Annals of Medicine and Surgery 79 (2022) 103952

Contents lists available at ScienceDirect

Annals of Medicine and Surgery

journal homepage: www.elsevier.com/locate/amsu

Case Report

Therapeutic actions in a 2-month-old child with teratoma and duodenal malrotation accompaniment

Parisa Rahmani [a], Parastoo Sharifi [a], Bahar Ashjaee [b], Maryam Ebrahimi [a,*]

[a] Pediatric Gastroenterology and Hepatology Research Center, Children's Medical Center, Pediatric Center of Excellence, Tehran University of Medical Sciences, Tehran, Iran

[b] Department of Pediatric Surgery, Pediatric Center of Excellence, Tehran University of Medical Sciences, Tehran, Iran

ARTICLE INFO

Keywords:
Intestinal malrotation
Teratoma
Upper GI series

ABSTRACT

Introduction: and importance: A mature mediastinal teratoma is a rare type of tumor that can contain fully developed tissues and it is usually considered benign, more common in female and may require surgical removal. The most common locations for teratomas are the tailbone (coccyx), ovaries, and testicles, but in this case, the teratoma was located in the mediastinum.

Case presentation: Furthermore, intestinal malrotation could potentially lead the patient to gastrointestinal (GI) obstruction and PO intolerance. Here, we present a 2-month-old patient with considerable manifestation of GI intolerance due to intestinal malrotaion accompanied by mature mediastinal teratoma.

Clinical discussion: An evaluation of malrotation should be part of every upper GI tract examination in pediatric patients, particularly newborns and infants.

Conclusion: Finally, in this case, surgical intervention could enhance both malrotation and mediastinal teratoma.

1. Introduction

Teratomas are neoplasms that can contain fully developed tissues and organs, including hair, teeth, muscle, and bone, and are foreign to the anatomic site of occurrence. They may appear in newborn, children, or adults. However, it has been reported that they are more common in females. Teratomas are usually benign in newborn, but still may require surgical removal.

They are most commonly detected in the sacrococcygeal region or gonads and are less frequent in other sites. Mature mediastinal teratoma typically manifests in CT scan as a heterogeneous anterior mediastinal mass containing either soft-tissue, fluid, fat, or calcium attenuation, or any combination of the four. CT is the imaging technique of choice in the evaluation of these lesions [1].

Intestinal malrotation, which is defined by a congenital abnormal position of the bowel within the peritoneal cavity, may lead to increased risks of bowel obstruction, midgut volvulus and also a potentially life-threatening condition [2]. The term "malrotation" is applied to wide range of intestinal anomalies, from readily apparent omphalocele in new born to asymptomatic nonrotation of the large and small bowel in adults [3,4]. Also, most patients with malrotation show signs and symptoms soon after birth. However, malrotation in the minority cases are diagnosed long after infancy and does not show typical clinical signs or symptoms [5]. So that, an evaluation for malrotation should be a part of every upper GI tract examination in paediatric patients, particularly neonates and infants. The upper GI series could help radiologist to distinguish a normal position of the bowel from maltreated tract by reconstructing position of the duodenojejunal junction [6]. Effective differentiation of a normal variant bowel location from malrotation, either in paediatrics or in adults, requires appropriate imaging technique and interpretation [7]. In this paper, our infrequent case has represented pathological features of mature mediastinal teratoma associated with adhesion to the oesophagus, aorta, and diaphragm accompanied by malrotation of the intestinal tract.

2. Case report

A 2-month-old girl was presented to our center with a complaint of inability to eat with progressive nonbilious vomiting episodes containing ingested particles since she was 1 week old. Consequently, the patient was experiencing complete PO intolerance and post-feeding coughs for 10 days prior to admission. Furthermore, the patient defecated once a week and the faecal matter was described as soft in nature. The patient's weight was 3200(g) at the time of birth and 2900(g) at the time of

* Corresponding author.
E-mail address: drebrahimi97989@gmail.com (M. Ebrahimi).

https://doi.org/10.1016/j.amsu.2022.103952
Received 4 April 2022; Received in revised form 2 June 2022; Accepted 2 June 2022
Available online 6 June 2022

admission and did not have history of any medical ailments. She also had a normal result of lumbar puncture to exclude meningitis. She also experienced unsuccessful reflux treatment (Nexium and Famotidine) in another centre. Since lactose intolerance was considered a possible diagnosis in another centre, Aptamil Pepti was administered 10 days prior to admission. Physical examination demonstrated tachycardia and dehydration that resolved later after appropriate fluid therapy. The abdominal examination showed soft palpation, without organomegaly or tenderness. In our hospital, sweat, metabolic and allergic tests were performed and the results were normally evaluated (shown in Table 1).

In her upper GI series exam (shown in Fig. 1) malrotation was detected, while her CT scan (shown in Fig. 2) illustrated a foreign body particle that was due to barium accumulation in the diverticula. Two operations were performed in this unit. The first was a left thoracotomy with an excision of left mediastinal mass that had an adhesion to the diaphragm and the oesophagus respectively. The second was one week after the first surgery and aimed to fix her intestinal malrotation. The duodenum rotated around its mesothelium and was detorted later. The Ladd band was released to resolve duodenal obstruction. All adhesion bands were cut from the ligamentum teres to the ileocecal junction. The base of the mesenteric artery was also freed. The lymph nodes were swollen, and a sample was sent to the pathology laboratory for investigation. Pathologic evaluation stated mature teratoma composed of multiple tissues like pancreas, hair follicles, stratified keratinized squamous epithelium and adipose and pancreatic tissue. In this study, human chorionic gonadotropin (HCG), as an appropriate marker for the progression and evaluation of treatment, was measured postoperatively. HCG can be secreted by various abnormal germ cells, and benign or malignant non-testicular teratomas. In non-seminomatous germ cell tumors (NSGCT), HCG is used in combination with alpha-fetoprotein (AFP) and lactate dehydrogenase (LDH). So that, β-HCG and AFP were requested (shown in Table 2). The patient was followed up at hospital clinic for two months and did not report any major complications. The case report is stated in accordance with SCARE 2020 guidelines [1].

Table 1
The result of abdominal examination.

Variable (normal range)	Presentation	Variable (normal range)	Presentation
White blood cells 109/L (4.00–10.00)	6.25	Cholesterol mg/dl (110–220) TG ng/dl (<250)	137 289
Neutrophils %	39%	Ca mg/dl (9–10.5)	8.6
Lymphocytes %	53%	P mmol/l (0.8–1.5)	5
Platelets 10^9/L (150–350)	463	Chloride mEq/l (98–106)	101
Hemoglobin g/dL (12–15)	9.9	Amonia mcg/dl (40–80)	33
C-reactive protein mg/dL (<0.5)	2	Gamma GT U/l (8–77)	13
Alkaline phosphatase U/L (40–109)	172	Mg mg/dl (1.5–2.4)	2.1
Aspartate transaminase (AST)IU/l (10–34)	32	TSH mcIU/ml (0.5–5) T4 ng/dl (0.9–2.4)	2.6 7.8
Alanine transaminase (ALT)IU/l (10–130)	12	Bill direct mg/dl (0–0.3) Bill total mg/dl (0.3–1.2)	0.2 0.6
Albumin g/dL (3.4–4.8)	3.7	U/A Glucose ± WBC (0–5) Protein ±	2+ 6–8 1+
K mmol/L (3.7–5.10)	4.1	INR (<1.1)	1
Na mmol/L (135–145)	139	CSF Glucose mg/dl (40–80) WBC(0–5) Protein mg/dl (15–60)	47 0 43
Creatinine mg/dL (0.3–0.7)	0.7	Pyruvate micro g/dl (300–900)	0.6
Blood Glucose mg/dl (70–105)	290	VBG PH HCO3 mmol/l (22–26) CO2 mmHg	7.25 17.7 41.2
BUN mg/dl (5–20)	40	ESR mm/h (<20)	8

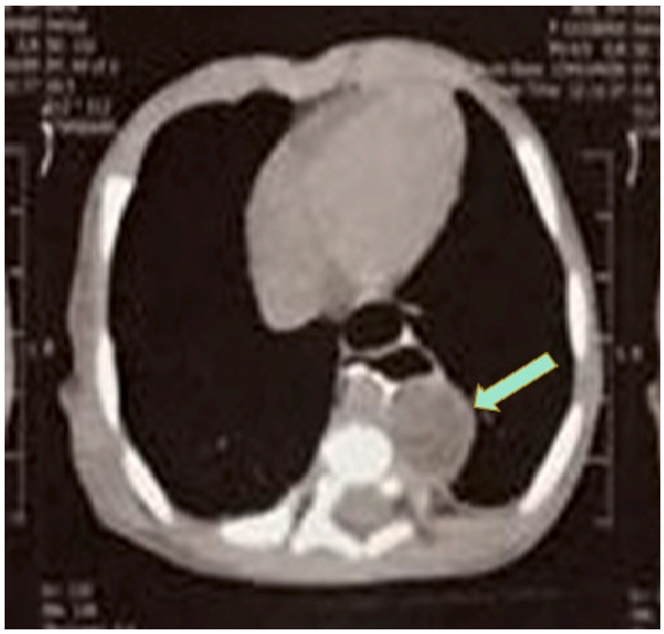

Fig. 1. Patient's chest CT scan with IV contrast is indicating mediastinal foreign body existence.

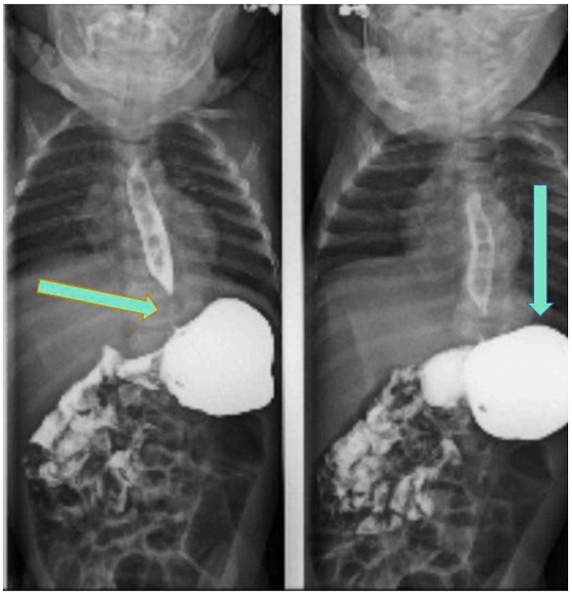

Fig. 2. Patient's upper GI series exam is illustrating intestinal malrotation.

Table 2
The results of β-HCG and AFP.

AFP IU/L (44–147)	60	BHCG Females Birth-3 months: ≤50 IU/L	0.2

3. Discussion

The highlights of the present case include: a teratoma in a female child arising from the wall of the oesophagus, aorta, diaphragm and its unusual association with duodenal malrotation. These associated anomalies in the gastrointestinal tract may be related to local developmental defects caused by the teratoma. Despite the existence of associated conditions, our patient was successfully operated successfully for teratoma as well as GI anomalies.

Majority of the teratomas can be diagnosed pre-operatively [8]. Diagnosis of teratoma in our case was not suspected initially probably due to rarity of mediastinal teratoma. An upper GI series exam also showed a mass in the mediastinum that led our suspicion to a foreign body while CT scan unveiled mediastinal teratoma. Following the diagnosis of teratoma, the patient underwent operation and a further pathological evaluation revealed mature teratoma. Intestinal malrotation could be considered a critical condition among paediatrics. In particular, the upper GI series exam plays a key role in the diagnosis of intestinal malrotation. However, previous studies have shown that not all cases could be diagnosed by the upper GI series exam [9]. Furthermore, the Ladd procedure and additional surgical interventions should be considered crucial in the evaluation of paediatrics with intestinal malrotation [10].

For future studies, we recommend consideration of teratoma among patients with cystic gastrointestinal lesions. Early diagnosis and treatment has excellent prognosis and can prevent the development of anomalies.

4. Conclusions

In this study, we could present a rare case of mediastinal teratoma accompanied by intestinal malrotation. Interestingly, it could be hypothesized that the existence of mediastinal teratoma with upper GI adhesions could potentially alter the risk of intestinal malrotation.

Ethical approval

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Sources of funding

No funding was secured for this study.

Author contributions

Dr. Parisa Rahmani: conceptualized and designed the study, drafted the initial manuscript, and reviewed and revised the manuscript.

Dr. Bahar Ashjaee and Dr. Parastoo Sharifi: Designed the data collection instruments, collected data, carried out the initial analyses, and reviewed and revised the manuscript.

Dr. Maryam Ebrahimi: Coordinated and supervised data collection, and critically reviewed the manuscript for important intellectual content.

All authors approved the final manuscript as submitted and agree to be accountable for allaspects of the work.

Conflict of interest

The authors deny any conflict of interest in any way or by any means during the study.

Research registration

N/A.

Guarantor

Parisa Rahman.

Consent to publish

Consent to participate from the under 16 years old was given by a parent or legal guardian.

Consent to participate

Patient consent was obtained prior to the surgery.

Patient consent

We confirm written consent was obtained for the publication of this case report from the parents. This report does not contain any personal information that could lead to the identification of the patient.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Availability of data and material

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103952.

References

[1] K. Moeller, P.A. Templeton, Mediastinal mature teratoma: imaging features, AJR Am. J. Roentgenol. 169 (1997) 985–990.
[2] P.J. Strouse, Disorders of intestinal rotation and fixation (“malrotation”), Pediatr. Radiol. 34 (11) (2004) 837–851.
[3] R.T. Maxson, P.A. Franklin, C.W. Wagner, Malrotation in the older child: surgical management, treatment, and outcome, Am. Surg. 61 (2) (1995) 135–138.
[4] P. Prasil, H. Flageole, K. Shaw, L. Nguyen, S. Youssef, J.-M. Laberge, Should malrotation in children be treated differently according to age? J. Pediatr. Surg. 35 (5) (2000) 756–758.
[5] D.W. Dietz, R.M. Walsh, S. Grundfest-Broniatowski, I.C. Lavery, V.W. Fazio, D. P. Vogt, Intestinal malrotation, Dis. Colon Rectum 45 (10) (2002), 758-1381.
[6] D. Jamieson, D. Stringer, Small Bowel. Babyn PS Pediatric Gastrointestinal Imaging and Intervention, second ed., Decker, Hamilton, Ontario, 2000, pp. 311–332.
[7] K.E. Applegate, J.M. Anderson, E.C. Klatte, Intestinal malrotation in children: a problem-solving approach to the upper gastrointestinal series, Radiographics 26 (5) (2006) 1485–1500.
[8] F.R. Long, S.S. Kramer, R.I. Markowitz, G.E. Taylor, C.A. Liacouras, Intestinal malrotation in children: tutorial on radiographic diagnosis in difficult cases, Radiology 198 (3) (1996) 775–780.
[9] S.A. Kapfer, J.F. Rappold, Intestinal malrotation—not just the pediatric surgeon’s problem, J. Am. Coll. Surg. 199 (4) (2004) 628–635.
[10] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg 84 (2020) 226–1500.