Case Report

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Littre hernia in childhood: A case report with a brief review of the literature

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ABSTRACT

A 3-year-old boy with a right-sided and painful inguinal swelling for the last 2 h was admitted to the emergency department. As there were no apparent peritoneal irritation findings, right-sided incarcerated inguinal hernia was reduced and the patient was scheduled for an elective herniorrhaphy. Perioperatively even though the sac seemed empty, it was opened in order to inspect its content. The adherence of Meckel's diverticulum (MD) to the base of hernial sac was realized and Littre hernia (LH) was diagnosed. The hernia was highly ligated after the wedge resection of the diverticulum and anastomosis. The patient was fed on the second postoperative day and discharged on the third postoperative day. Despite numerous presentations of LH in the adult age group in the literature, there are limited data about the disease in the childhood period. Even though the scarcity of the data, there are some so-called rules for LH in childhood like protruding more common through umbilical hernias, containing heterotopic tissues more frequent than adult age and incarcerating/strangulating more often. The aim of this study is to review the reported LH cases, present a new case and discuss the features of LH in childhood period.

Key words: Children, littre hernia, meckel's diverticulum in hernial sac

INTRODUCTION

Littre hernia (LH) is the protrusion of a Meckel diverticulum (MD) through a potential abdominal opening with a container sac. This clinical entity has been described after the autopsy findings of two patients by Alexis de Littre for the first time as 'ileal diverticula in the inguinal hernia' in 1700. But the concept of "Littre hernia" has only been improved after a century, as J Frederich Meckel defined the true congenital diverticulum of the gut which is now entitled with his name.[1]

MD commonly complicates before the age of 2 and because of this reason, it is accepted as a childhood disease. In 75% of the patients, as the tip of MD is free in the abdomen it is actually more prone to protrude through any abdominal opening.[1] But this presentation is quite rare in childhood and the true incidence of LH is unknown. Despite numerous presentations of LH in the adult age group in the literature, there are limited data about the disease in the childhood period. Albeit the lack of informative data, there are some so-called rules for LH in childhood like protruding more common through umbilical hernias, containing heterotopic tissues more frequent than adult age and incarcerating/strangulating more often.[1]

The review of English literature of LH in the childhood period from the beginning of the 20th century revealed 69 cases in case reports [Table 1] and case series [Table 2].[2-34] The aim of this study is to review these cases, present a new case and discuss the features of LH in the childhood period.

CASE REPORT

A 3-year-old boy with a right-sided and painful inguinal swelling for the last 2 h was admitted to the emergency department. He was neither complaining of nausea/vomiting nor did have either episode of gastrointestinal bleeding or intestinal obstruction. The physical examination revealed a right-sided incarcerated inguinal hernia with no apparent peritoneal irritation findings. After the manual reduction, the patient was scheduled for an elective open herniorrhaphy. Perioperatively the widened hernial sac was dissected through the deep inguinal ring. As the sac was opened in order to inspect its

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Table 1: Littre hernia cases

	Age	Sex	Symptoms and signs	Manual reduction	Additional perioperative findings	Heterotopic mucosa
Bunts[2]	NB	F	UH		MD adherent to the inguinal ring	
	7 years	NM	Incarcerated UH	Partially completed		
	NB	NM	Incarcerated UH			
	NM	NM	UH			
	1 days	F	Incarcerated UH			
	Beyond NB	NM	UH		Perforation of MD	
	NM	M	Incarcerated IH			
Bird[3]	NB	M	Faecal fistula from UH		Everted MD in the umbilical opening	
Rendle-Short and Havard[4]	14 days	M	Incarcerated R-sided IH			
Davis[5]	3,5 weeks	M	Incarcerated R-sided IH			
Kline[6]	4 weeks	M	Incarcerated R-sided IH			
Baillie[7]	7 weeks	M	Incarcerated R-sided IH	Partially completed		
Wollgast and Hilz[8]	2,5 months	M	Incarcerated R-sided IH			
Gracey and Williams[9]	18 months	M	Incarcerated R-sided IH		MD and appendix in the hernial sac	
Rutherford and Akers[10]	Beyond NB	NM	Incarcerated IH			
	Beyond NB	NM	Strangulated IH			
Kabbani and Lewis[11]	2 months	NM	R-sided IH			+*
Kremer and Morton[12]	14 months	M	Incarcerated UH			
Krausz *et al*.[13]	6 weeks	M	Bilateral incarcerated IH	Completed in the R side Failed in the L side	Gangrenous L-sided testicle	
Mishalany *et al*.[14]	10 months	M	Incarcerated R-sided IH	Partially completed	Tip of the MD adherent to testicle	+*
Ravikumar *et al*.[15]	8 months	M	R-sided IH		Tip of the MD adherent to the hernial sac	+*,**,***
Das *et al*.[16]	NM	NM	NM		MD adherent to hernial sac fundus	
	NM	NM	NM		MD adherent to hernial sac fundus	
Kang *et al*.[17]	2 days	M	L-sided DH			
Messina *et al*.[18]	16 days	M	Incarcerated R-sided IH	Partially completed	Tip of the MD adherent to root of scrotum	
	12 days	M	Incarcerated R-sided IH			
Chirdan *et al*.[19]	8 years	M	Incarcerated UH	Failed		
Vaos[20]	2 years	M	Incarcerated R-sided IH		Perforation of MD	+*
Ein *et al*.[21]	NM	NM	IH			
	NM	NM	IH			
Chan *et al*.[22]	11 months	M	L-sided IH			
	3 years	M	Incarcerated R-sided IH	Failed		
Al-Mandil *et al*.[23]	Beyond NB	M	Incarcerated IH			
Akin *et al*.[24]	7 years	M	Incarcerated R-sided IH			+*
Komlatse *et al*.[25]	18 months	F	Incarcerated UH			
Presented case	3 years	M	Incarcerated R-sided IH		MD adherent to base of the hernial sac	+****

NB = Newborn, M = Male, F = Female, NM = Not mentioned in the article, IH = Inguinal hernia, UH = Umbilical hernia, DH = Diaphragmatic hernia, R-sided = Right sided, L-sided = Left sided, MD = Meckel's diverticulum, *gastric mucosa; **jejunal mucosa; ***colonic mucosa; ****pancreatic mucosa

content, the MD was seen. It was found adherent to the base of the hernial sac circumferentially [Figure 1]. The diverticulum was dissected from the hernial sac and high ligation of the hernia sac was completed after the wedge resection of the diverticulum and anastomosis. The intestinal contents were addressed to the intraperitoneal cavity. The patient was fed on the second postoperative day and discharged on the third postoperative day. The histopathological findings revealed MD with an ectopic pancreatic mucosa.

DISCUSSION

LH is the protrusion of a MD through any potential abdominal opening. It is a rare perioperative finding in adults with an incidence of 4-10% and even rarer in the childhood period.[1]

This literature review containing the presented case divulged 70 cases of LH from early 1900s to these days and by the help of these cases we aimed to discuss the features of LH in childhood. LH seems to be a problem

Table 2: Littre hernia cases presented with congenital abdominal wall defects

	MD/umbilical cord hernia	MD/exomphalos minor	MD/exomphalos major
Williams[26]	1/1		
Aitken[27]	7/23	1/9	
Nicol and MacKinlay[28]		7/25	1/24
Wakhlu and Wakhlu[29]		8/45	0/17
Hale and Agrawal[30]		1/1 *	
Tourne *et al*.[31]		1/1	
Kumar *et al*.[32]		4/27	0/26
Mavridis *et al*.[33]		1/1*	
Pal *et al*.[34]	2/4		

*Presented with faecal fistula, MD = Meckel diverticulum

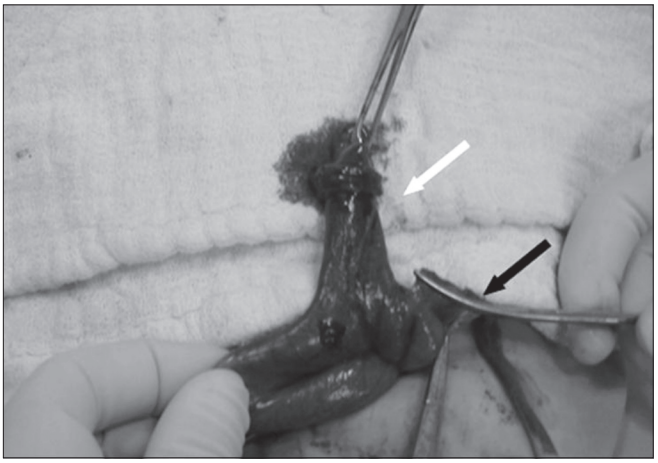


Figure 1: The Meckel's diverticulum and adjacent ileum in the inguinal sac (the white and the black arrows indicate the Meckel's diverticulum and the medial wall of the hernial sac, respectively)

of infancy generally in male gender. Fifty one of these patients presented with LH in the infancy period and the vast majority of these case reports and case series are composed of male gender. Even though LH can be seen at any age, the infancy period seems to be the most risky period. It is probably attributed to the tendency of incarceration at this age group.

Even though the half of the LH is located in the inguinal region in the adult age, the most common site is accepted as 'umbilical hernia' for children with a reported incidence of 85%.[1] This literature review revealed that not solely umbilical hernias but any kind of persisting umbilical openings (including umbilical hernias, umbilical cord hernias, exomphalos minor and major) are the most common sites for LH. This common location is due to the foetal umbilicus being a component of omphalomesenteric duct.[1] Apart from the previous reports, the prevalence of this presentation is found lower in this series like 64.7%. (44/68; as total number of the cases with known location of LH is 68). The second common site is the inguinal region with a prevalence of 33.8% in all age groups (23/68). Diverse from adulthood, the protrusion from the femoral region seems negligible in childhood. That is probably due to the rarity of femoral hernias in the childhood period. Most interesting site for protrusion of MD in this series is through a diaphragmatic hernia in a baby.[17]

Despite the difficulty of preoperative diagnosis of a LH, findings like incomplete manual reduction of an incarcerated hernia, hernial faecal fistulas and previous history of rectal bleeding were accepted to alert the clinician about a LH.[14] Even though the manual reduction was the procedure of choice in most of the patients in this series, it was found to be not completed in four patients and failed in three. Only three patients of this series presented faecal fistulas. The history of rectal bleeding or repetitive abdominal pain was not present in any of the cases in this review.

The complication rate of LH in this series is about 38.5%. (27/70). Twenty four of these patients incarcerated and one of the incarcerated patient experienced intra-abdominal perforation. Three of the patients in the complication group presented with faecal fistula, one patient through an umbilical hernia and two babies through exomphalos sacs. No other sides for faecal fistulas were reported even in cases who admitted hospital lately. In terms of fistula formation, umbilical region seems to be at risk for when compared to the other abdominal openings. It is not only due to the absence of muscular layers but also due to the lesser amount of subcutaneous tissue at the umbilicus in children.

The presence of heterotopic mucosa of LH is a rare finding in this series and 66.7% of the cases with heterotopic mucosa (4/6) were found to be complicated either by adhering to the adjacent tissues or by perforating. Six cases presented with adhesions to the adjacent tissues and pathological findings of half of these patients revealed heterotopic mucosa. The formation of adhesions is said to be due to either inflammatory processes related to the heterotopic mucosa or congestion and exudation due to closed loop obstruction in heterotrophy free MDs.[7,14]

In conclusion, the anamnesis of a LH is no longer different from any hernia whether complicated or not. While evaluating a newborn with a congenital abdominal wall defect, a MD through any umbilical opening should be kept in mind especially for the ones

with small defects (exomphalos minor and umbilical cord hernia). Quick decision and early intervention in such cases could preclude perforation and related faecal fistula formation. Meanwhile while evaluating a child beyond newborn period with an incarcerated hernia, an incomplete or failed manual reduction should bring forth the possibility of a LH. Furthermore, querying the history of rectal bleeding or repetitive abdominal pain in such cases should increase the suspicion of a LH.

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