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RESEARCH ARTICLE

SYMPTOMATIC MECKEL'S DIVERTICULUM IN THE NEWBORN: REVIEW OF THE LITERATURE

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Abstract

Meckel's diverticulum accounts for 2 to 3% of gastrointestinal tract malformations in children. It is a distal remnant of the incompletely obliterated omphalo mesenteric duct that occurs at 4 weeks' gestation, and is located approximately 40 to 60 cm from the last ileal loop. In the majority of cases, its presentation is asymptomatic and discovery is incidental. The symptomatic form represents 4% and is rare in children, including bleeding, occlusion, inflammation and intermittent umbilical oozing. The symptomatic form is exceptional in newborns. This work is a descriptive study of the literature regarding reported cases of newborns diagnosed with symptomatic Meckel's diverticulum either as a picture of intestinal obstruction or by spontaneous perforation of the DM. The objective of our study is to describe the cases of newborns operated in emergency for symptomatic Meckel's diverticulum and to evaluate the surgical management.

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Introduction:-

Meckel's diverticulum accounts for 2 to 3% of gastrointestinal tract malformations in children.

It is a distal remnant of the incompletely obliterated omphalo mesenteric duct that occurs at 4 weeks' gestation, and is located approximately 40 to 60 cm from the last ileal loop.

In the majority of cases, its presentation is asymptomatic and discovery is incidental. The symptomatic form represents 4% and is rare in children, including bleeding, occlusion, inflammation and intermittent umbilical oozing. The symptomatic form is exceptional in newborns.

This work is a descriptive study of the literature regarding reported cases of newborns diagnosed with symptomatic Meckel's diverticulum either as a picture of intestinal obstruction or by spontaneous perforation of the DM.

The objective of our study is to describe the cases of newborns operated in emergency for symptomatic Meckel's diverticulum and to evaluate the surgical management.

Patients and Methods:-

We performed a search in the "MEDLINE" database, "SCOPUS" from initial date 1988 to until 2012, with the following keywords: "newborn AND symptomatic Meckel's diverticulum" and "newborn AND complicated Meckel's diverticulum" in French and in English.

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We were thus able to group 34 articles dealing with this subject for which demographic, clinical, therapeutic and evolutionary data were analyzed; 1st degree references were also analyzed.

Results:-

We grouped 36 newborns with symptomatic Meckel's diverticulum, of which 12 were non-perforated Meckel's diverticula and 24 perforated.

A male predominance was noted: 5 boys for one girl.

The average age at diagnosis was 5.72 days (minimum H3 and maximum 24 days) with a median of 4 days.

The mean gestational age was 35.9 days, the lowest was 26 days and the highest was 41 days.

The average birth weight is 2043g; the minimum weight is 500g and the maximum weight is 4500g.

All newborns in the 1st category (n=12) (non-perforated Meckel's diverticulum) presented with associated bilious vomiting in:

- 2 newborns: medium to heavy rectal bleeding.
- 1 newborn: lethargy
- 1 newborn presented a pneumo scrotum with abdominal distension
- 1 newborn: palpable mass on the right
- 2 newborns presented with acute intestinal invagination

16 newborns in the 2nd category (n=24) (perforated Meckel's diverticulum) presented with pneumoperitoneum on standard abdominal-pelvic radiography, i.e. 66.6%:

- 1 newborn with sepsis
- 4 newborns presented with a picture of low neonatal occlusion without pneumoperitoneum, i.e. 16.6%.

- 2 neonates presented with meconium discharge through the omphalocele; 1 neonate had an antenatal diagnosis of anencephaly and

omphalocele

- Associated malformations were found in 3 cases, i.e. 13%, distributed as follows

1. Hirschsprung's disease in 1 case
2. "VACTERL" syndrome in 1 case
3. Omphalocele and anencephaly in 1 case
4. Omphalocele in 1 case

- 1 case of a SARS-COV2 positive newborn was reported

In our study; 15 premature newborns with gestational age ≤ 37 SA or 40% having as characteristic of the pregnancy:

- 1 case of oligohydramnios
- 1 case of bleeding and failure of tocolysis
- 1 case of maternal HELLP syndrome
- 1 case of maternal pyelonephritis and appendicitis
- 1 case Circular cord
- 1 case Twin pregnancy
- 1 case of prolonged laborious delivery

31 newborns benefited from a terminal resection-anastomosis taking away Meckel's diverticulum; in 5 newborns, a stoma was performed and then re-established.

The anatomopathological study revealed:

- The presence of gastric heterotopia in 5 cases (15.15%)
- The presence of pancreatic heterotopia in 2 cases (6%)
- No tissue heterotopy in 14 cases (42.42%)
- The maximum reported size of the Meckel Diverticulum is 6cm*5cm and the minimum 1.5cm*1.7cm
- The Meckel Diverticulum was found from 20cm to 60cm from the last ileal loop.

The evolution was favorable in the majority of cases:

- 1 case of release having benefited from ileostomy then recovery after 3 weeks
- 1 case of sepsis requiring a 28-day postoperative stay.

The tables below summarize the data from our results:

Table A: Cases of newborns with imperforate Meckel's diverticulum complicated by intestinal obstruction (NR= not reported; M= Male; F= Female).

	<u>Sex</u>	<u>Gestational age(SA)</u>	<u>Weig ht(g)</u>	<u>Age(D ays)</u>	<u>Clinicalpresenta tion</u>	<u>Contents of theDM</u>	<u>DM size Incm</u>	<u>Tissueheter otopia</u>	<u>Typeofob struction</u>
Leconte andal,1988[1]	M	NR	NR	8	Vomiting sbilious	NR	6*5	No	Compression extrinsic
Fromthe Hunt19 93[2]	M	33	1955	10	Vomiting sbilious	Curdledmilk	10	Gastric	Compression intraluminal
Goyal et al1993[3]	NR	NR	NR	24	Drowsinessa nd vomitings	NR	1.5*1.7	No	Iliums uture
Donnelly etal,1998[4]	NR	NR	500	NR	Vomitingbi liousness and rectalbleeding	NR	NR	NR	Intraluminalco mpression
Sy et al,2002[5]	M	36	2520	1	Straight massandileus	Blooddigested/m eco nium	6*4*3	No	Volvulus
TaekYuanda l2005[6]	M	39	3920	1	Vomiting sbilious	NR	3*3*2	No	Invagination
Sinhaetal, 2009[7]	M	36	2700	6	Biliousvo miting	NR	6*4*3	No	Intraluminal Compression
	M	41	3420	5	Biliousvomi ting Rectorrhagia massive		2.5*4* 3	Gastric	NR
Bertozzi andal2013[8]	M	38	3400	15	Vomiting sbilious	Mucus	3*3*2	Gastric	NR
KumarDas [9] et al2015	F	Eventually	2854	20	Biliousvomit ing	NR	3.4	NR	Invagination
LouatiH and Al2017[10]	M	Eventually	2350	1	Biliousvomit ing	NR	NR	NR	Invagination
Oukhouya MA et al2018[11]	M	NR	NR	6	Biliousvo miting	NR	3*2*3	NR	Iliums uture

Table B: Cases of newborns with Meckel's diverticulum complicated by perforation: (NR= not reported; M= Male; F= Female)

<u>Author</u>	<u>Sex</u>	<u>Gestational age (SA)</u>	<u>Weig ht(g)</u>	<u>Age</u>	<u>Clinicalpr esentatio n</u>	<u>Tissuehe terotopi a</u>	<u>Histology</u>	<u>Malformativeassociation</u>
Coppese t al,199 1[12]	M	32	1780	3	Pneumo-scrotum /pneumoperiton eu m	No	Inflammationandnec rosis	No

Ford1992[13]	NR	37	1900	1	Pneumoperitoneum	Pancreatic	Inflammationandnecrosis	VACTERLsyndrome
Yeh et al,1996[14]	M	NR	NR	8	Obstructionintestinal	No	Inflammationandnecrosis	No
Gandyandal, 1997[15]	M	Term	4500	4	Intestinablobstruction	Pancreatic	Inflammation	No
Kumaret al,1998[16]	M	NR	2300	5	Intestinablobstruction	No	Noinflammation	No
Zahraeet al,2003[17]	NR	Atterm	2070	3	Sepsis	No	Inflammation	No
Sy etal, 2006[18]	F	40	3200	3	Pneumoperitoneum	No	Inflammation	Hirschsprungdisease
Changetal,2006[19]	M	33	2040	1	Pneumoperitoneum	No	Muscle defect	No
Oyachiet al,2007[20]	M	Term	3060	17	Intestinablobstruction	No	Inflammation	No
Aguayoe et al,2009[21]	NR	28	810	6	Pneumoperitoneum	No	Inflammation	No
Alkanetal,2009[22]	F	38	2800	1	Pneumoperitoneum	No	Inflammation	No
Khan et al2012 [23]	F	29	650	6	Pneumoperitoneum	NR	Inflammation	No
Bertozzietal2013 [24]	M	34	2500	5	Pneumoperitoneum	No	withoutinflammation	No
Cranksontetal 2013 [25]	M	Eventually	NR	2	Pneumoperitoneum	NR	NR	No
Smolkinetal2013 [26]	M	28	1200	5	Pneumoperitoneum	NR	NR	No
	M	26	750		Pneumoperitoneum	NR	NR	No
Borji et al2014 [27]	M	29	1400	H3	Pneumoperitoneum	NR	NR	No
Alvare s Al2015 [28]	M	30	940	10	Pneumoperitoneum	Gastric	Diverticulitisandsevere peri-ulcerationdiverticular	No
Orelaruet Al2018[29]	M	32	1300	3	NR	NR	Inflammation Necrosis	No

Wang YJet al2019 [30]	M	27	1370	1	Pneumoperitone um	No	Inflammation	No
McKel vieetal 2019 [31]	F	30	1200	3	Pneumoperitone um	Gastric	Inflammation	No
TaziMa ndal 2019[32]	M	Eventually	3500	1	Ruptured Omphalo cele	Meconium	3.5	Omphalocele
Bindi .et al2 020 [33]	M	Eventually	NR	3	Pneumoperitone um	NR	NR	No SARSOC2Positive
Urri aSoto etal20 21[34]	M	Eventually	NR	1	Meconi umdisc hargefr omthe omphalocele	NR	NR	Anencephalyandomphalocele

Discussion:-

Meckel'sdiverticulum(MD)resultsfromincompleteregessionoftheyolkductotherwiseknown as the omphalo mesenteric duct; this obliteration anomaly occurs around the 5thSA[35]

TheDMmostcommonlypresentsas ablindpouchattachedtotheanti-mesentericrimcentred by a feeding artery. It varies in size and may be connected to the posteriorabdominalwallby afibrous flangeorremnant [36]

DMisacommon malformationofthegastrointestinaltractinchildren representing2%.Itis symptomatic in 60% after the age of 3 years with a male predominance; thesymptomatic presentation of DM is: hemorrhage 40%; intestinal obstruction 30% anddiverticulitis20% [37].

ThetoptwomechanismsofdigestiveobstructionoftheDMisacuteintestinalintussusception and volvulus (46% and 24% respectively); other less common mechanismsareinternal herniationand inflammationoftheDM [38]

TheoccurrenceofDMperforationis10%ofsymptomaticchildren inthe firstyearoflife[39]

There are few published cases and case series on symptomatic neonatal DM and this isnecessarilyexplainedbythe rarityofDMasanetiologysponsible forneonatalocclusion(etiolgiesof lowerneonatal occlusions,ulcerativenecroticperitonitis)

Inour study,36 cases ofnewborns operatedforsymptomaticDMwereidentified anddividedintotwocategories:

1. Category 1: the DM is symptomatic and not perforated
2. Category 2: the DM is symptomatically perforated

Our study included very low birth weight babies (Minimal: 500g) with extremes ofprematurity at26SA.Antenataldiagnosis was reportedin2cases.

Maternal complications (HELLP syndrome; cord circular; bleeding) were reported in 6preterminfants,suggestinganassociation betweenpregnancyandmaternalcomplicateddelivery and neonataldistress andDMperforationin preterminfants.

The main clinical presentation all ages combined was bilious vomiting and abdominaldistension first; 2 cases presented with small to large amounts of rectal bleeding and

1 case was admitted with sepsis. Premature babies were readmitted with occlusive syndrome and associated respiratory distress.

The initial management took place in a neonatal intensive care unit with good conditioning. Borg et al., was the first to perform a laparotomy in the preterm infant and found that respiratory distress is more of a direct consequence of abdominal distension by perforation of the DM [27].

Ford and Woolley [10], suggested that excessive ventilation in babies with esophageal fistulas is likely to result in DM perforation; this is increased by the coexistence of anal imperforation [13].

The radiological workup revealed pneumoperitoneum in 16 newborns and variable numbers of hydroaerosal levels in the colon and the small intestine in the other cases.

Surgical exploration objectified:

- DM located 20-60 cm from the last ileal loop
- 3 cases of acute intestinal invagination on a non-perforated DM
- 1 case of volvulus on DM with fibrous flanges no perforated.
- 24 perforated DMs
- Almost the majority of newborns have had terminal resection-anastomosis with the DM removed.
- Tissue heterotopia is present in 7 cases
- Inflammation, necrosis and hemorrhage are almost present in the histological study

The postoperative evolution was favorable in the majority of cases with an average stay of 7 days, which suggests that the one-stage treatment by terminal anastomosis resection even in premature and very low birthweight babies is effective.

Conclusion:-

Symptomatic Meckel's Diverticulum is a rare and heterogeneous entity in the newborn. It can take on several clinical presentations but the most common presentation is neonatal obstruction associated with respiratory distress in premature and low birthweight babies.

The association with maternal stress during pregnancy and delivery has been described and remains an avenue to be explored as to its link with DM perforation.

Apart from its rare symptomatic character, the diagnosis of asymptomatic DM should always be suspected to ensure early and appropriate surgical treatment.

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