



A PORTAL FOR RESEARCH, INNOVATION AND KNOWLEDGE

APSP JOURNAL OF CASE REPORTS

OFFICIAL JOURNAL OF THE ASSOCIATION OF PAEDIATRIC SURGEONS OF PAKISTAN

ISSN 2218-8185

JAN - JUN

VOLUME 1 (1); 2010

AJ  CR

EMAIL: Editors@apspcaserep.com

<http://www.apspcaserep.com>

APSP Journal of Case Reports: A New Beginning

Bilal Mirza, Jamshed Akhtar*

In a book, Orphan Diseases: New Hope for Rare Medical Conditions, author Wendy Murphy in introductory pages quotes William Harvey, an English physician (1578 – 1657)

"Nature is nowhere accustomed more openly to display her secret mysteries than in cases where she shows tracings of her workings apart from the beaten paths; nor is there any better way to advance the proper practice of medicine than to give our minds to the discovery of the usual law of nature, by careful investigation of cases of rarer forms of disease".^{1,2}

This is apt to the discipline of pediatric surgery, as unusual anomalies are seen frequently in clinical practices. At times one tries to seek opinions and learn from experiences of other colleagues in dealing with such patients. Case reports thus become an important source of documenting such experiences and observations. In hierarchy of evidence in scientific literature, they are placed at lowest level but even then, are the important contribution to the medical literature. According to Karl Popper any proposition or theory is scientific only if it is falsifiable. Thus logically an observation can falsify an assertion. A reported case may shift the existing thinking paradigm.³

Case reports have made important contributions in relation to many clinical conditions. Cases of HIV / AIDS have been documented initially as case reports. Many adverse drug reactions came into light when such incidences were reported under this category of manuscript. They are of educational value and guide us to future areas of research. In selecting case reports for this journal we ensure that they must address or reveal new scientific information. We have also added segments like images and letters to the editor, so as to provide educationally rich clinical observations to our readers.

Many scientific journals have stopped publication of case reports. Various reasons have been cited though according to some publishing of case reports affects the impact factor.⁴ Also as research became an integral part of teaching, training and promotions, authors are now inclined towards more elaborate study designs, thus their interest in this category of publication has declined. This

domain is usually left to the juniors in the discipline. The APSP Journal of Case Reports, an online, free access periodical, from the platform of the Association of Paediatric Surgeons of Pakistan, is one such portal through which authors can present their experiences to the world.

The first issue of this journal is taken out in record time. One of the editors (Bilal Mirza) remained instrumental in this regard. He contributed greatly in developing and maintaining the website of the journal as well. All articles presented in this are peer reviewed by senior pediatric surgeons both from Pakistan and abroad. Elaborate guidelines are available on our website both for the authors and reviewers. We hope you will contribute to this first ever electronic journal dedicated to case reports from Pakistan. Your participation by submitting quality manuscript shall improve the standard of the journal and increase its readership.

REFERENCES

1. Murphy W. Apart from beaten path In Orphan Diseases: New Hope for Rare Medical Conditions, Twenty-First Century Books Millbook Press Incorporated Connecticut 2002: pp 9. Available at, <http://books.google.com> accessed on July 25, 2010.
2. William Harvey. Today in Science History. Available at http://www.todayinsci.com/H/Harvey_William/HarveyWilliam-Quotations.htm accessed on July 25, 2010.
3. Falsifiability. Available at <http://en.wikipedia.org/wiki/Falsifiability> accessed on July 24, 2010.
4. Kidd M, Hubbard C. Introducing Journal of Medical Case Reports. J Med Case Rep 2007;1:1.

Dr. Jamshed Akhtar

Editor, APSP J Case Reports
Department of Paediatric Surgery
National Institute of Child Health Karachi
Email:jamjim88@yahoo.com

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](https://creativecommons.org/licenses/by/3.0/)

How to cite

Mirza B, Akhtar J. APSP Journal of Case Reports: A new beginning. APSP J Case Rep 2010; 1: 1.

An Unusual Case of Gastroschisis

Bilal Mirza,* Afzal Sheikh

ABSTRACT

Gastroschisis is an abdominal wall defect through which intestine and rarely other organs eviscerate. It is less frequently associated with anorectal malformations. Abnormal size and shape of the defect is rarely identified in these patients. We report a case of gastroschisis with an unusual abdominal wall defect, imperforate anus and an ectopically placed vestibule. The defect was extended from right side of umbilicus to the perineum. There was evisceration of entire gastrointestinal tract (GIT), liver, gallbladder and urinary bladder. The defect was not manageable with a spring loaded silo and a sterilized blood bag was used to cover the defect. The unusual defect, associated anomalies and evisceration of unusual viscera are the main reasons for reporting the index case.

Key words: Gastroschisis, Imperforate anus, Evisceration.

INTRODUCTION

Gastroschisis refers to an abdominal wall defect situated usually on the right side of umbilical cord, through which intestine and rarely other abdominal viscera are eviscerated, with no covering membrane or sac. It is rarely associated with anorectal malformations.^{1,2}

The most accepted theory about the development of the defect is based on the resorption of right umbilical vein resulting in a defect on the right side of umbilical cord.^{1,3} We report an usual case of gastroschisis in association with anorectal malformation. It may provide some insight as to the etiology of this anomaly.

Address: Department of Paediatric Surgery, The Children's Hospital and Institute of Child Health Lahore, Pakistan

E-mail address*: blmirza@yahoo.com

* (Corresponding author)

Received on: 10-06-2010

Accepted on: 01-07-2010

<http://www.apspjcaserep.com>

© 2010 Mirza et al

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](https://creativecommons.org/licenses/by/3.0/)

CASE REPORT

A 24-hour-old female baby, product of spontaneous vaginal delivery, at term, in a village, weighing 2kg presented to emergency room with evisceration of intestine from abdomen. The patient was hypothermic and cyanosed. On examination baby had temperature of 96F, respiratory rate 45/min and pulse 80/min. Patient was placed under infant warmer and oxygen inhalation given via mask. Warm IV fluids infused and antibiotics started. After stabilization, the patient was shifted to the operation theatre.

The abdominal defect was about 7cm in diameter and on right side of the umbilical cord. It was extending down to the perineum. The entire GIT was eviscerated along with liver, gallbladder and urinary bladder. Spleen, ovaries and uterus were lying inside the abdominal cavity. The intestine, gallbladder and urinary bladder were matted together and a thick peel covered them in toto. Anus was absent. There was a labial prominence on right side of the defect in perineum while on left side an abnormal vestibule harbouring two openings, found. From one opening meconium was coming and from other urine passed out. There was another labial prominence lateral to the vestibule. There was no bladder or cloacal extrophy [Image].



Image Defect on right side of umbilicus and extending down to the perineum; evisceration of matted and massively edematous intestinal loops, liver and gallbladder; imperforate anus and ectopically placed vestibule showing meconium inside.

It was not possible to close the defect primarily because of very limited abdominal capacity, much increased size of abdominal viscera and unusual defect. We tried spring loaded silastic silo, but it was not technically compatible with the size and shape of the defect, a sterilized blood bag was then used as silo. Post operative course remained stormy. Later baby developed septic shock and died.

DISCUSSION

The term gastroschisis (Gastro = belly, Schisis = separation) was used, for the first time, by Taruffi in 1894, and it refers to an abdominal wall defect situated usually on the right side of umbilical cord through which mid-gut and rarely other abdominal organs eviscerate.^{1,4}

Other viscera that may eviscerate are stomach, duodenum, colon, urinary bladder, gonads, liver whole or some part of it, and gallbladder.¹ In our case the entire intestine, from stomach to rectum, was eviscerated along with liver, gallbladder and urinary bladder.

The defect in gastroschisis is usually small (<4cm) as compared to omphalocele. The defect usually lie on the right side of umbilical cord but left sided gastroschisis has been reported in literature.^{1,5} In our patient the defect was in usual location but extended down to the perineum.

Two most important theories regarding the development of the defect are resorption of right umbilical vein and failure of mesodermalization. According to the first theory, the defect probably resulted due to failure of the umbilical coelom to develop. The elongating intestine then has no space to expand and ruptures out in amniotic cavity. This event occurred on the right side of umbilical cord due to resorption of right umbilical vein at 4th week of gestation. According to the other, but not generally accepted theory, there is failure of mesodermalization of the anterior abdominal wall with absence of abdominal wall components.³ In our case the defect was unusual and resorption of right umbilical vein theory cannot explain it adequately. Moreover, the existence of left sided defects, also points towards some other etiology of the defect. Following considerations can be made regarding the index case;

1. It may be a case of caudal fold defect
2. It may be a combined defect i.e. caudal fold defect and gastroschisis
3. It may be a new type of malformation
4. It may be a case of gastroschisis with unusual defect

The first two theories cannot explain the defect in absence of bladder or cloacal exstrophy which are associated with caudal fold defects. Various defect have been reported in literature and include a defect in the left hypochondrium and bilateral defects, but, none of them had associated eviscerated bowels and usual relationship with umbilical cord, as typically characterized in the gastroschisis.^{6,7} These cases could be considered as new malformations, however, in our case the location of the defect and evisceration of abdominal content preclude its place in new malformation. The fourth option may be valid in our case. The defect, in the index case, was too long and can not result from mere resorption of right umbilical vein. This strengthens the notion of true absence of the abdominal wall that could be the result of failure of mesodermalization process.

Gastroschisis has low association with congenital malformations as compared to the omphalocele. The reported associated anomalies are intestinal atresia, malrotation, choledochal cyst, cleft lip and palate, and sternal clefts. Its association with anorectal malformations is rarely reported.^{1,4,5,8,9} In our case the anal pit was absent and an abnormal opening with meconium inside was present in ectopically placed vestibule. This might be a recto-vaginal fistula because urethral opening was lying in normal relation at ectopic site.

In our case the patient presented after 24 hours of birth with massively matted and edematous eviscerated contents. Though early cover was applied to eviscerated structure following optimization still baby went into sepsis and died. If timely referral, ideally antenatal diagnosis,

had been made baby might be saved. To conclude, right umbilical vein resorption theory cannot explain every patient of gastroschisis, and failure of mesodermalization theory can be put forward to explain the unusual defect found in the reported case.

REFERENCES

1. Mirza B, Ijaz L, Sheikh A. Evisceration and necrosis of gall bladder and evisceration of urinary bladder in a patient of gastroschisis. *Afr J Paediatr Surg* 2010;7:43-4.
2. Rao SR, Rao TR, Gajula SS, Rao AV. Rare case of fetal gastroschisis with aplasia of the foot and external genital organs. *Congenit Anom* 2008;48:140-1.
3. Grosfeld JL, O'Neill JA, Coran AG, Fonkalsrud EW, Caldamore EW. Editors Paediatric surgery. 6th ed. USA: Mosby Elsevier; 2006. p. 1162-7.
4. Serber J, Stranzinger E, Geiger JD, Teitelbaum DH. Association of gastroschisis and choledochal cyst. *J Pediatr Surg* 2009;44:E23-E26.
5. Suver D, Lee SL, Shekherdimian S, Kim SS. Left-sided gastroschisis: higher incidence of extraintestinal congenital anomalies. *Am J Surg* 2008;195:663-6.
6. Carrera IA, Pitarch V, Garcia MJ, Barrio AR, Martinez-Frias ML. Unusual congenital abdominal wall defect and review. *Am J Med Jenet* 2003; 119: 211-3.
7. Hebbar S, Pratapkumar N. An unusual case of Foetal Gastroschisis with Limb aplasia and Extra-corporal liver Calicut Medical Journal 2005; 3(3):e2 URL: <http://www.calicutmedicaljournal.org/2005/3/3e2>
8. Khalil BA, Murphy H, Jones MO, Ellis I, Lamont GL. Gastroschisis associated with cleft lip and palate. *J Pediatr Surg* 2007;42:E9-E10.
9. Lee TC, Barshes NR, Nguyen L, Karpen SJ, Quiros-Tejeira RE, Carter BA, et al. Gastroschisis and biliary atresia in a neonate: uncommon presentation or common precipitant. *Eur J Pediatr Surg* 2005;15:434-6.
10. Dabbas N, Muktar Z, Ade-Ajayi N. GABBY: An *ex vivo* model for learning and refining the technique of preformed silo application in the management of gastroschisis. *Afr J Paediatr Surg* 2009; 6:73-6.

How to cite

Mirza B, Sheikh A. An unusual case of gastroschisis. *APSP J Case Rep* 2010; 1: 2.

Atresia of the Ascending Colon: A Rarity

Haroon Mansoor,* Naila Kanwal, Mahmood Shaukat

ABSTRACT

Atresia of the colon is among the rare types of all gastrointestinal atresias. Ascending colon is the rarest site of all the colonic atresias. The authors report a case of a 4-day-old male baby who presented with the features of distal intestinal obstruction. At laparotomy type I atresia of the ascending colon, just distal to cecum, was found. Primary ceco-colic anastomosis along with a covering ileostomy was performed. Ileostomy was reversed 3 weeks later.

Key words: Atresia of ascending colon, Neonatal intestinal obstruction, Early ileostomy reversal

INTRODUCTION

Atresia of the ascending colon is one of the rarest causes of neonatal intestinal obstruction. Reported incidence of colonic atresia is 1 in 20,000 live births of which ascending colon is the rarest.¹

Due to rarity of the disease large series have not been reported in the literature. Colonic atresia occurs in descending order of frequency at sigmoid, splenic flexure, hepatic flexure and ascending colon respectively.²

It may be associated with anomalies of other systems. Mortality is usually high when treatment is delayed for more than 72 hours. However prognosis is satisfactory with early diagnosis and proper management.³

Rarity of the disease prompted the authors to report this case.

CASE REPORT

A 4-day-old term male baby was born through cesarean section, to an otherwise healthy primigravida, at a peripheral private clinic. No prenatal problem was detected on routine antenatal visits.

The baby did not pass meconium till 4th day when he developed marked abdominal distension along with other features of intestinal obstruction. At the time of admission to our hospital along with distension mild dehydration was also present. There was no other apparent associated anomaly. Rectal stimulation was inconclusive.

Plain x-ray of the abdomen in erect posture showed multiple air fluid levels suggestive of distal small bowel obstruction.

A diagnosis of distal small bowel atresia was made. Baby was optimized by fluid and electrolytes replacement. Parenteral antibiotics along with vitamin K were administered.

Laparotomy was performed through a right upper transverse incision. Operative finding was type I atresia of the ascending colon just distal to hugely distended cecum [Image 1] [Image 2]. A ceco-colic anastomosis was performed by opening the lumens of dilated cecum and micro ascending colon through a longitudinal incision on their anterior walls. Intervening mucosal septum was excised and the defect closed transversely with covering ileostomy.

Address: Department of Paediatric Surgery, Mayo Hospital Lahore, Pakistan

E-mail address*: fariharoon@yahoo.com

* (Corresponding author)

Received on: 08-06-2010

Accepted on: 10-07-2010

<http://www.apspcaserep.com>

© 2010 Mansoor *et al*

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](https://creativecommons.org/licenses/by/3.0/)



Image 1: Atresia of the ascending colon with hugely distended cecum



Image 2: Mucosal septum of type I atresia and narrow lumen of ascending colon.

The recovery was smooth and patient discharged on 7th postoperative day. After 3 weeks a contrast radiograph through ileostomy was performed to confirm the distal patency before ileostomy reversal [Image 3].

DISCUSSION

Colonic atresia accounts for 1.8-15% of intestinal atresias.⁴ Ascending colon is the rarest site of colonic atresia. Due to its rarity it is usually not thought of in the differential diagnosis of neonatal intestinal obstruction.⁵

Delayed recognition of symptoms increases the risk of complications like perforation and sepsis. Etiology of this anomaly is still debated. Commonly accepted theory is that of in-utero vascular accidents in the early gestation.⁶ Colonic volvulus, intussusception, incarceration and strangulation of internal hernias in-utero, are also the probable etiological factors. Failure of recanalization after the solid cord stage as in duodenal atresia is also considered to be the cause of colonic atresia. Due to the rarity of the disease available literature is scanty. Associated anomalies like abdominal wall defects (gastroschisis),⁷ musculoskeletal disorders, small gut atresia, ocular and facial anomalies are common.⁸

APSP J Case Rep 2010; 1: 3

Colonic atresia in babies with gastroschisis seems to result from the bowel compression with narrowing of abdominal defect.⁸

Association of small bowel atresia and Hirschsprung's disease is of paramount importance.⁹ Rectal biopsy is recommended in patients who initially were treated for colonic atresia and had slow return of gut functions.¹⁰

Uncomplicated right colonic atresia can be treated with primary anastomosis with little morbidity whereas staged reconstruction with proximal diversion is advised in sigmoid and left colonic atresia to avoid the complications of anastomosis.¹⁰

Preservation of ileocecal valve is desired for future growth of the child. Due to hugely dilated cecum and the enormous disparity between the cecum and atretic ascending colon in the reported case primary cecocolic anastomosis with covering ileostomy seems appropriate. However the operative strategy depends on the clinical state of the patient and the safety of the procedure should always be a priority. In the case presented staged procedure was adopted and it resulted in early recovery and discharge of the patient. Stoma care is an issue in these cases especially with ileostomy where effluent is more fluid in nature. To address this issue an early reversal was performed in our patient. It thus appears to be an appropriate approach in a set up where stoma care may be an issue.



Image 3: Contrast radiograph through ileostomy showing cecum and narrow lumen of unused distal colon.

REFERENCES

1. Benson CD, Lotfi MW, Brough AJ. Congenital atresia and stenosis of the colon. *J Pediatr Surg* 1968; 3: 253.
2. Karnak I, Ciftci AO, Senocak ME, Tanyel FC, Buyukpamukcu NI. Colonic atresia: surgical management and outcome *Pediatr Surg Int* 2001; 17: 631-5.
3. Etensel B, Temir G, Karkiner A, Melek M, Edirne Y, Karaca I, et al. Atresia of the Colon *J Pediatr Surg* 2005; 40: 1258-68.
4. Ruggeri G, Libri M, Gargano T, Pavia S, Pasini L, Tani G, et al. Congenital colonic stenosis: a case of late-onset. *Pediatr Med Chir* 2009; 31: 100-3.
5. Winters WD, Weinberger E, Hatch EI. Atresia of the colon in neonates: Radiographic findings *Am J Radiol* 1992; 159: 1273-6.
6. Hoyme H, Higginbottom MC, Jones KL. Vascular etiology of disruptive structural defects in monozygotic twins. *Pediatrics* 1981; 67: 288-91.
7. Saxonhouse MA, Kays DW, Burchfield DJ, Hoover R, Islam S. Gastroschisis with jejunal and colonic atresia and isolated colonic atresia in dichorionic, diamniotic twins. *Pediatr Surg Int* 2009; 25: 437-9.
8. Baglaj M, Carachi R, MacCormack B. Colonic atresia: a clinicopathological insight into its etiology. *Eur J Pediatr Surg* 2010; 20: 102-5.
9. Draus JM Jr, Maxfield CM, Bond SJ. Hirschsprung's disease in an infant with colonic atresia and normal fixation of the distal colon. *J Pediatr Surg* 2007; 42: e5-8.
10. Kim CW, Superina RA and Ein S. Colonic atresia combined with Hirschsprung's disease: A diagnostic and therapeutic challenge. *J Pediatr Surg* 1995; 30: 1216-7.

How to cite

Mansoor H, Kanwal N, Shaukat M. Atresia of the ascending colon: A rarity. *APSP J Case Rep* 2010; 1: 3

Ileal Duplication Cyst Causing Recurrent Abdominal Pain and Melena

Muhammad Ali Sheikh,* Tariq Latif, Masoom Ali Shah, Imran Hashim, Jameel Akhtar

ABSTRACT

Alimentary tract duplications are rare congenital anomalies. The presentation depends on their anatomical location, size and other characteristics. The most common variety is small bowel cystic duplication. We report a case of an eight years old girl who presented with recurrent abdominal pain and melena. Radioisotope technetium scan showed increased uptake of tracer in right lower abdomen and a diagnosis of Meckel's diverticulum made. At surgery a cystic, communicating, ileal duplication found which was resected along with adjacent gut. It is thus reiterated that while investigating children with recurrent abdominal pain and melena, gut duplications must be included in the differential diagnosis.

Key words: Alimentary tract duplications, Pain abdomen, Melena

INTRODUCTION

Alimentary tract duplications (ATD) are rare congenital anomalies.¹ These lesions may be present anywhere from mouth to anus but commonly found in small bowel.² The reported incidence of these lesions is 1 in 4500.³ In 1937, Ladd coined the term "duplications of the alimentary tract" thus simplifying the terminology which previously used to be confusing.⁴ The term is applied to those lesions that had well developed coat of smooth muscle, mucosa representing some part of alimentary tract with close anatomic relation with some portion of gastrointestinal tract.

Duplications of small bowel can cause life threatening complications like volvulus, intussusception, gut perforation and massive bleeding. Herein we report one such case that remained elusive till laparotomy was performed.

Address: Department of Paediatric Surgery,

Shaikh Zayed Hospital Lahore, Pakistan

E-mail address*: drali444@yahoo.com

* (Corresponding author)

Received on: 18-07-2010

Accepted on: 30-07-2010

<http://www.apspcaserep.com>

© 2010 Sheikh et al

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](http://creativecommons.org/licenses/by/3.0/)

CASE REPORT

An eight years old girl was admitted in medical ward through emergency department because of recurrent abdominal pain and melena for the last one month. She had similar complaints since the age of 3 years. The symptoms usually settled within few days. At the time of admission she was otherwise healthy but pale looking with tenderness around umbilicus. Her weight was 19 kg, hemoglobin 6.8 gram per cent and hematocrit 21 percent. Her coagulation profile was within normal limits. Abdominal radiograph and ultrasound examination were reported as insignificant.

Radioisotope technetium scan done for suspected Meckel's diverticulum showed increased tracer uptake in right lower abdomen close to urinary bladder suggestive of ectopic gastric mucosa (Image 1). She was prepared for surgery with transfusion of packed cells and initially underwent laparoscopy. The lesion could not be localized so laparotomy was performed. The omentum was found adhered to a lesion in the mid ileum and gut was twisted around it. On separation about 4×5 centimeter cystic structure found in the mesentery of ileum two feet proximal to the cecum (Image 2). The cystic structure with adjacent part of ileum was resected and continuity of gut restored with a primary anastomosis. The duplicated part was found communicating with both proximal and distal parts of gut resected. The post-operative recovery was uneventful. Histology of resected



Image 1. Radioisotope scans showing abnormal uptake in lower abdomen (white arrow)

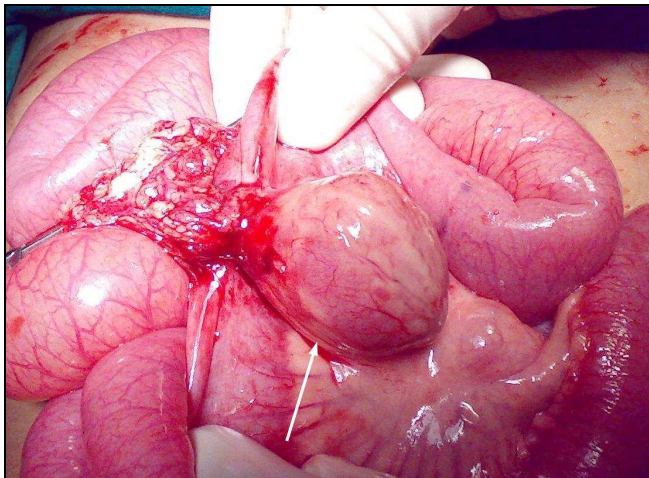


Image 2. Cystic duplication of ileum (white arrow)

gut confirmed the presence of gastric mucosa in duplicated part.

DISCUSSION

Alimentary tract duplication cysts are hollow, epithelium-lined, cystic, spherical or tubular structures that are attached to the wall of gastrointestinal tract.⁴ They may or may not communicate with the intestinal tract. Different theories had been put forward to explain the occurrence of enteric duplications but no single theory could account for all the known variants. The split notochord theory was postulated to explain the formation of neuroenteric duplications and associated vertebral anomalies.⁴⁻⁶ Some duplications of foregut and hindgut may result from partial *APSP J Case Rep* 2010; 1: 4

twinning. Others especially of ileum, may occur as a result of persistent embryological diverticula. Some portions of the alimentary tract undergo a solid stage during development followed by recanalization; therefore duplications may result from aberrant luminal recanalization.⁴ Finally intrauterine insults such as trauma or hypoxia could cause these anomalies at any level of alimentary tract thus etiology may be multi-factorial. Duplications may be found along esophagus in thorax, mid gut and in hindgut with involvement of urinary and genital system.⁷ The most common location is ileum and majority are cystic in nature.^{2,6} More than 50% of the cystic duplications in all locations have gastric mucosa. In our patient the lesion was of cystic variety with ectopic gastric mucosa.

The clinical presentation of these lesions can vary according to the age of the patient as well as anatomical location. Some may remain asymptomatic and identified on routine physical examination or during investigations for other problems.² The small bowel ATD can be an anchor point for intussusceptions or may result in volvulus, whereas long tubular duplications with proximal communication drain poorly and retention of intestinal contents can obstruct adjacent intestine. Gastric mucosa in duplication can cause ulceration, bleeding or perforation. These duplications can mimic inflammatory bowel diseases as documented by Puligandla, because six of their patients were treated as atypical Crohn's disease.² The widespread use of prenatal ultrasound scan has allowed these lesions to be detected early in gestation. With prenatal diagnosis treatment can be instituted before the onset of symptoms. Laparoscopy can be used as a diagnostic modality in cases of recurrent abdominal pain.

The diagnosis is usually not established before surgery as occurred in our patient where Meckel's diverticulum was suspected based upon radioisotope findings thus duplications must be considered in differential diagnosis of recurrent abdominal pain and occult gastro intestinal bleeding. Cystic duplications can be resected easily along with adjacent intestine and same was possible in case reported here. Presence of adhered omentum at surgery points towards episodes of inflammation of the cyst. Nowadays laparoscopy is gaining importance in managing different pediatric surgical conditions. This minimally invasive procedure was used in this patient but had to be converted to open approach, as the lesion could not be identified. With increasing learn curve we hope to use this technique more liberally.

REFERENCES

1. Khan HF, Qazi A, Burki T. Duplications of the small intestine. In: Khan NZ, Qazi A eds. Human

- duplications and accessory organs: A nature's disadvantage. Agha Gee Printers. 1999: 29-41.
2. Puligandla PS, Nguyen LT, St-Vil D, Flageole H, Bensoussan AL, Nguyen VH et al. Gastrointestinal duplications. *J Pediatr Surg* 2003;38:740-4.
 3. Schalamon J, Schleef J, Hollwarth ME. Experience with gastrointestinal duplications in childhood. *Arch Surg* 2000;385:402-5.
 4. Lund DP. Alimentary tract duplications. In: O'Neil JA, Rowe MI, Grosfeld JI, Fonkalsrud EW, Coran AG eds. *Pediatric Surgery*. Vol II. 6th Ed. St. Louis: Mosby Year Book Inc. 2006:1389-98.
 5. Choi SO, Park WH, Kim SP. Enteric duplications in children: an analysis of 6 cases. *J Korean Med Sci* 1993;8:482-7.
 6. Holcomb GW III, Gheissari A, O'Neill JA Jr, Shorter NA, Bishop HC. Surgical management of alimentary tract duplications. *Ann Surg* 1989;209:167-74.
 7. Jan IA, Shahzad M, Anwar H, Burki T, Dawood M. Hindgut duplications presenting as perianal masses. *J Coll Physicians Surg Pak* 2001;11:463-4.
-

How to cite

Sheikh MA, Latif T, Shah MA, Hashim I, Akhtar J. Ileal duplication cyst causing recurrent abdominal pain and melena. *APSP J Case Rep* 2010; 1: 4

Delayed Recognition of Type I Sigmoid-Colon Atresia: The Perforated Web Variety

Ghulam Mustafa,* Bilal Mirza, Zahid Bashir, Afzal Sheikh

ABSTRACT

Colonic atresias are the rare malformations of the colon and constitute about 1.7 to 15% of all gastrointestinal (GI) atresias. A 6-month old infant presented with recurrent episodes of sub-acute intestinal obstruction since birth. During the index admission, patient had clinical signs of complete intestinal obstruction. The patient was operated and type I sigmoid-colon atresia found which on further exploration turned out to be of perforated mucosal web variety. The resection of the involved part of colon and a primary end to oblique colo-colic anastomosis was performed.

Key words: Colonic atresia, Colonic stenosis, Perforated colonic web, Intestinal obstruction

INTRODUCTION

There are three anatomical types of colonic atresia, the less frequent being type I. This is characterized by a mucosal diaphragm, completely occupying the lumen, without sero-muscular interruption.¹ At times this web has a small opening because of which complete obstruction does not occur. This results in delayed recognition of this anomaly with life threatening consequences. Presented here is one such infant in whom this anomaly was not picked up promptly.

Address: Department of Paediatric Surgery, The Children's Hospital and Institute of Child Health Lahore, Pakistan

E-mail address*: missyou2009@yahoo.com
* (Corresponding author)

Received on: 10-06-2010 Accepted on: 01-07-2010

<http://www.apspjcaserep.com> © 2010 Mustafa *et al*

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](https://creativecommons.org/licenses/by/3.0/)

CASE REPORT

A 6-month-old male infant presented to emergency room with abdominal distension, bilious vomiting and constipation for two days. There was a history of repeated episodes of such events since birth.

On clinical examination the patient was febrile, with temperature of 100F, pulse 120/min and respiratory rate 28/min. Abdomen was distended with visible bowel loops. Bowel sounds were absent. Digital rectal examination revealed empty rectum.

Abdominal radiograph showed air fluid levels [Image 1]. Nasogastric tube was passed and intravenous fluids and antibiotics started and surgery planned.

At laparotomy dilated sigmoid colon found. On tracing it further distally a narrowed segment reached [Image 2]. On longitudinal enterotomy over the narrowed area, a web found which had an opening in its center. The margins of the colon over the web and few centimeters of colon around the stenosed area were inflamed and tear easily. This portion of colon was resected and an end to oblique primary colo-colic anastomosis performed.



Image 1: Xray abdomen erect posture showing air fluid levels indicating intestinal obstruction.



Image 2: Operative picture showing a distended colon proximal to the web

The post operative recovery was uneventful. Naso-gastric tube was removed on 4th post operative day and patient started orally. On 7th post-operative day patient was discharged. The histopathology of the specimen revealed inflammation and presence of ganglion cells. Patient remained well at 6 months follow up.

DISCUSSION

Gut atresias are commonly found in small intestine and colon is a rare site for this anomaly. The reported incidence of colonic atresia is 1 in 20,000 live births. In a series of 277 patients over a period of 25 years, Vecchia et al, reported colonic atresia in 8% cases only.¹⁻³

The most commonly accepted theory of development of intestinal atresia is based on vascular accidents occurring during the course of fetal development and this mechanism is supposed to be the most probable etiology of the colonic atresia and stenosis.¹

There are three types of colonic atresia. Type I atresia is characterized by a diaphragm inside the lumen and the serosal surfaces of intestine, proximal and distal to atresia, are uninterrupted. Type III colonic atresia is the most frequently reported type.^{1,4} In our case the there was a web (type I) which was perforated at the centre.

The preoperative diagnosis of colonic atresia and stenosis can be made accurately without the help of various sophisticated diagnostic modalities. Abdominal radiograph may show non-specific intestinal obstruction and in about 10% of cases pneumoperitoneum may occur. Any newborn with a suspicion of intestinal atresia on clinical grounds and plain radiograph findings should have a contrast enema to delineate the unused colon and ascertain any colonic atresia or stenosis. In case of colonic atresia the contrast will typically fill the lumen of distal unused colon, terminating at a point adjacent to the segment that is most distended with luminal air – the cutoff point.^{1,3,5-7} The age of our patient was 6-month thus possibility of colonic atresia was ruled out though stenosis was not suspected. In this case mechanical obstruction of the small intestine was the pre-operative diagnosis.

Various types of surgical procedures are adopted for this anomaly depending upon anatomical location, type of atresia and condition of the baby, though primary repair without any covering stoma is recommended in all types. Due to its association with Hirschsprung's disease a biopsy of the distal colon and rectum has been advised in all cases. The experience of local coloplasty technique in such cases is very limited and thus not recommended yet.^{1,4,5}

A review of literature revealed only one case of congenital colonic stenosis that presented in the infantile age (4-month).⁶ In our case, the patient presented in acute obstruction at 6 months of age though he had recurrent episodes of intestinal obstructions since birth that was not investigated properly. A timely recognition of symptoms would have reduced prolonged morbidity.

REFERENCES

1. Oldham KT, Arca MJ. In: Grosfeld JL O'Neill JA Jr, Coran AG, Fonkalsrud EW, Caldamone AA. editors. Pediatric Surgery. 6th ed. Chicago: Year Book; 2006. p. 1493-1501.

2. Siu KL, Kwok WK, Lee WY, Lee WH. A male newborn with colonic atresia and total colonic aganglionosis. *Pediatr Surg Int* 1999;15:141-2.
 3. Vecchia LKD, Grosfeld JL, West KW, Rescorla FJ, Scherer LR, Engum SA. Intestinal atresia and stenosis: a 25 year experience with 277 cases. *Arch Surg* 1998;133:490-7.
 4. Chadha R, Sharma A, Roychoudhury S, Bagga D. Treatment strategies in the management of jejunoileal and colonic atresia. *J Indian Assoc Pediatr Surg* 2006;11:79-85.
 5. Fairbanks TJ, Kanard RC, Del Moral PM. Colonic atresia without mesenteric vascular occlusion. The role of the fibroblast growth factor 10 signaling pathway. *J Pediatr Surg* 2005;40: 390-6.
 6. Ruggeri G, Libri M, Gargano T, Pavia S, Pasini L, Tani G, et al. Congenital colonic stenosis: a case of late-onset. *Pediatr Med Chir* 2009; 31:130-3.
 7. Kannan S, McGreevy PS, Fullerton TE. Nonsteroidal anti-inflammatory drug induced duodenal web. *S D J Med* 1997; 50: 393-4.
-

How to cite

Mustafa G, Mirza B, Bashir Z, Sheikh A. Delayed recognition of type I sigmoid-colon atresia: The perforated web variety. *APSP J Case Rep* 2010; 1:5

CASE REPORT

OPEN ACCESS

Gastric Duplication Cyst Presenting as Acute Abdomen: A Case Report

Kanchan Kayastha*, Afzal Sheikh

ABSTRACT

Gastric duplication cysts are rare variety of gastrointestinal duplications. Sometimes they may present with complications like hemorrhage, infection, perforation, volvulus, intussusception and rarely neoplastic changes in the gastric duplication cyst. We present one and half year old male child who developed sudden abdominal distension with pain and fever for two days. Ultrasound revealed a cystic mass in the hypochondrium and epigastric regions. On exploration an infected and perforated gastric duplication cyst was found. Surgical excision of most part of cyst wall with mucosal stripping of the rest was performed. Histopathology confirmed the diagnosis of gastric duplication cyst. Early surgical intervention can result in good outcome.

Key words: Gastric duplication cyst, Acute abdomen, Peritonitis

INTRODUCTION

Gastric duplication cysts constitute about 2-7 % of all gastrointestinal duplications. Majority of gastric duplication cysts are large and non-communicating. Most of the cases of gastric duplication cysts present in early age, however, in some cases patient may remain asymptomatic for a long period and might present with sudden onset of abdominal distension, pain, signs of obstruction, peritonitis etc.¹ In this report we present a patient with complicated gastric duplication cyst.

CASE REPORT

A male baby of one and half year presented in emergency with fever, abdominal pain and distension for two days. Fever was high grade and not associated with rigors and chills. Pain abdomen was continuous and patient had with few bouts of non bilious, non projectile vomiting. The past medical and surgical history was unremarkable.

Address: Department of Paediatric Surgery, The Children's Hospital and Institute of Child Health Lahore, Pakistan

E-mail address*: Kanchan_kayastha@hotmail.com
* (Corresponding author)

Received on: 22-07-2010 Accepted on: 05-08-2010

http://www.apspcaserep.com © 2010 Kayastha *et al*

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](http://creativecommons.org/licenses/by/3.0/)

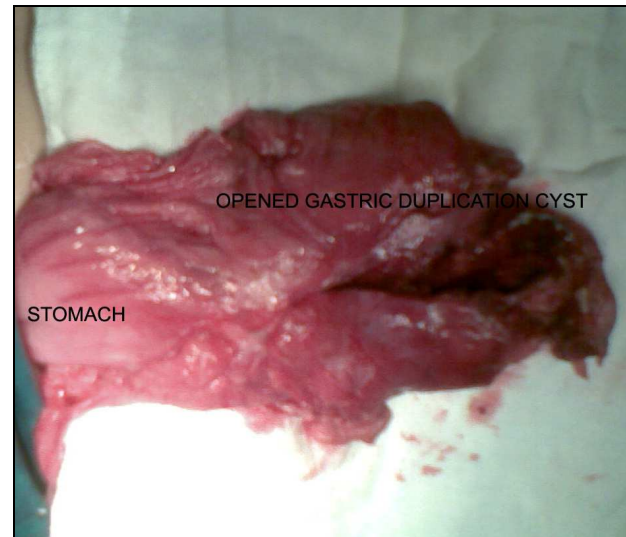


Image is showing gastric duplication cyst (surgically opened) attached intimately at greater curvature of stomach

General physical examination revealed temp 101°F, pulse 100/min and respiratory rate 30/min. On abdominal examination generalized rigidity and guarding were present. No mass or viscera could be palpated. Bowel sounds were audible. A clinical diagnosis of acute peritonitis was made. X-ray abdomen was unremarkable and ultrasound showed a cystic structure of 4cm x 5cm size in the left hypochondrium and epigastrium. Urgent laparotomy was planned after initial stabilization.

At operation a cyst having intimate contact with the greater curvature of the stomach was found. It was

perforated at its posterior surface and about 500ml of pussy fluid was drained from the peritoneal cavity. The cyst was not communicating with the stomach but shared a common wall. Excision of major part of cyst was done with mucosal stripping of the residual wall attached to the stomach. Peritoneal lavage was done and a drain placed. Post-operative recovery was uneventful. Patient was discharged on 8th day following surgery. Histopathology revealed gastric mucosa and smooth muscles in the wall of cyst. At six months follow up patient remained well.

DISCUSSION

Gastric duplications are very uncommon congenital anomalies. They usually arise at greater curvature of stomach. Almost all gastric duplications are cystic in nature. Usually they are non-communicating. In our case gastric duplication was at greater curvature, cystic in nature and non-communicating. The clinical presentations of gastric duplication cysts depends upon site, size, communication with part of the alimentary tract and associated complications.^{2,3}

In complicated cases patient may present with an acute abdomen, peritonitis or even pancreatitis. Other complications include hemorrhage, infection, perforation of the cyst and compression on surrounding structures. Clinical features thus vary. Our patient presented with fever, pain and abdominal distension suggestive of complications like infection and perforation of the cyst.^{4,5}

The differential diagnoses of gastric duplication cyst include omental cyst, mesenteric cyst, choledochal cyst, ovarian cyst, hydronephrosis etc. Ultrasound abdomen, contrast GI studies, CT-scan and MRI are helpful diagnostic modalities. In our case ultrasound abdomen did show a cyst.^{1,5}

The treatment of gastric duplication cyst is surgical resection of the cyst. Due to its close connection with the adjacent gut usually it is very difficult to completely excise therefore partial resection with the mucosal stripping of the remaining cyst is recommended as done in present case. The diagnosis of duplication cyst is based upon presence of GIT mucosa in cyst with smooth muscle coat in the wall. Cyst must have an intimate contact with any part of GIT. In our patient all these features were present. Early diagnosis and prompt surgical intervention with optimal surgical procedure carries a good prognosis.^{1,3,6}

REFERENCES

1. Agha F, Gabriele O, Abdulla F. Complete gastric duplication. *Am J Radiol* 1981;137: 406-7.
2. Di Pisa M, Curcio G, Marrone G, Milazzo M, Spada M, Triana M. Gastric duplication associated with pancreas divisum diagnosed by a multidisciplinary approach before surgery. *World J Gastroenterol* 2010;16:1031-3.
3. Sinha C.K, Nour S, Fisher R. Pyloric duplication in newborn: A rare cause of gastric outlet obstruction. *J Indian Pediatr Surg* 2007;12: 34-5.
4. Kumar K, Joshi M, Vishwanath N, Akhtar T, Oak S. Neonatal lingual gastric duplication cyst: A rare case report. *J Indian Assoc Pediatr Surg* 2006;11:97-8.
5. Kuraoka K, Nakayama H, Kagawa T, Ichikawa T, Yasui W. Adenocarcinoma arising from gastric duplication cyst with invasion to the stomach: a case report with literature review. *J Clin Pathol* 2004;57: 428-31.
6. Lund D.P. Alimentary tract duplications. In: O'Neil JA, Rowe MI, Grosfeld JL, Fonkalsrud WE, Coran AG. Editors. *Pediatric Surgery*, 6th ed, Philadelphia: Mosby, 2006:1389-98.

How to cite

Kayastha K, Sheikh A. Gastric duplication cyst presenting as acute abdomen: A case report. *APSP J Case Rep* 2010; 1: 6

CASE REPORT

OPEN ACCESS

A Giant Lymphatic Cyst of the Transverse Colon Mesentery

Tayyaba Batool, Soofia Ahmed, Jamshed Akhtar*

ABSTRACT

Mesenteric cysts are not uncommon in pediatric age group but giant lymphatic cysts of mesentery are reported infrequently. This is a report of six years old female who had vague abdominal pain with distension for two years. Investigations revealed a large cystic mass in abdomen. On exploration a giant lymphatic cyst in the mesentery of transverse colon found. More than 1500 ml of milky fluid was drained. The cyst was unilocular and appeared to be the collection of lymph (chyle) between two leaves of the mesentery of the transverse colon. It is postulated that trauma to or malformation of lymphatics at the root of mesentery might have lead to this pathology.

Key words: Lymphatic cyst, Mesenteric cyst, Lymphatic malformation, Abdominal mass

INTRODUCTION

Mesenteric cyst is a term applied to any cyst found in mesentery. Lymphatic cyst is a type of mesenteric cyst which is of lymphatic origin. It is usually a benign lesion. A closely related pathology is cystic hygroma which is also of lymphatic origin. This is usually found in the retroperitoneum. These are more common in small bowel mesentery but rarely reported in colonic mesentery.^{1,2}

Lymphatic cysts usually contain pale yellow fluid. Chylous cysts are infrequently seen. Chylous cysts are classified as rare variants of mesenteric cysts. It constitutes 7.3% to 9.5% of all abdominal cysts.²

We report a case of giant mesenteric cyst at unusual location and with unique features.

Address: Department of Paediatric Surgery, National Institute of Child Health Karachi, Pakistan

E-mail address*: jamjim88@yahoo.com
* (Corresponding author)

Received on: 22-07-2010 Accepted on: 11-08-2010

<http://www.apspcaserep.com> © 2010 Batool *et al*

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](http://creativecommons.org/licenses/by/3.0/)

CASE REPORT

A six years old female child weighing 12 kg, admitted with history of gradual abdominal distension that was noticed by the mother for the last 2 years but did not seek any medical advice as there were no other associated complaints. With an increase of abdominal girth and vague abdominal pain, parents took the child to a nearby general practitioner who advised ultrasound and referred the child to our facility. On examination child appeared comfortable with abdominal fullness. Abdominal examination did not reveal a distinct mass though vague fullness was present oriented more in vertical direction. There was a definite feel of fluid thrill on palpation.

Ultrasound done previously showed cystic mass in abdomen. With female gender a differential diagnosis included ovarian cyst in addition to mesenteric and omental cyst. CT scan abdomen was advised. This showed a large cystic mass occupying almost whole of the abdomen and located under anterior abdominal wall with viscera pushed posteriorly [Image 1] [Image 2]. A diagnosis of omental cyst was made at this stage.

Laparotomy was performed. On opening peritoneum a huge lymphatic cyst was found [Image 3]. It was delivered out with difficulty and that too after partially evacuating it. More than 1500 ml of milky fluid was drained out. The anatomy was then identified more clearly. It was found between leaves of transverse colon mesentery and extending into its root [Image 4]. The redundant leaves of mesentery were excised without jeopardizing blood supply of colon and left open so as to prevent re-accumulation of fluid. Drain was kept and wound closed.

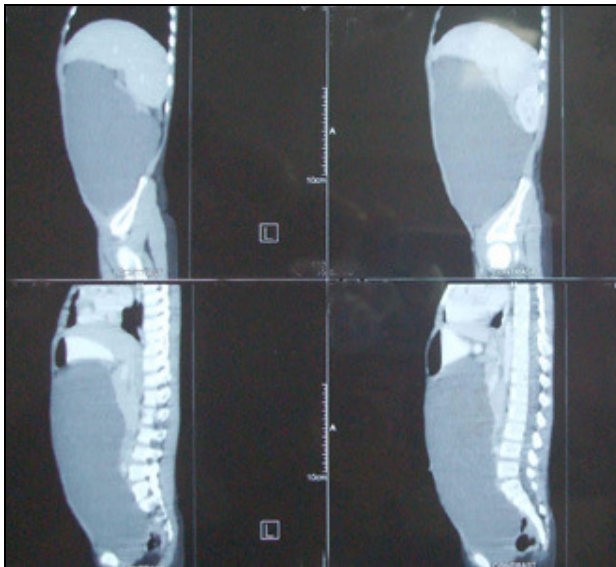


Image 1: CT scan, sagittal section showing huge unilocular cyst

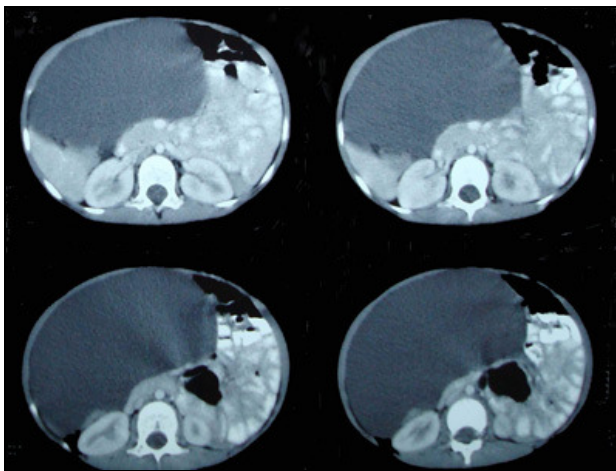


Image 2: CT scan showing position of the cyst just beneath the anterior abdominal wall with compression of the adjacent viscera



Image 3: Operative view of chylous cyst

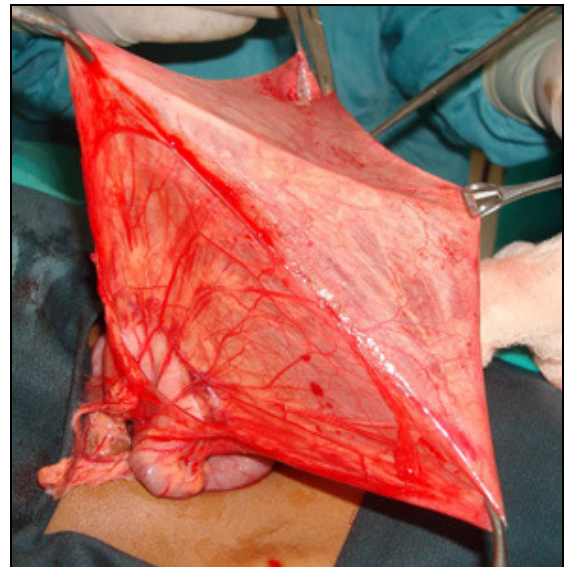


Image 4: Stretched out leaves of mesentery following drainage of fluid

Post-operative course was uneventful and patient discharged on 4th day. Biopsy was reported as consistent with mesenteric cyst. Parents were counseled again as to possible recurrence of the lesion and a regular follow up was planned. At three months follow up child is well. On ultrasound a small hypoechoic area of 4cm x 4cm was noted in mid abdomen. A repeat ultrasound after a month is advised to note the progress of the lesion.

DISCUSSION

Cyst of lymphatic origin can be found within peritoneum (omentum, mesentery) as well as in the retroperitoneal area. One of the theories proposed that these cysts represent benign proliferations of ectopic lymphatics. These channels lack communication with the main lymphatic system. Other theory suggested that due to the failure of joining venous system embryonic lymph channels gradually dilate. Still one of the theories suggested that as a result of non-fusion of leaves of mesentery, lymphatic fluid accumulates within this, supposedly a dead space. They can also result from trauma to the lymphatic channels.³ Usually these cysts are multilocular or multiseptated. Huge unilocular cysts are rare. In the patient reported here it was a huge unilocular cyst within the leaves of the mesentery of transverse colon.

These cysts mostly remain asymptomatic and only come into attention when they cause abdominal fullness / distension or vague discomfort. In this patient it caused fullness and mild abdominal pain because of which parents seek advice. Though chronic symptoms predominate acute presentation is not uncommon which includes intestinal obstruction, volvulus, hemorrhage into

cyst, rupture etc.^{4,5} Clinical examination at times may not pick these cysts as distinct masses, rather a feeling of ascites (fluid accumulation) may be the only sign, as happened in our patient.

Abdominal ultrasonography usually provides working diagnosis in these patients and other investigations may be avoided though a more detailed picture can be provided by CT scan abdomen which may help in planning surgery. In reported patient CT scan revealed excellent details of the lesion. Thus where possible this modality may be used for diagnostic purposes.

Surgery is usually a straight forward affair. At times lesion can be removed completely without sacrificing any adjacent organ though resection of involved segment of small bowel, if limited area is involved, is more appropriate. The lesions of the mesentery can be excised as complete as possible, though in difficult cases and if cysts are at critical location de-roofing of the cysts is another approach, a type of marsupialization. In our patient the leaves of mesentery were extremely stretched out. Redundant part was excised easily. No well defined cyst found in this case. The histopathology reported the walls of lesion as of mesenteric origin with mesothelial lining. Considering lack of well defined limits of the lesion there are chances of recurrence of cysts in such cases. A regular follow up is thus advised. In our patient same is being followed.

This case had many unusual features, namely being unilocular, of huge size containing milky fluid, in the mesentery of transverse colon and with apparently no distinct cystic structure. It appeared that mesenteric

leaves simply stretched out due to accumulation of fluid. Presence of milky fluid within transverse colon mesentery may be explained on the basis of either malformed ruptured lymphatic channels of some trauma that might have caused leak of lymphatic fluid at the base of mesentery. This case may add to host of postulations related to lymphatic mesenteric cysts.

REFERENCES

1. Iida S, Furukawa K, Terada Y, Sugisaki Y, Yoshimura K, Tajiri T. A case of a mesenteric cyst in the sigmoid colon of a 3 year old girl. *J Nippon Med Sch* 2009;76:247-52.
2. Rattan KN, Nair VJ, Pathak M, Kumar S. Pediatric chylolymphatic mesenteric cyst - a separate entity from cystic lymphangioma: a case series. *J Med Case Rep* 2009; 3:111.
3. Pantanowitz L, Botero M. Giant mesenteric cyst: A case report and review of the literature. *Internet J Pathol* 2001;1. Available at file:///C:/Documents%20and%20Settings/PC/Desktop/New%20Folder%20(2)/Mes%20cys.htm
4. Orobitg FJ, Vázquez L, De Franceschini AB, Ramos-Ruiz E. Mesenteric cyst of lymphatic origin: a radiopathological correlation and case report. *P R Health Sci J* 1994;13:171-4.
5. Kumar S, Agrawal N, Khanna R, Khanna AK. Giant lymphatic cyst of omentum: a case report. *Cases J* 2009;2:23.

How to cite

Batool T, Ahmed S, Akhtar J. A giant lymphatic cyst of the transverse colon mesentery. *APSP J Case Rep* 2010; 1: 7

CASE REPORT

OPEN ACCESS

Broken Piece of Silicone Suction Catheter in Upper Alimentary Tract of a Neonate

Bilal Mirza,* Muhammad Saleem, Afzal Sheikh

ABSTRACT

Esophageal foreign bodies (FB) are common in adults and children. These are rarely reported in infants and neonates. A 2-day-old newborn was referred to our hospital with history of accidental intrusion of soft silicone suction catheter into the upper gastrointestinal tract (GIT). X-ray chest and abdomen confirmed the presence of suction tube in esophagus and stomach. The suction catheter was retrieved successfully at direct laryngoscopy.

Key words: Esophageal foreign body, Neonate, Laryngoscopy

INTRODUCTION

Esophageal foreign bodies are commonly encountered in adults and children.^{1,2} The common esophageal foreign bodies in children are coins, beads, fish bone etc. Small and smooth gastro-esophageal FBs that can pass through the pylorus may be observed for spontaneous passage through anus. However, large / sharp FBs may need active intervention to avoid their symptoms and complications.^{2,3}

Esophageal FBs in infants are rare; rarer still is their occurrence in newborn. Only four cases of neonatal GIT foreign bodies have been reported in Pubmed.¹ We report probably youngest of all the neonates with esophageal foreign body.

CASE REPORT

A two-day-old male baby was referred to our institution with complaints of vomiting after every feed since birth. The referral letter revealed that the baby was born through spontaneous vaginal delivery in a private hospital; and during newborn resuscitation, the silicone suction catheter was accidentally detached from the suction machine piping and baby swallowed it.

Since then the patient started vomiting after every feed.

The baby was vitally stable. A radiograph of chest and upper abdomen showed a tube curled up in stomach and esophagus. Patient was taken to the operation theatre and with direct laryngoscopy under general anesthesia a soft silicon suction catheter was retrieved from the upper esophagus. [Image 1] [Image 2].

Patient showed uneventful recovery and was symptoms free at 3 months follow up.

DISCUSSION

Esophageal foreign bodies are rarely encountered in neonates. After that age, when the milestone of grasping and putting everything in mouth are achieved, the incidence of FB in GIT starts rising. About 80% of all pediatric GIT foreign bodies occur between 6 months and 3 years of age. Zameer *et al* extensively reviewed the literature regarding GIT foreign bodies in neonates. They were able to find only three cases.¹

The usual symptoms of a FB in esophagus are nausea, vomiting, dysphagia, respiratory difficulty, neck pain and hematemesis. The symptoms usually depend upon the type, size and nature of the FB. Esophageal FB can, sometimes, create fatal complications such as esophageal perforation, subcutaneous emphysema, retro-esophageal abscess, pneumothorax and pneumomediastinum, esophago-aortic fistula, mediastinitis and lung abscess.^{1,3-6}

There are three types of esophageal FBs. Small esophageal FBs such as beads, buttons, rubber pieces,

Address: Department of Paediatric Surgery, The Children's Hospital and Institute of Child Health Lahore, Pakistan

E-mail address*: blmirza@yahoo.com
* (Corresponding author)

Received on: 12-06-2010 Accepted on: 11-07-2010

http://www.apspcaserep.com © 2010 Mirza *et al*

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](https://creativecommons.org/licenses/by/3.0/)



Image 1: Retrieval of the FB with direct laryngoscopy

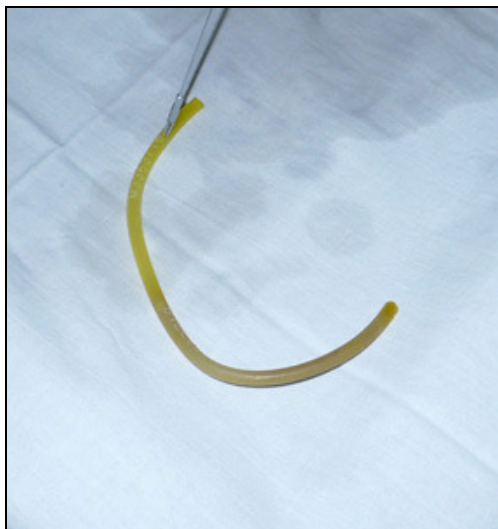


Image 2: Silicone suction catheter after retrieval

and button batteries etc; large FBs such as impacted bolus of meat, coins, ornaments and fish bone; and lastly the long FB such as piece of wood, long piece of bone and bezoars. The reported soft FBs are pieces of rubber, rubber-eraser, rubber pallets and buttons, pieces of plastic bags, paper, pieces of clothes, meat, surgical gauze and sponges.^{1,4,6-8}

Most of small FBs do not need any active management as about 80% of all FBs pass spontaneously from GIT.⁴ Large and long FBs usually need intervention for their retrieval.⁴⁻⁶ It is estimated that only 10-20% of all GIT foreign bodies require endoscopic retrieval, whereas, only

1% require surgical exploration.¹ The management options for esophageal FBs are; retrieval by direct laryngoscopy; esophagoscopy and by open method.^{1,4,7,8} Small foreign bodies usually at the level of cricopharyngeus are removed by direct laryngoscopy. The examples are coin, beads and fish bone retrieval. The foreign bodies below this level usually require esophagoscopy or open surgical procedures for their removal.^{1,2,4,6}

Our case is unique because of unusual age of presentation, the mode of intrusion and the FB residing in stomach and esophagus that was retrieved successfully with direct laryngoscopy. It is inferred that casual newborn resuscitation carries a number of life threatening risks for the patient. It may end up in iatrogenic-FBs in the alimentary tract of newborn as happened in this case. Moreover, early referral to a specialized tertiary centre and prompt management can avoid many complications.

REFERENCES

1. Zameer M, Kanojia RP, Thapa BR, Rao K. Foreign body oesophagus in a neonate: A common occurrence at an uncommon age. *Afr J Paediatr Surg* 2010; 7:114-6.
2. Nayak SR, Kirtane MV, Shah AK, Karnik PP. Foreign bodies in the cricopharyngeal region and oesophagus (a review of 226 cases). *J Postgrad Med* 1984;30:214.
3. Pandya VK, Malik R, Lakhole M. Rigid 12 inches long impacted foreign body in upper gastro-intestinal-tract. *Ind J Radiol Imag* 2006;16:495-7.
4. Al Shehri GY, Al Malki TA, Al Shehri MY, Ajao OG, Jastaniah SA, Haroon KS, et al. Swallowed foreign body: Is interventional management always required? *Saudi J Gastroenterol* 2000;6:84-6.
5. Baraka A, Bikhazi G. Oesophageal foreign bodies. *Br Med J* 1975;1:561-3.
6. David SS, Subbiah B. Foreign bodies in the air and food passages in children. *Indian Paediatr* 1973;10:183-4.
7. Patil KK, Patil SK, Gorad KP, Panchal AH, Arora SS, Gautam RP. Intraluminal migration of surgical sponge: Gossypiboma. *Saudi J Gastroenterol* 2010;16:221-2.
8. Bakhshaeekia A, Hosseini SM, Razmi T, Shamsaefar A. Bowel perforation by crumpled paper in a patient presenting with acute abdominal pain. *Saudi J Gastroenterol* 2009;15:52-4.

How to cite

Mirza B, Saleem M, Sheikh A. Broken piece of silicone suction catheter in upper alimentary tract of a neonate. *APSP J Case Rep* 2010; 1: 8

Hyposkillia and Critical Thinking: Lost Skills of Doctors

Tayyaba Batool

Dear Sir,

Imperforate hymen is the most frequent obstructive anomaly of the female genital tract. It can be diagnosed on prenatal ultrasound as bladder outlet obstruction, due to hydrocolpos or mucocolpos¹ and postnatally on physical examination of the perineum and abdomen. No additional tests or investigative tools are required for diagnosis per se. We share our experience of two index cases of imperforate hymen where clinical diagnosis was missed.

Case 1: A 21-day-old female baby was presented with complaint of urinary retention. Since bladder was palpable on examination, catheterization was done by the nursing staff on advice of a resident. Urinalysis and ultrasound were advised which turned out to be insignificant. On 5th day of admission it was noticed that despite catheterization lower abdomen was still distended. Perineal examination done at that time revealed imperforate hymen [Image 1].

Case 2: Twelve years old female presented with lower abdominal mass. She was catheterized elsewhere for urinary retention but mass persisted. She was admitted and CT scan abdomen advised which picked up a cystic mass in pelvis [Image 2]. Surgery planned and on operation table perineal examination revealed an obvious bulge at vaginal orifice.

Hymenotomy was performed in both the cases.

Critical thinking is about how we approach to the problems, questions and issues.² Learning clinical skills is an art and critical thinking is part and parcel to acquire this art.

Hyposkilliacs are “physicians who cannot take an adequate medical history, cannot perform a reliable physical examination, cannot critically assess the information they gather, cannot create a sound management plan, have little reasoning power, and communicate poorly. They learn to order all kinds of tests and procedures but don't always know when to order [them] or how to interpret them”³



Image 1: A neonate with imperforate hymen - An obvious clinical finding not to be missed.



Image 2: CT scan showing distended vagina and displaced urinary bladder.

The requisite of producing good clinicians is to create an environment of inquiry. The learner or clinician will analyze and evaluate the patient and disease with his/her cognitive skills only if his brain is primed to do so. Otherwise the culture of technological overuse will keep on flourishing; and we will end up in producing clinicians unable to pick up simple clinical diagnoses with poor basic knowledge and skills. This is high time to revisit the

training methods for junior doctors, to be addressed conscientiously.

REFERENCES

1. Hillard PJA, e- medicine, Imperforate Hymen, Department of Gynae/Obs, Stanford University Medical Centre: Updated Feb 24, 2010. Accessed on June 13, 2010. Available from; <http://emedicine.medscape.com/article/269050-overview>
2. Facione PA. Critical Thinking: What it is and why it counts? 2010 update. Insight assessment. Millbrae, CA: The California Academic Press. 2010 Available from; http://www.insightassessment.com/pdf_files/what&why2006.pdf
3. Fred HL. Hyposkillia – Deficiency of Clinical Skills. Tex Heart Inst J. 2005; 32: 255-7.

Tayyaba Batool

Address: Department of Paediatric Surgery,
National Institute of Child Health Karachi, Pakistan.

Email: docnich2003@yahoo.com

Received on: 10-06-2010

Accepted on: 06-07-2010

<http://www.apspcaserep.com>

© 2010 Batool

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](http://creativecommons.org/licenses/by/3.0/)

How to cite

Batool T. Hyposkillia and critical thinking: Lost skills of doctors. APSP J Case Rep 2010; 1: 9

Overlapping Features of Caudal Regression Syndrome and VACTERL Complex in a Neonate

Lubna Ijaz,* Afzal Sheikh

Dear Sir,

Caudal regression syndrome (CRS) is characterized by a group of heterogeneous anomalies involving the distal spinal cord and vertebral column, genitourinary system, hind gut and limbs. The malformation may range from minor anomalies of spine and spinal cord to the extreme, the sirenomelia. Various authors pointed out an overlap of spectrum of anomalies in CRS and VACTERL (vertebral, anorectal, cardiac, tracheo-esophageal, renal and limb anomalies) complex.¹⁻³ A case of a neonate is presented in whom the spectrum of congenital anomalies overlapped between CRS and VACTERL complex.

A 1-day-old male neonate weighing 2.8 kg presented to the emergency room of our institution with imperforate anus and absent left lower limb. The baby was a product of consanguineous marriage. The newborn was vitally stable and examination revealed imperforate anus, left lower amelia, sacral agenesis, megameatus intact prepuce, left sided undescended testis (UDT) and a small tag of tissue in place of the absent limb [Image].

Patient was admitted and routine neonatal care provided. Baby was subjected to various investigations. X-ray invertogram revealed high gas shadow in abdomen and sacral agenesis. Ultrasonography of the abdomen showed absent left kidney. Parents were counseled about the condition and a sigmoid loop colostomy performed for imperforate anus and discharged after two days.

CRS is a heterogeneous group of anomalies that may involve distal spine, spinal cord, genitourinary system, hind gut, and limbs; whereas, existence of VACTERL complex in a patient is considered where at least three of six anomalies of this complex are present.¹⁻³

Few authors have reviewed more than 150 patients of sirenomelia and depicted at least three out of six anomalies of VACTERL complex in more than 98% of the patients.



Image: Photograph of the patient showing spectrum of anomalies including imperforate anus, left lower amelia, left UDT, megameatus and a skin tag in place of absent limb

Shah et al¹, in 2006, reported a case of sirenomelia associated with esophageal atresia and tracheo-esophageal fistula.^{1,4,5} Our patient had four out of six anomalies of VACTERL complex and also fulfilled the criteria of caudal regression syndrome.

It therefore appears that caudal regression syndrome and VACTERL complex may be the same spectrum of anomalies that coexist in various combinations. As an overlap of the anomalies encountered, the use of the term caudal regression syndrome may be reserved for those cases having only caudal spinal and spinal cord anomalies, an event in isolation too rare, without multiple anomalies that usually overlap with the VACTERL complex. Moreover, it appears that both multi-anomaly-CRS and VACTERL anomalies may have similar etiological factors, a point that can be debated as more of such cases are reported.

REFERENCES

1. Shah DS, Tomar G, Preetkiran. Sirenomelia. *Indian J Radiol Imaging* 2006; 16:203-4.
2. Fayyaz A, Ilyas M, Iqbal O. Pre-natal diagnosis of caudal regression syndrome. *J Coll Physicians Surg Pak* 2007; 17: 425-6.
3. Taori KB, Mitra K, Ghonga NP, Gandhi RO, Mammen T, Sahu J. Sirenomelia sequence (mermaid): Report of three cases. *Indian J Radiol Imaging* 2002; 12:399-401.
4. Duncan PA, Shapiro LR, Klein RM. Sacrococcygeal dysgenesis association. *Am J Med Genet* 1991; 41: 153-61.
5. Stocker JT, Heifetz SA. Sirenomelia: a morphological study of 33 cases and review of the literature. *Perspect Pediatr Pathol* 1987; 10: 7-50.

*Corresponding author: **Lubna Ijaz**

Address:

Department of Paediatric Surgery,
The Children's Hospital & The Institute of Child Health,
Lahore, Pakistan.

Email: doctorlubnabil@yahoo.com

Received on: 17-06-2010

Accepted on: 27-06-2010

<http://www.apspcaserep.com>

© 2010 Ijaz *et al*.

This work is licensed under a [Creative Commons Attribution 3.0 Unported License](https://creativecommons.org/licenses/by/3.0/)

How to cite

Ijaz L, Sheikh A. Overlapping features of caudal regression syndrome and VACTERL complex in a neonate. *APSP J Case Rep* 2010; 1: 10

Pancreatic Calcification

Bilal Mirza

An 8-year old girl presented to the emergency room with the complaints of acute epigastric pain and vomiting for two days. Vomiting was non-bilious. There was a past history of recurrent self-limiting similar episodes during the last one year. Patient was prescribed proton pump inhibitors (PPI) and oral suspensions such as sucralfate. There was no history of trauma, rash and jaundice.

General physical examination was unremarkable. There was mild tenderness in the epigastrium on palpation; otherwise abdomen was soft and not distended. Hemoglobin was within normal limits and WBC's count of 10,500. Random blood glucose, serum electrolytes and renal functions were in normal range. Serum amylase was 113 i.u (Normal <96 i.u.). An abdominal radiograph was requested that delineated calcifications in the region of pancreas. [Image1]



Image 1: Plain x-ray abdomen showing calcification in the region of pancreas (arrow)

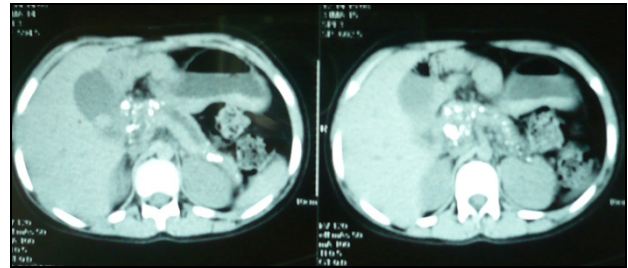


Image 2: CT scan of abdomen revealing a dilated pancreatic duct, pancreatic calcification and gallstones

CT scan of abdomen showed a dilated pancreatic duct and calcification in the pancreas and gallstones [Image 2]. A diagnosis of chronic recurrent pancreatitis with gall stones made and patient provided supportive medical therapy that ameliorated her symptoms. Further workup and possible surgical intervention is planned on her.

DISCUSSION

Pancreatic calcification is a diagnostic feature of chronic pancreatitis even in the absence of the clinical signs and symptoms. Pancreatic calcification is seen on radiographs in about 30-50% of patients with chronic pancreatitis in adults. Pancreatic calcification is rarely reported in children below ten years, however, its incidence increases after this age¹

The etiology of the chronic relapsing pancreatitis in children includes; trauma, anatomic abnormalities of the pancreatobiliary ducts, hereditary causes, systemic diseases, choledocholithiasis and cholelithiasis, cystic fibrosis, drugs, inflammatory bowel disease, and infections.^{1,2}

Recurrent attacks of the pancreatitis results in progressive damage of the exocrine as well as endocrine functions of the pancreas. Later on, proteinaceous plugs get deposited in the pancreatic ducts and with the passage of time they calcify and can be picked up radiologically. During the relapse of the pancreatitis, patient may present with the features of the acute pancreatitis such as severe epigastric pain and vomiting.³

In pediatric age group the important cause of the chronic pancreatitis is the congenital anomalies of the pancreas

and pancreatobiliary ducts. The important malformations that may present with chronic pancreatitis are choledochal cyst, anomalous union of pancreatobiliary ducts without choledochal cyst, pancreas divisum and annular pancreas. Gallstone disease is another frequently identified cause of the chronic pancreatitis in children. It is usually associated with hemolytic disorders such as thalassemia and hereditary spherocytosis.^{1,2}

The anatomical abnormalities of the pancreatobiliary ducts can be diagnosed using modalities like ultrasonography, computed tomography (CT) scan, endoscopic retrograde pancreatography (ERCP) and magnetic resonance pancreatography (MRCP).^{1,4}

During the relapse of the pancreatitis the supportive management includes nothing by mouth, nasogastric decompression in case of recurrent vomiting, analgesics, PPI, parenteral fluids, antibiotics and nutrition. Some times octreotide infusion has to be instituted in cases of intractable pain.¹

The surgical management of chronic relapsing pancreatitis depends upon the etiological factors. In case of gallstones a cholecystectomy may offer relief of the symptoms, however, in case of congenital malformations of the pancreatobiliary systems, major surgical interventions are needed. The procedures may range from simple sphincterotomy and sphincteroplasty to the pancreatectomy and pancreatojejunostomy (Puestow, Duval procedure) or pancreatogastrostomy (Smith procedure). It should be kept under consideration that the mortality and morbidity is high in patients undergoing surgical interventions for chronic pancreatitis.¹

REFERENCES

1. Miyano T. In: Grosfeld JL O'Neill JA Jr, Coran AG, Fonkalsrud EW, Caldamone AA. editors. Pediatric Surgery. 6th ed. Chicago: Year Book; 2006. p. 1671-8.
2. Lesniak RJ, Hohenwarter MD, Taylor AJ. Spectrum of causes of pancreatic calcifications. Am J Radiol 2002;178:79-86.
3. Rajasuriya K, Thenabadu PN, Leanage RU. Pancreatic calcification following prolonged malnutrition. Am J Dis Child 1970;119:149-51.
4. Iwao Y, Ritsuko S, Hiroshi O. Imaging: MRCP and ERCP. Jap J Pediatr Surg 2004;36:445-50.

Bilal Mirza

Address: Department of Paediatric Surgery,
The Children's Hospital & The Institute of Child Health
Lahore, Pakistan.

Email: blmirza@yahoo.com

Received on: 28-06-2010 Accepted on: 25-07-2010

<http://www.apspicaserep.com> © 2010 Mirza

This work is licensed under a [Creative Commons Attribution3.0UnportedLicense](https://creativecommons.org/licenses/by/3.0/)

How to cite

Mirza B. Pancreatic calcification. APSP J Case Rep 2010; 1: 11

IMAGES

OPEN ACCESS

Sirenomelia

Kanchan Kayastha

A baby was delivered to a 28-year old mother with fused limbs, no feet, absent external genitalia and imperforate anus

The newborn at arrival in emergency room of our institution was hypothermic and cyanosed. Neonate was resuscitated. Clinical examination following resuscitation revealed fused lower segment of the body below pelvis into a single limb with no feet. Posture of the lower torso was that of alphabetic letter L when viewed from the back. There were no openings for urogenital system and anal opening was also absent [Image].

The ultrasound of the patient revealed bilateral renal agenesis, and the gonads and urinary bladder were also not visualized. A sigmoid loop colostomy was formed for imperforate anus. The clinical condition of the baby deteriorated afterwards and expired at the end of first day of life.

DISCUSSION

Sirenomelia is a rare congenital anomaly characterized by partial or complete fusion of lower limbs and usually associated with other severe anomalies. It is considered, by many authors, as severe form of caudal regression syndrome. The associated anomalies may include bilateral renal agenesis, complete or partial agenesis of genitourinary system, imperforate anus, absence or ambiguous external genitalia, single umbilical artery, lung hypoplasia and vertebral and cardiac anomalies.^{1,2} In our case the associated anomalies were complete agenesis of urogenital system and imperforate anus.

Etiology of sirenomelia is uncertain and various theories have been proposed to explain its origin. An embryonic insult to caudal mesoderm between 28-32 days of gestation and vascular hypo-perfusion (arterial steal) has been proposed as possible factors. Others associated the condition with maternal diabetes mellitus, exposure to teratogens and genetic predisposition.³

Depending upon the degree of fusion of lower limbs and feet, sirenomelia can be classified into three forms

Simpus apus (no feet, one tibia, one femur)

Simpus unipus (one foot, two tibia, two fibula, two femur)

Simpus dipus (two feet, two fused legs) flipper like popularly known as mermaid.⁴

In our case the exact type of the anomaly cannot be proposed in absence of radiographic findings, however, clinically the baby can be placed in simpus apus group as the patient did not have feet.

In expert hands the antenatal sonographic diagnosis of sirenomelia can be made as early as 18 weeks of gestation and a better option of termination of pregnancy can be advised in order to avoid physical and psychological stress to parents and the family.



Patient with imperforate anus and absent external genitalia. Lower limbs fused into a single limb

REFERENCES

1. Das BB, Rajegowda BK, Bainbridge R, Giampietro FP. Caudal regression syndrome versus sirenomelia: a case report. *J Perinatol* 2002; 22:168-70.
2. Valenzano M, Paoletti R, Rossi A, Farinini D, Garlaschi G, Fulcheri E. Sirenomelia. Pathological features, antenatal ultrasonographic clues, and

review of current embryogenic theories. Human Reprduct Updat 1999; 5:82-6.

3. Jaiyesimi F, Gomathinayagam T. Dixit A, Amer M. Sirenomelia without vitelline artery steal. Annals Saudi Med 1998; 18: 542-4.
4. Bhardwaj AP. Sirenomelia. Indian Pediatr 2004; 41: 196.

Kanchan Kayastha

Address: Department of Paediatric Surgery, The Children's Hospital & The Institute of Child Health Lahore, Pakistan.

Email: Kanchan_kayastha@hotmail.com

Received on: 05-06-2010 Accepted on: 25-07-2010

<http://www.apspicaserep.com> © 2010 Kayastha

This work is licensed under a [Creative Commons Attribution3.0UnportedLicense](#)

How to cite

Kayastha K. Sirenomelia. APSP J Case Rep 2010; 1: 12