped** in designing , conducting the work and article .

Abstract

Teratomas are bizarre neoplasms that most commonly occur in infancy and childhood. Since surgical therapy is the primary treatment modality for any type of teratoma, we as anesthesiologists encounter these cases when they come for surgical removal. Providing anesthesia services for these cases is challenging because of the age at presentation, the complexity of the surgical condition, the inherent anomalies associated with it, and also the existing literature gaps in anesthesia management for these cases. In order to close this knowledge gap and to support future anesthesia planning, this case series was framed.

Key words: Teratoma; regional anesthesia; sacrococcygeal; gastric; retroperitoneal; anti-NMDAR Encephalitis.

GLOSSARY OF TERMS

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| 1) ASA – American society of anesthesiologists | 6) OPD -Out patient department |
| 2) AFP - Alphafetoprotein | 7) CT- Computed tomography |
| 3) SCT- Sacrococcygeal teratoma | 8) β -HCG – Beta human chorionic gonadotrophin |
| 4) OETT- Oral endotracheal tube | 9) HPE- Histopathological examination |
| 5) 2D ECHO – Two dimensional echocardiography | |

Introduction

The word Teratoma is derived from the Greek word teraton, which means monster. Teratomas are relatively common embryonic neoplasms that arise from totipotent cells and contain elements that represent all three germ layers. Teratomas can be found in almost any organ, but tend to develop more frequently in the midline or paraxially and are almost always considered foreign to the anatomical region in which they are found. In infancy and childhood, the most common teratomas

in the sacrococcygeal region (60-65%), gonadal (10-20%), mediastinal (5-10%), presacral (5%) are rarely intracranial, retroperitoneal, cervical and gastric (<1%) .¹

Clinical diagnosis of teratoma can be challenging because the disease occurs in unusual locations and has unusual presentations that almost always require correlation with radiological, laboratory, and histopathological findings to confirm the diagnosis. Since pure teratomas have the potential to become malignant and malignant teratomas²⁻⁵ to metastasize, it is very important to make an

accurate early diagnosis and proceed with treatment. The definitive therapy for any type of teratoma is surgical excision, and the prognosis is excellent with complete excision of the lesion. Adjuvant chemotherapy or radiation therapy is recommended only if there are features suggestive of recurrence or malignancy following surgical excision, as suggested by clinical radiological examination and serum markers, which are quite rare. With the priority of the surgical procedure over other adjuvant measures in tumor treatment, we as anesthesiologists very often encounter these cases in order to ensure anesthesia during these surgical interventions. Although it is a very common pathology that requires surgical excision, there is little literature on anesthesia management for these surgical procedures. The aim of this case series, therefore, is to address the perioperative concerns that are unique to the anesthesia management of different types of teratomas to aid future anesthesia planning. Written parental consent was obtained for their children to be included in these case series and for publication.

Case description

This case series includes 6 cases with the diagnosis of teratoma in the newborns up to 16 years of age who underwent surgical tumor removal under anesthesia in our facility between August 2018 and January 2021. Intraoperative monitoring according to the standards of the American Society of Anesthesiologists was followed. Since the tumors are highly vascular, 2 large-caliber intravenous cannulas for fluid resuscitation and, where necessary, invasive lines were secured.

Adequate blood and blood products were readily available and all resuscitation equipment with emergency medication was kept ready in the operating room (Table 1, Table 2).

Discussion

Gastric teratomas are extremely rare and were first reported by Eusterman and Sentry in 1922.⁶ About 94% of the reported cases were in male infants or newborns. Typical symptoms are abdominal lumps, distension, and vomiting, but intramural tumors with gastrointestinal bleeding and gastric perforations have also been described in

the literature.⁷ They manifest as palpable masses in 75% and abdominal distension in 56%.² Other rare manifestations are secondary to peritonitis after gastric perforation and tumor rupture. Most of these typical findings were observed in our case (Table 1, Figure 1). In addition, the newborn had 2 seizures with a hemoglobin value of 4.9 g / dl on admission and was on a dopamine infusion at 10 µg / kg / min to maintain hemodynamics due to persistent hematemesis. Since we could not find any evidence in the literature for anesthesia management of gastric teratomas, our anesthesia management (Table 2) was based predominantly on the precautions to be taken in view of the critical condition of the patient, the 2D ECHO findings and the symptoms presented. Therefore, it is imperative to add more evidence of anesthetic management of gastric teratomas to the literature to aid future planning.

Sacroccygeal teratoma (SCT) is the most common tumor in the neonatal period with an incidence of 1 in 35-40 thousand live births and a female to male ratio of 4:1.⁸ Associated congenital malformations are observed in 12-15% of cases and occur more often with presacral tumors. Perioperative concerns related to this case management include position related circulatory and respiratory distress, the possibility of venous air embolism (VAE)⁹ due to the opened sinuses with resection of a large vascular tumor, surgical injuries that cause damage to the pelvic nerve leading to bowel and bladder dysfunction, and an alarming complication of tumor lysis that leads to cardiac arrest. In anticipation of all of these reasonable precautions have been taken. Careful anesthetic (Table 2) and surgical management with close monitoring followed which resulted in successful management of the case. Our case management differed from others in the choice of a side position (Figure 2) to secure the airways in order to avoid injury to the sacral mass, which is a major challenge in this age group.

Retroperitoneal teratomas are rare entities that make up 3.5-4% of all germ cell tumors in children and have a female to male ratio of 2: 1. The incidence is bimodal, with peaks in the first 6 months of life and early adulthood. Because of their location, they are usually not identified until they have grown to gigantic proportions. Retroperitoneal teratomas are the third most common primary

TABLE 1: Preanesthetic evaluation

Serial no	Age/ Gender/ Weight	History	GPE	Systemic Examination	Lab Investigation	Radiological investigation	Clinical Diagnosis
Case 1	1 Month Male 2Kg	Birth history: Preterm baby delivered by LSCS C/C: Abdominal distension, Black coloured vomitus and stools since 2 days. 2 episodes of seizures	Pallor present	P/A examination: Distended, Globular shape, Veins on abdomen prominent. Abdominal girth 11 cms. Abdominal mass felt, Bowel sounds heard	Hemoglobin -4.9g/dl increased to 6.8 g/dl after 90 ml of blood transfusion 2D ECHO: Mild TR, Borderline PAH, PASP -30mmHg	USG abdomen and pelvis Heterogenous mass of 10.8x 8.8x6.4cm	Gastric teratoma? ? Neuroblastoma
Case 2	2Month Female 5 Kg	Birth history: FTNVD C/C: Swelling over lower back since birth	NAD	Local examination: sacral mass of size 14 cm x 12 cm involving coccyx extending to both buttocks	2D ECHO: PFO / OS-ASD, Left to Right shunt, Trivial TR	USG KUB: 11 x7 cm solidcystic lesion in presacral region and perineal region suggestive of Sacral teratoma	?Sacrocoycegal teratoma
Case 3	13 Years Female 22 Kg	Birth history: FTNVD result of consanguineous marriage C/C: Pain abdomen and vomiting on and off since 4 months Abdominal distension since 1 week	Mental retardation present, Deaf and mute. Down syndrome facies present	P/A examination: Mass per abdomen occupying lower abdomen and umbilical region, nodular surface, mass mobile on transverse axis, lower border cannot be made out	WNL Karyotyping - Downs syndrome	USG Abdomen and pelvis: Large heterogenous lesion measuring 12x13 cm with both solid and cystic components is seen arising from pelvis	Retroperitoneal teratoma? Germ cell tumor ovary?
Case 4	13 Years Female 35 Kg	C/C: Pain abdomen and mass per abdomen since 2 months. Menstrual H/O: attained menarche 2 months back	NAD	Per abdominal examination: Tenderness over right iliac fossa and hypogastrium present No guarding or rigidity	WNL	USG Abdomen and pelvis: Thin walled cystic lesion in lower abdomen of 10x4 cm size Right adnexal mass of 4.1 x 4.3 x 3.9 cm	Right ovarian an cyst with torsion
Case 5	16 Years Female 35 Kg	C/C: Pain in right loin and vomiting since 3 days. Past H/O: underwent left ovarian cystectomy an year back	NAD	Per abdominal examination: Tenderness present over right iliac fossa	WNL	USG Abdomen: Thick walled cystic lesion measuring 8.1 x 7.6 cm in right ovary ? Germ cell tumor	Right ovarian cyst? ? Germ cell tumor
Case 6	16 Years Female 50 Kg	C/C: Pain abdomen and fever on and off since 2 months	NAD	P/A examination: Soft, no distension, Tenderness present over hypogastrium, no guarding / rigidity	WNL	USG Abdomen & Pelvis: Thin walled cystic lesion in lower abdomen of 10.3 x 10.8 cm -? Germ cell tumor MRI pelvis: Mesenteric cyst of 10.1 x 5.8 x 8.3 cm	?Left paraovarian cyst or hydrosalpinx

H/O- History; C/C- Chief complaints; FTNVD - Full term normal vaginal delivery; NAD - No abnormality detected; USG- Ultrasonography; MRI - Magnetic resonance imaging; WNL - Within normal limits

TABLE 2: Perioperative course

Serial no	Surgical procedure and I/O finding	Anesthetic management	Post operative period	Histo pathological report
Case 1	Emergency laparotomy and resection of tumor Finding: Tumor arising from greater curvature and posterior wall of stomach with endogastric invasion. Duration: 90 minutes	General anesthesia with controlled ventilation Rapid sequence induction Intubated with 3.0mm plain portex OETT Maintenance: 50% O2 in air with Sevoflurane (1-2%) Left femoral central venous catheter secured Dopamine infusion titrated to response, 70 ml PRBC and 20 ml of FFP to maintain hemodynamics Decided for postoperative elective ventilation Blood loss: 30 ml Urine output: 20 ml	1st POD 60 ml of FFP and 30 ml PRBC transfused 2nd POD Dopamine infusion stopped 3rd POD Extubated 15th POD Discharged	Immature gastric teratoma grade 3 stage 1
Case 2	Surgical excision of tumor with coccygectomy Finding: Sacrococcygeal teratoma Altmann's type 2 Duration: 120 minutes	General anesthesia with controlled ventilation Inhalational induction After confirming for mask ventilation, succinylcholine used to facilitate intubation Intubation performed in lateral position with 3.5mm ID plain portex OETT to avoid injury to the sacral mass Analgesia – intravenous paracetamol, opioids, Caudal was not given due to the mass in sacral region Extubated on table Blood loss: 80ml managed with crystalloids	4th Post operative day : Discharged	Mature teratoma- sacrococcygeal region
Case 3	Laparotomy and tumor excision Finding: Left ovary replaced by huge tumor occupying pelvis in relation to ovary and rectum. Tumor ruptured due to fragile wall. Duration: 120 minutes	Combined spinal epidural anesthesia SAB - with 2ml of 0.5% Bupivacaine (Heavy) Epidural - combination of lignocaine and bupivacaine administered in increments to achieve the desired sensory block level with minimal hemodynamic disturbances Blood loss: 250ml replaced with PRBC	7th Post operative day : Discharged and advised chemotherapy AFP-1210ng/ml Received 6 cycles of PEB chemotherapy On further follow up with CT and AFP levels – no features of relapse or recurrence detected	Malignant mixed germ cell tumor with yolk sac components.
Case 4	Emergency laparotomy Finding: Right ovarian cyst with torsion of 6 rotation with gangrenous changes Duration: 80 minutes	Combined spinal epidural anesthesia SAB - with 2ml of 0.5% Bupivacaine (Heavy) Through Epidural catheter - combination of lignocaine and bupivacaine administered in increments to achieve the desired sensory block level with minimal hemodynamic disturbances	5th Post operative day : Discharged	Immature teratoma (incidental finding)
Case 5	Laparotomy and tumor excision Finding: Left ovarian cyst with torsion, cyst replacing the entire ovary Duration: 90 minutes	Combined spinal epidural anesthesia SAB - with 2.5 ml of 0.5% Bupivacaine (Heavy) Through Epidural catheter - combination of lignocaine and bupivacaine administered in increments to achieve the desired sensory block level with minimal hemodynamic disturbances	5th Post operative day : Discharged	Immature teratoma (confirmatory finding)
Case 6	Laparotomy and tumor excision Finding: Left paraovarian cyst of 10x5 cm with fibrial end adherent to cyst and ovary separate from the cyst. Duration: 60 minutes	Combined spinal epidural anesthesia SAB - with 2.6 ml of 0.5% Bupivacaine (Heavy) Through Epidural catheter - combination of lignocaine and bupivacaine administered in increments to achieve the desired sensory block level with minimal hemodynamic disturbances Blood loss: 50ml	4th Post operative day : Discharged	Immature teratoma (confirmatory finding)

POD- Postoperative day; **SAB-** Subarachnoid block; **PEB-** Cisplatin , Etoposide, Bleomycin; **PRBC-** Packed red blood cells; **FFP-** Fresh frozen plasma

retroperitoneal neoplasm in childhood, ranking behind neuroblastoma and nephroblastoma, and 10% of these tumors are malignant. Although these tumors are mostly asymptomatic, compression of the surrounding structures can cause abdominal distension and pain, as well as nausea and vomiting.¹⁰ Radiographic imaging plays an important role in the diagnosis of retroperitoneal teratoma. In our case too, the demographic characteristics and clinical appearance were typical as in the literature. In addition, the patient was deaf and dumb at the pre-operative assessment with Down syndrome and no congenital heart defects (Table 1). The surgical excision was performed under combined spinal epidural anesthesia. A long-term study showed that complete surgical resection is associated with the best survival rates for primary retroperitoneal tumors, which supports our decision to operate.¹¹ Since there is no evidence in the literature for the anesthetic management of these cases, the management in our case (Table 2) was mainly based on the precautions to be taken in the face of a large abdominal tumor causing impaired respiratory and circulatory functions, which prompted us to continue with regional anesthesia. We also anticipated and were prepared for other complications such as blood loss and anaphylaxis due to tumor spillage and infection that resulted in successful treatment of the case.

Mature cystic teratoma (MCT) is the most common benign neoplastic lesion of the ovaries in adolescents, accounting for approximately 70% of benign ovarian neoplasms in women under 30 and 50% of pediatric tumors.¹² Due to the risk of ovarian torsion, spontaneous rupture, and malignant ovarian cancer, the recommended life-saving treatment is surgical removal as soon as possible, followed by long-term follow-up to detect early relapse or involvement of the contralateral ovaries. The previous case reports have shown that surgical excision is performed under general anesthesia because of its association with anti-N-methyl-D-aspartate receptor (NMDAR) encephalitis, which occurs in approximately 38-58% of all cases and 94% of ovarian teratomas. It is important to understand the pharmacological interaction between the anesthetics and the disabled NMDAR and to develop an appropriate anesthetic plan because NMDAR is an important target for commonly used anesthetics with the possibility of autonomic instability. Since there were no clinical signs indicating

encephalitis in any of the three cases (Table 1) of our case series and there were no clear indications of an interaction of local anesthetics with disabled NMDAR, we continued with the combined spinal epidural anesthesia (Table 2, Figure 3) for the surgical procedure.

The age group of pediatrics and newborns poses challenges for anesthesia management per se. In addition, the literature on anesthesia management for teratomas is very limited and therefore these situations are fraught with challenges and surprises. Based on our observations in this case series, we are of the opinion that with the support of preoperative radiological imaging to determine the extent of the tumor to guide the anesthesia and surgical planning and an individual anesthesia plan, taking into account the pressure effects of the tumor on the cardiovascular, respiratory system and the risk of pulmonary aspiration of gastric contents. We should also be aware of other complications such as VAE, anaphylaxis and shock and be prepared to handle them when they arise as they can lead to disaster if unknown and unanticipated.

Although teratomas share a common modality of treatment, the anesthetic implications differ depending on the anatomical location of the teratoma. A thorough pre-anesthetic assessment, careful preparation, vigilant intraoperative monitoring, and a customized anesthetic plan will definitely provide a positive outcome.

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