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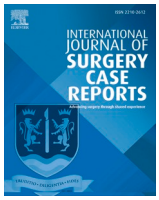

Case report

Case report: A unique case of benign teratoma in the posterior mediastinum associated with anterior meningomyelocele in a one-year-old child

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ABSTRACT

Introduction and importance: Benign cystic teratoma considered an extragonadal germ cell tumor that can present at any age and mostly located in the anterior mediastinum, only 3 %–8 % are in the posterior mediastinum. Meningomyelocele is an abnormal herniation of the meninges that located in most cases posteriorly in lumbo-sacral spine. Cervicothoracic meningomyelocele are rare entities resemble only 1 %–5 % of all neural tube defects. The presence of both anterior meningomyelocele (MMC) an benign teratoma is very rare and this association in the thoracic column has never been mentioned before in the medical literature.

Case presentation: We present the case of a one-year-old child, who was admitted to our hospital with a complaint recurrent vomiting episode, and respiratory distress, with no improvement in symptoms after conservative treatment. Computed tomography showed an anterior heterogeneous meningomyelocele that extend posteriorly to the upper lobe of right lung tissue. MRI confirmed the presence of the meningomyelocele in addition to a heterogenous cystic structure within. Thoracotomy was indicated and the meningomyelocele was carefully resected and sent to histopathology analysis which showed the presence of a benign teratoma accompanying the meningomyelocele.

Clinical discussion: Meningomyeloceles and teratoma are rarely associated, especially in the thoracic spine. Mediastinal tumors should be taken into consideration when a mass is found. A thorough imaging investigations is crucial in establishing the diagnosis along with histopathology after complete resection.

Conclusion: In the presence of posterior mediastinal meningomyelocele with heterogeneity, a histopathological examination of the specimen should be performed to exclude the mediastinal tumors.

1. Introduction

The most frequent intrathoracic lesions in children are mediastinal masses, and the anterior mediastinum considered the most common site [1]. Germ cell tumors (GCT) mostly occur in the gonads. However, in rare cases they could be present in extragonadal locations. The most common localization of extra-gonadal GCT in infants is in the mediastinum, which represent merely 1–3 % of all germ cell neoplasms [2]. Benign cystic teratoma considered an extragonadal germ cell tumor that can present at any age and mostly located in the anterior mediastinum with a close relationship to the thymus [3–5]. Only 3 %–8 % are in the posterior mediastinum [5,6].

meningomyelocele is a protrusion of meninges and spinal cord through a defect in the spinal column, it is usually found at the lumbar and sacral areas whereas Cervicothoracic meningomyelocele are rare entities resemble only 1 %–5 % of all neural tube defects.

Anterior thoracic meningomyelocele considered very rare with only 3 known case reports in the English-language Medical literature [7].

This case represents an association between anterior thoracic meningomyelocele and mature teratoma which was never described in the medical literature before.

The work has been reported in line with the SCARE criteria and the revised 2020 SCARE guidelines [8].

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Abbreviations

MMC	meningomyelocele
CT	computed tomography
MRI	Magnetic resonance imaging
GCT	Germ cell tumors
LDH	lactate dehydrogenase
AFP	alpha-fetoprotein
β HCG	Human chorionic gonadotropin
GI	gastrointestinal

2. Presentation of case

2.1. Patient information

We describe the case of a one-year-old male child who came to the emergency department with a complaint of post-feeding vomiting with dyspnea and tachycardia. The story began at the first month after birth with recurrent vomiting episodes especially after feeding and at the age of 3 months, a dyspnea was developed, so he was admitted several times in a rural hospital and treated with antibiotics and bronchodilators without any improvement. The child was referred to our emergency department with mild respiratory distress, tachycardia, and mild dehydration.

No urogenital or other gastrointestinal problems were reported and there was no relevant medical or family history.

2.2. Clinical findings

Clinical examination revealed tachycardia with a respiratory distress, soft rales, and wheezing sounds in the upper right lobe upon auscultation.

2.3. Diagnostic assessment

Chest X-Ray showed a mass in the right upper hemithorax with a spinal curve convex to the left.

Computed tomography (CT) scan of the chest revealed multiple thoracal vertebral segmentation anomalies and a large anterior cystic mass that measures 30 × 29 × 26 mm herniating into the thoracic cavity through a bone defect at the level of T3–T4 vertebrae, so it was assumed to be meningomyelocele as initial diagnosis (Fig. 1C).

MRI T2-weighted images confirmed the meningomyelocele that extends laterally into the chest cavity, posterior to the upper lobe of the right lung, and measures 44 × 39 × 26 mm, in addition to a heterogenous cystic structure within (Fig. 1A–B).

Syringomyelia was also noticed at a level above the meningomyelocele measuring 10 mm.

Laboratory investigations were all within normal ranges, Tumor markers (a-FP and β -HCG) were normal.

2.4. Therapeutic intervention

Open surgery was indicated based upon the given clinical and radiological pictures. The procedure was performed at our tertiary university teaching hospital. It was done by a fifth year senior Pediatric Surgery residents with five years of surgical experience and by a Pediatric Surgery specialist with 32 years of Pediatric Surgery experience. In addition to a neurosurgeon with 30 years' experience of neurosurgery.

The procedure was carried-out Under general anesthesia without complications, a right posterolateral thoracotomy was performed to reach the posterior mediastinum where the cystic cavity was found to be adjacent to the right lung (Fig. 2), the capsule was opened carefully and a quantity of cerebral spinal fluid came out, the spinal cord and nerve roots were gently dissected from surrounding durra matter, the cyst was fully resected and sent to histopathological examination, the dura was sutured and then a drainage chest tube was placed.

Histopathology revealed a presence of mature teratoma without immature or malignant components, with Skin, brain, GI mucosa, smooth muscles, cartilage contents next to other types of tissue which consist with mature teratoma (Fig. 3A–C).

The child had uneventful recovery, the respiratory symptoms disappeared, the movements were completely intact, the chest tube was removed after 5 days of surgery, and he was discharged home after 2 days.

He has been followed up as an outpatient for 6 months following his operation, and he was scheduled for regular visits to the pediatric surgery clinic for evaluation, which were unremarkable.

3. Discussion

Mature teratomas are neoplasms derived from endodermal, mesodermal, and ectodermal origin. They are the most usual form of germ cell tumor (GCTs). The incidence rate is equal in both men and women, and they are mostly benign tumors [9]. Because of the slow growth, Benign mediastinal teratomas remain asymptomatic and they usually diagnosed accidentally when a chest X-ray or CT scan is made for other indications [6,10]. When symptomatic, the symptoms are usually respiratory due to compression of adjacent structures: respiratory distress, dyspnea, pneumonia, cough, wheezing, and chest pain. In addition, the mass might cause a chest wall deformity or of spinal cord compression [11]. Generally, with benign mature mediastinal teratoma, the levels of serum the tumors markers (β HCG, lactate dehydrogenase (LDH), and AFP) remain within the normal ranges [12], because the benign teratoma (pure mature teratoma) does not secrete AFP and β HCG. Moreover, Serum levels of AFP, β HCG, and LDH considered very important in the noninvasive diagnostic of GCTs. High levels of serum AFP or ß HCG indicate malignant components such as yolk sac tumor or choriocarcinoma, and rules out pure mature teratoma or seminoma [13]. Another core diagnostic modality is CT scan. It shows the presence of multiple-tissue elements within a well-defined cystic mediastinal mass [4], 50 % of the cases contains Fatty and cystic components, and sometimes a fat-fluid level within the mass strongly suggests the diagnosis [14,15]. MRI helps also with detecting a heterogenous mediastinal mass containing a variable mixture of fat, fluid, soft tissue, and calcifications, or the invasion of surrounding structures [16].

Meningocele is an abnormal herniation of the spinal cord and meninges through a neural foramina or a bony defect in a vertebral body, and 80 % of them located posteriorly in lumbosacral spine and considered as subtype of the closed form of spinal dysraphism On the other hand, Anterior spinal meningomyelocele are very rare and generally found in the anterior aspect of the thoracic or sacral spine. These frequently occur as a complication of generalized mesenchymal dysplasia, in particular, and usually associated with tethering of the spinal cord. [11]. Symptoms are related to size and its relationship with surrounding structures and might include back pain, paraparesis, shortness of breath, coughing, and palpitation as well. Imaging modalities such as CT scan and MRI are essential for establishing the diagnosis as well as for showing the relationship between the meningomyelocele and its surrounding structures.

Surgical excision considered the golden standard in treatment of teratomas, and it is important to resect the mass because of the compressive effect on adjacent tissues. Additionally, all benign tumors and cyst in the mediastinum can transform into malignancy over the time [17].on contrary, there is no need for surgery in Occult spina bifida in most cases unless the patient presents symptoms. Untethering surgery is performed if new or progressive symptoms develop. Prophylactic surgery is recommended for the tethered cord to prevent irreversible urinary problems when they occur [18].

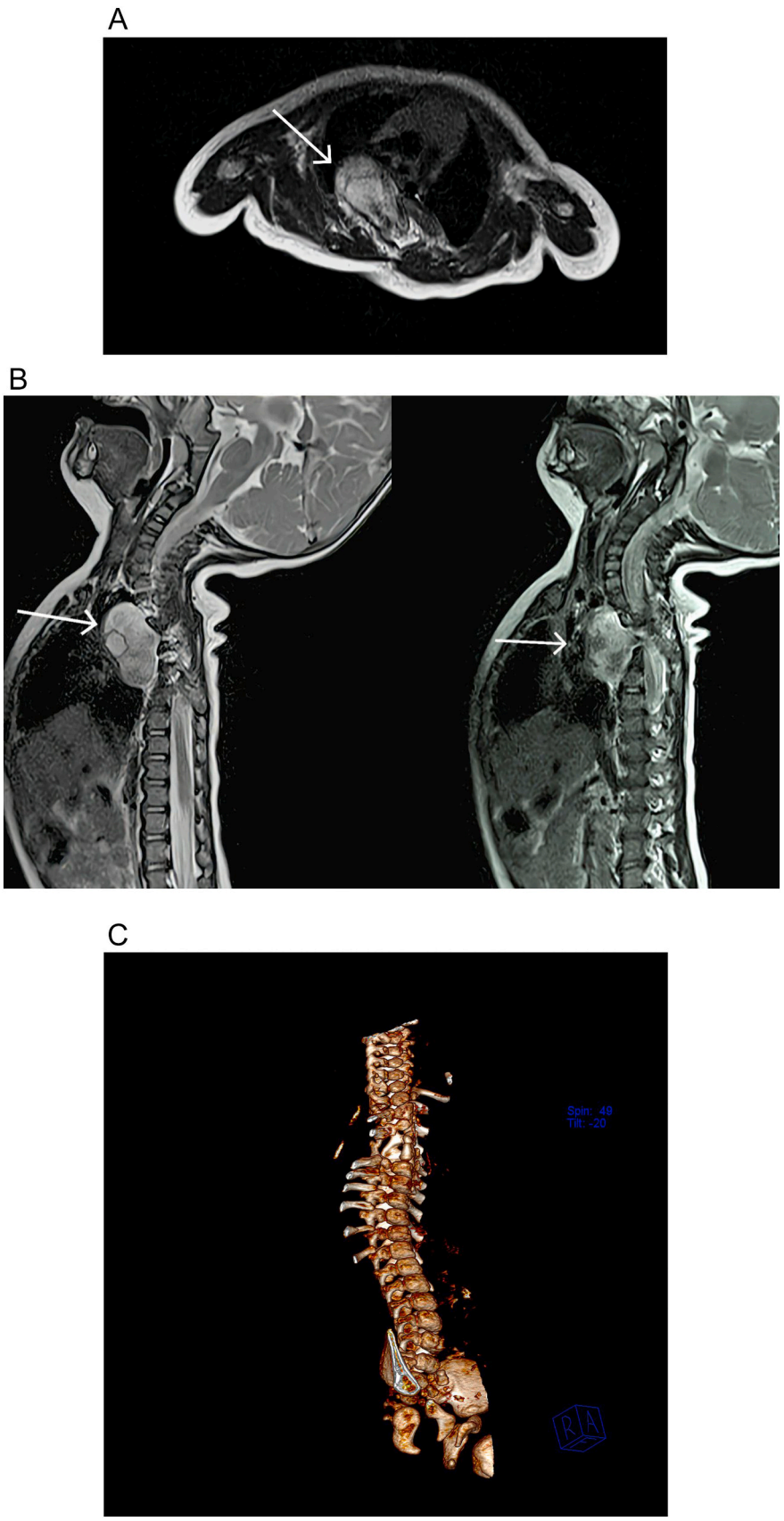

Fig. 1. A cross sectional MRI T2-weighted image of the Chest showing the meningomyelocele in the posterior mediastinum (white arrow) that measures (44 × 39 × 26 mm). It contains several septa with several connections to the cerebrospinal fluid With retraction of the thoracic spinal cord.
B sagittal MRI T2-weighted image, showing that the meningomyelocele (white arrow) located at the level of T4-T5-T6-T7. in addition to a heterogenous cystic structure within.
C: CT reconstruction image showing vertebral anomalies. T4, T5 hemivertebra, and T6&T7 Fusion.

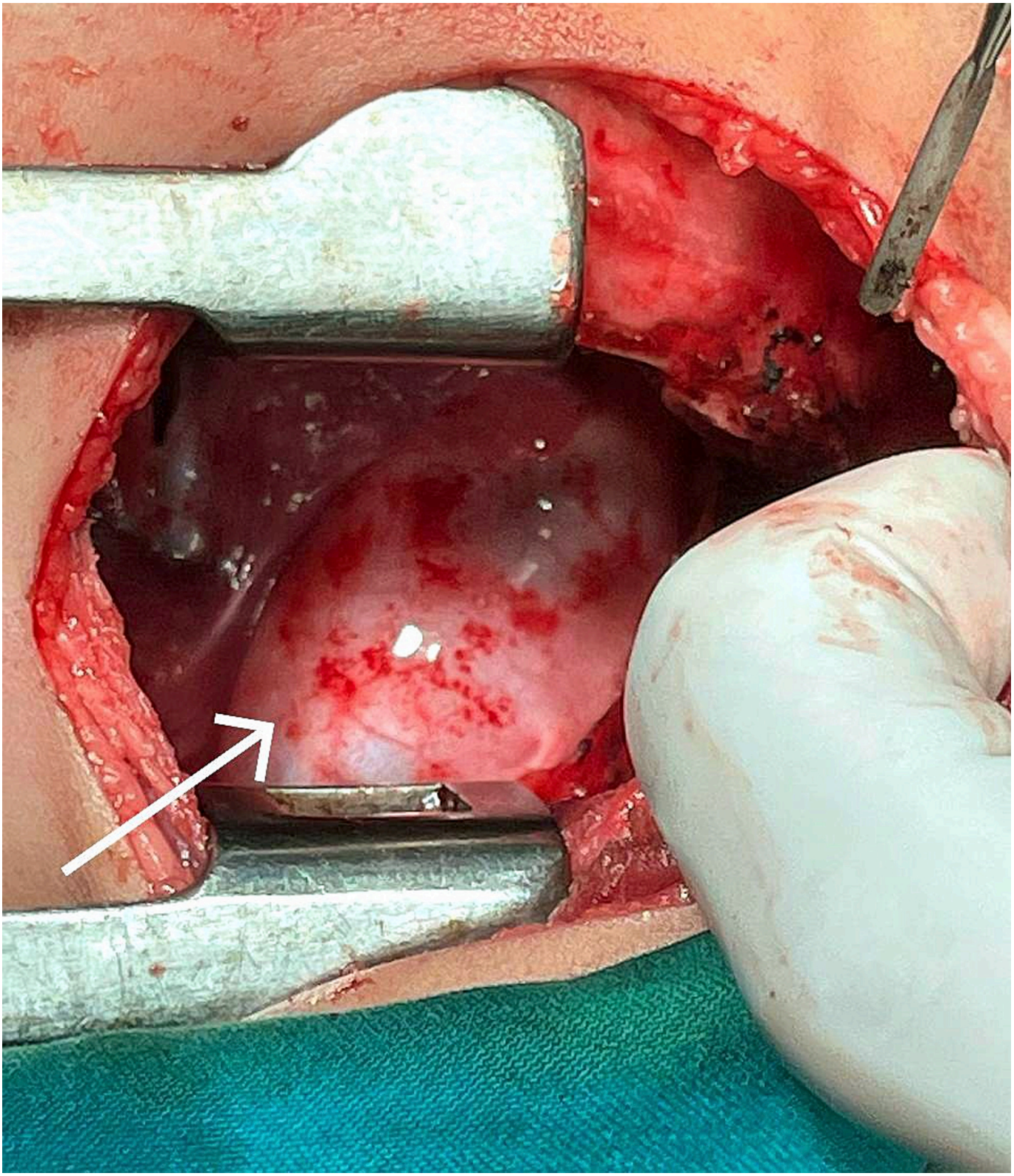

Fig. 2. Intraoperative image prior to excision. White Arrow points toward anterior meningomyelocele, that is located in posterior mediastinum.

If stayed Untreated, benign mediastinal teratoma can cause several complications like lung atelectasis, adjacent tissue compression, infection, perforation in hemithorax and cardiac tamponade [19]. Usually, the cause of Cystic rupture is the infection [17]. benign teratomas have a very excellent prognosis after excision even when complete excision is not possible. Additional treatments such as Postoperative irradiation or other adjuvant measures are not indicated [20].

4. Conclusion

This case represents the first reported anterior thoracic meningomyelocele associated with mature teratoma in the posterior

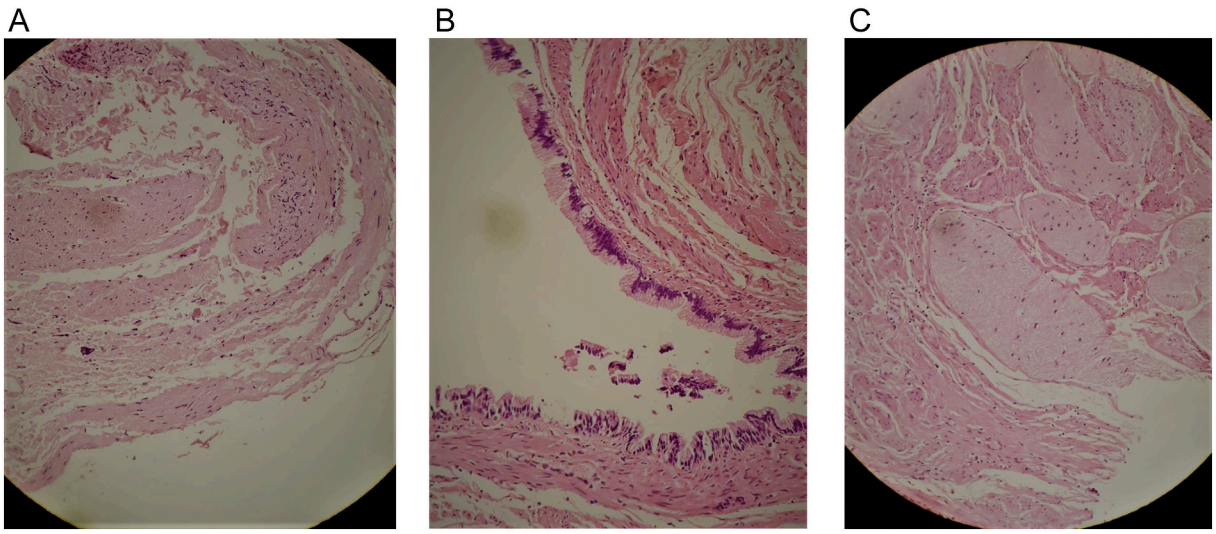

Fig. 3. A: H&E stained section showing glial tissue and fibrous stroma (magnification × 40).
B: H&E stained section showing mature intestinal mucosa (magnification × 40).
C: H&E stained section nerve fibers and smooth muscle (magnification ×40).

mediastinum in a child. It demonstrates the importance of histopathology in establishing an accurate diagnosis of posterior mediastinal masses, even in the presence of thorough imaging investigations, when there is a slight doubt about the diagnosis.

Consent of patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Availability of data and materials

The datasets generated during and/or analyzed during the current study are not publicly available because the Data were obtained from the hospital computer-based in-house system. Data are available from the corresponding author upon reasonable request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

Institutional review board approval is not required for deidentified single case reports or histories based on institutional policies.

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Guarantor

Khaled Alomar.

CRediT authorship contribution statement

RA, KA, RSA: Who wrote, original drafted, edited, visualized, validated, literature reviewed the manuscript.

FAS: General Surgery senior resident who was the first assistant in the surgery. Supervision, project administration, and review of the manuscript.

GM: Neurosurgery specialist, who performed and supervised the operation.

HD: Head of Pediatric surgery department in Pediatrics' university hospital, supervision, and review of the manuscript.

KA: Conceptualization, resources, and the corresponding author who submitted the paper for publication.

All authors read and approved the final manuscript.

Declaration of competing interest

Not applicable.

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