FOETAL-VITELLINE DUCT CYST WITH HINDGUT AGENESIS- A RARE CASE REPORT

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ABSTRACT

BACKGROUND

This is a case of 22-year-old female patient with 14 weeks live foetus, underwent routine ultrasound scan where a large intraabdominal cyst was found. Foetus has been aborted and later subjected to autopsy where it has been reported a small developed gut with mid gut anomaly of intestinal vetilline cyst with hindgut agenesis. Ultrasound scan was done with Philips HD 7 ultrasound machine, 3.5-5 MHz probe.

KEYWORDS

Vitelline Duct, Hindgut Agenesis, Yolk Sac, Gene Alleles, Autopsy, Omphalomesenteric Duct.

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BACKGROUND

Omphalomesenteric duct/vitelline duct serves as communication between midgut and yolk sac in early embryonic life and usually obliterates between 9-18th week of gestation. The vitelline duct is an embryonic remnant of yolk sac. If yolk stalk does not obliterate completely, various portions may persist giving rise to many entities like Meckel's diverticulum, fistulas to umbilicus including an omphalomesenteric duct cyst. The cysts are lined by columnar/cuboidal epithelium. Eventually, these cysts may contain gastric, colonic or small intestinal epithelium.

CASE REPORT

A 22-year-old female patient came for routine antenatal checkup. Patient had vague lower abdominal pain for 6 days, which got relieved on taking rest. A routine antenatal ultrasound scan was done for the patient where a live foetus corresponding to 14 weeks 3 days of gestation- measurements include-

BPD	-	26.9 mm
FL	-	13.6 mm
AC	-	123.5 mm

With foetal cardiac activity (FHR-146/m) with fundo anterior grade 1 placenta, there was a large anechoic cystic lesion arising from foetal abdomen approximately measuring 5.3 x 4.13 x 3.3 cms taking no flow on colour mapping, cyst noted to be completely indenting proximal structures of the foetal abdomen. The above-described cyst was occupying entire abdominal cavity of the foetus pushing mediastinal structures as well.

Liver appeared normal with non-visualisation of bowel loops. The cord appearing to be having two vessels, however, not imaged properly due to large abdominal cyst. Foetal limbs and spine were normal. Foetal brain was normal. Patient has been counseled about condition of the foetus, later foetus was terminated by obstetricians and sent for autopsy and histopathology.

Autopsy Findings

Gross Examination

Weight of the foetus	-	100 g.
Length of the foetus	-	15 cms.
Chest circumference	-	13 cms.

A cyst noted in abdominal cavity measuring 5.5 cm attached to ileum at one end and other end to umbilical cord. Contents of the cyst- clear serous fluid with 15-20 mL of volume. Internal surface of the cyst appears smooth. Part of the mid gut and hindgut were not developed, structures arising from these parts were not present. No development of urinary bladder and genital system from urogenital sinus noted.

Placenta-

Placenta was measuring 11 x 6 x 3 cm with gray brown clots. Cut section showing two vessels.

Microscopic Examination of Placenta-

Placenta showing plenty of chorionic villi with sheets of decidual cells and focal areas of haemorrhages. Sections of cord revealed structure of two vessel separated by fibrous tissue in one vessel. There were areas of haemorrhages.

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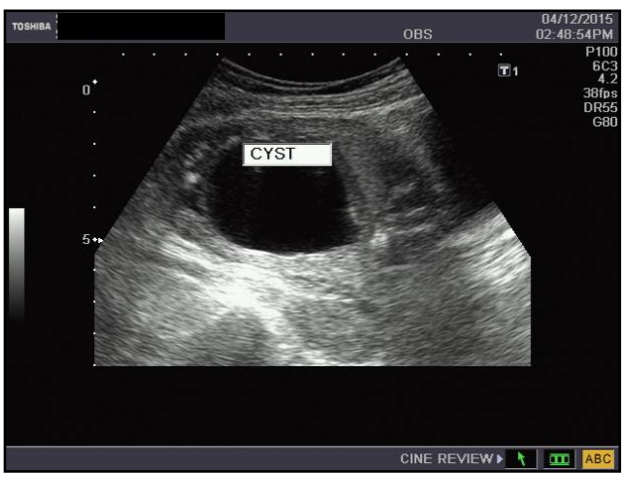
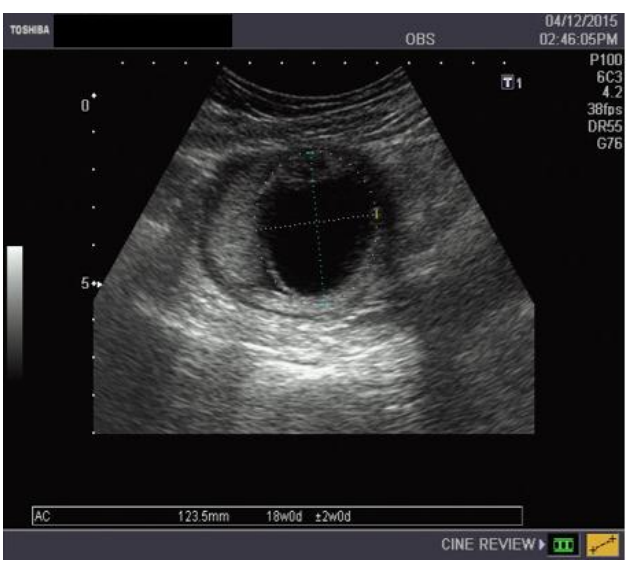
Microscopic Examination of Abdominal Cyst-

Sections from cyst showing lining of low columnar to low cuboidal epithelium noted. In few places, cyst wall is thickened and lined by single layer of cuboidal epithelium. Wall shows smooth muscle of intestinal type and occasional nerve bundles. The proximal end of cyst shows normal intestinal mucosa with infantile villous and muscular wall, distal end shows fibrous wall of umbilical cord.

Images of the Case[1,2]

- USG.
- USG was done using 3.5-5 MHz curvilinear array transducer for both gray scale and Doppler imaging.
- On USG, there is a round well-defined anechoic cystic lesion noted taking no flow on colour Doppler indenting proximal structures of the abdomen.





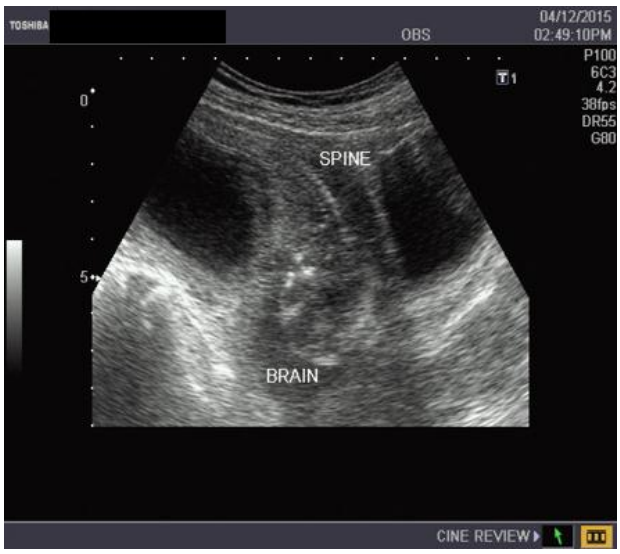
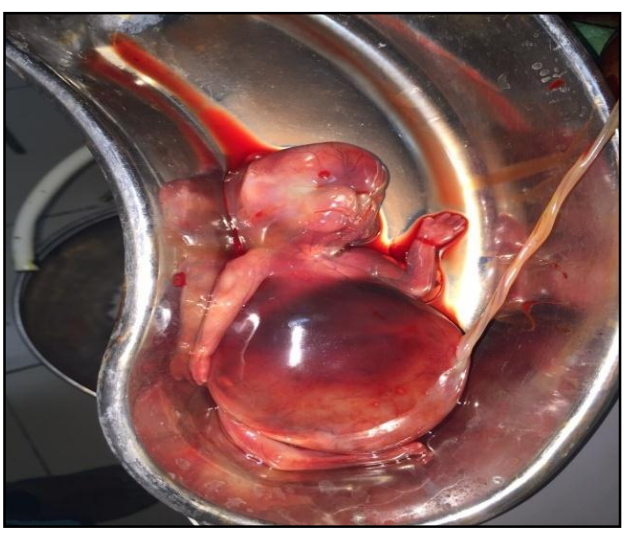
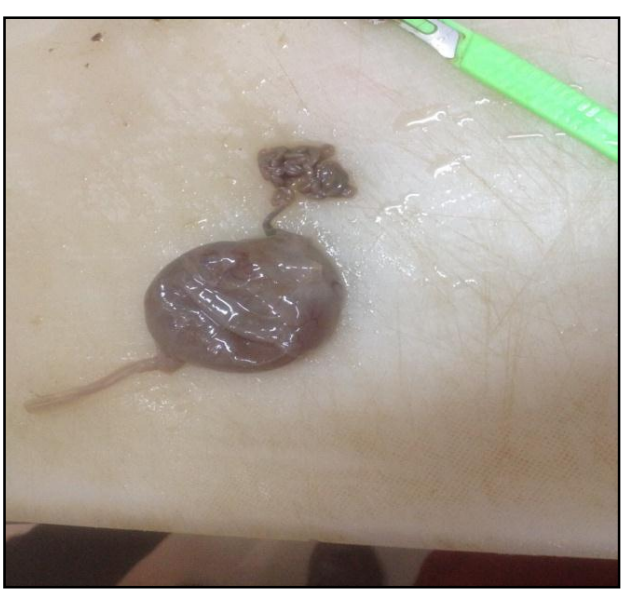


Image of Aborted foetus



Autopsy Images



DISCUSSION

Development of Hindgut

Hindgut extends from the mid gut (middle of transverse colon) to the body wall and is supplied by the inferior mesenteric artery. Hindgut consists of the developing distal transverse colon, descending colon, rectum and anal canal to pectinate line. Terminal portion of hindgut is enlarged as cloaca develops into the rectum and upper 2/3rd of anal canal, while its anterior subdivision, urogenital sinus, develops into bladder and in the female, the urethra and vestibule, while in male, the prostatic urethra.

Hindgut Agenesis[3]

Complete absence of hindgut structures is called hindgut agenesis.

There is very limited literature describing hindgut agenesis.

Laboratory research is still going on to rule out the causes and its genetic relations. One of the American Laboratories has come out with few genes, which are responsible for hindgut agenesis by conducting research on mice.

The allelic composition of genes responsible are-
.M1 X/1 tm 1/M1X/1 tm 1 = robb.
.Nr 6 a/tm 2 coo/Nr 6 a/tm Z Coo.
.Sma d L tim z rob/Snad L tim 1.Z rob.
.Tg (sox2-cre) 1 Amc/o.

Foetal intra-abdominal cyst are common lesions found during routine antenatal ultrasound scans.

These cystic lesions can arise from different entities.

Physiological intra-abdominal cysts are-

- Foetal gastric dilatation/foetal stomach bubble (can be pathological, if there is a gastric outlet obstruction).
- Normal foetal gallbladder.

Pathological intra-abdominal cysts are-

- No colour flow.
- Foetal choledochal cyst.
- Foetal ovarian cyst tends to be one of the commonest of intra-abdominal/pelvic cysts.
- Foetal hydronephrosis.
- Foetal renal cysts.
- Foetal urinoma formation secondary to posterior urethral valve.
- Enlarged foetal urinary bladder - foetal megacystis.
- Foetal enteric cyst(s).
- Foetal enteric duplication cyst.
- Foetal omental cyst.
- Foetal mesenteric duplication cyst.
- Cystic-appearing double bubble due to duodenal atresia.
- Meconium pseudocyst.
- Foetal urachal cyst.

With colour flow-

- Umbilical venous varix- particularly in intrahepatic position.
- Cystic neoplasms.
- Foetal intra-abdominal cystic teratoma- may demonstrate some colour flow.[4,5]

CONCLUSION

Correlating radiological and pathological findings of above-mentioned case, a diagnosis of mal developed gut with midgut anomaly of intestinal vetilline cyst with hindgut agenesis has been offered. To our knowledge, a similar case has not been reported so far.

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