In The Name of God

Iranian Journal of Pediatric Surgery

Official Journal of Iranian Society of Pediatric Surgeons

With the Collaboration of:
Pediatric Surgery Research Center, Research Institute for Children's Health, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

The Iranian Journal of Pediatric Surgery is published biannually by the Iranian Society of Pediatric Surgeons and Pediatric Surgery Research Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

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6. Skandalakis JE, Gray SW, Ricketts R: The esophagus, in Skandalakis JE, Gray SW (eds): Embryology for Surgeons. Baltimore, MD, Williams & Wilkins, 1994, pp 65-112

Paper presented at a meeting:
7. Bealer JF, Vanderwall K, Adzick NS, et al: A new treatment option for patients with congenital diaphragmatic hernia. Presented at the 14th annual meeting of the International Fetal Medicine and Surgery Society, Newport, RI, May 3-6, 1996.

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Original Articles

Case Reports

From the editor

Since the first appearance of COVID-19, especially during the first pandemic wave, emergency room visits decreased; although emergency surgeries remained the same. The main concern at the time was for elective surgeries, which according to different studies were postponed at pandemic waves resulting in increased surgery waiting times. This increase in "waiting time" was detrimental for some fields of surgery such as oncologic surgeries and the decision to postpone, to call a surgery elective or emergent was made by the surgeon; especially those surgeons who were part of the authorities. This is why in the first chapter of Schwartz text book of surgery a surgeon is not defined only as someone who has knowledge and can perform a surgical technique but is a person with the ability to make important decisions and be a leader. The past 2 years have been very demanding for the medical community and we have made it work with as much grace and dignity as possible. So this is a shout-out to all of us out there who have done the best they can and are still fighting to improve the outcome.

Editor- In- Chief

Ahmad Khaleghnejad Tabari MD

Professor of Pediatric Surgery

Outcomes of Hepaticoduodenostomy over T-tube against Roux-en-Y Hepaticojejunostomy to Restore Bilio-enteric Continuity after Choledochal Cyst Excision in Children

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How to cite this article:
Thakur VK, Yadav R, Chaubey D, Keshri R, Hasan Z, Kumar V, Prasad R, Kumar R, Rahul SK. Outcomes of Hepaticoduodenostomy over T-tube against Roux-en-Y Hepaticojejunostomy to Restore Bilio-enteric Continuity after Choledochal Cyst Excision in Children. Iranian Journal of Pediatric Surgery 2021; 7 (2):68 - 80.

DOI: https://doi.org/10.22037/irjps.v7i2.34571

Abstract

Introduction: Any surgical procedure which would restore the bilio-enteric continuity after excision of the choledochal cyst with minimal complications would be a feasible alternative to Hepaticojejunostomy using a Roux loop of jejunum. To determine the outcomes of Hepaticoduodenostomy done over T-Tube against Roux-en-Y Hepaticojejunostomy for bilio-enteric reconstruction after excision of choledochal cyst.

Materials and Methods: This study was retrospectively done on all patients of choledochal cysts (Types 1 and 4) operated between January, 2014 and December, 2019. The clinical details, intra-operative and post-operative results of patients who underwent Roux-en-Y Hepatico-Jejunostomy (Group-1) and Hepatico-duodenostomy over T-Tube (Group -2) for establishing bilio-enteric continuity after excision of choledochal cyst were compared and analyzed statistically.

received: 15 April 2021
accepted: 9 May 2021
Published online: November 2021

Results: 78 patients of choledochal cysts were operated during this period with 31 patients in Group-1 and 47 in Group-2; there was no difference in the mean age or size of the cysts in the two groups; Type 1 cysts were the most common with female preponderance in both groups.

Group-2 patients had lesser intra-operative time and fewer numbers of sutures were used during surgery. There was no difference in the incidence of anastomotic leaks, strictures, cholangitis or adhesive obstruction and reoperation rates between the two groups. Group 2 showed increased nasogastric bilious aspirates in 19.15% of cases which improved on conservative management.

Conclusion: Bilio-enteric reconstruction using Hepaticoduodenostomy over T-Tube is a simpler, lower pressure and less time taking anastomotic technique with comparable complication rates when compared to Roux-en-Y Hepaticojejunostomy in the management of choledochal cysts.

Keywords

- Choledochal cyst
- Hepaticoduodenostomy
- T-Tub

Introduction

Surgical management of Choledochal cyst (CDC) is complex and determined by the type of CDC. The most common types of CDC – type 1 and 4 are managed by excision of CDC and re-establishing bilio-enteric continuity by anastomosing the healthy biliary channel to a bowel loop. This can be variously done by using either a Roux loop of Jejunum or the duodenum (open or laparoscopic methods). Several investigators have compared the two approaches and most of them have favored Hepatico- jejunostmy using Roux loop of jejunum (HJ).[1-3] At our center, two different units practice two different procedures; one of the two units does open HJ, while the other does open hepaticoduodenostomy (HD)to restore bilio-enteric continuity after CDC excision. In the unit performing HD, two patients had anastomotic leak which was very difficult to manage and increased the morbidity and hospital stay of those patients. So, this unit now practices a modified HD technique where a low-pressure anastomosis is fashioned over a T-tube ensuring its further protection. We present a comparative study between all our patients who had this modified HD over T- tube and those who had a Roux-en-Y HJ.

Materials and Methods

This was a retrospective study that was conducted on all patients of type 1 and type 4 choledochal cysts operated in the

department of Pediatric Surgery at a tertiary care center from January, 2014 to December, 2019 after taking clearance from the institutional ethics committee. All other types of choledochal cysts (Types 2, 3 and 5) and cases which presented with perforation of the choledochal cysts were excluded from the study.

At our center, patients of CDC are randomly allocated to one of the two units for surgical management. One unit performs open HJ after CDC excision and all these patients constituted Group 1. The other unit does open HD over a T-Tube after CDC excision and all patients operated by this technique constituted Group 2. While allocation of patients, a simple rule was followed that patients presenting to a particular unit's outpatients' care would be operated by the same unit. This also was the reason for the different number of patients in the two groups.

To calculate the sample size, search was done for similar comparative study in literature. In one such study, Liem et al., reported operative time of 220 ±60minutes and164±51 minutes for HJ and HD groups, respectively.[3]Considering an alpha error of 0.05 and power of 80% and using the values of mean, standard deviation and percentage of decrease in operative time (25.45%) from this data, we got a sample size of 36 (18 for each group). Considering a 10% loss to follow-up, we calculated a sample size of 20 for each group. So, we planned to compare at least 20 patients in each group.

Operative records of patients were recovered from the monthly operative census and hospital medical records. A total of 78 patients were included in the study. The patient characteristics (including age at the time of surgery, sex, type of choledochal cyst on Magnetic Resonance Cholangiopancreatogram (MRCP), size of choledochal cyst); intraoperative parameters (like size of the cyst during surgery, operative time, blood loss, any significant intraoperative complication; number of sutures used); Postoperative findings (including number of days for drain removal, number of days for start of oral feeding, wound infection, anastomotic leak, number of days of hospital stay, postoperative adhesion, cholangitis, any anastomotic stricture in long term follow-up, cosmetic aspects, evidence of malabsorptive symptoms, diarrhea, malnutrition) and overall cost of surgery were compared between the two groups. Patients were followed up in the outpatient's department (three-monthly in the first year, six-monthly for next two years and yearly thereafter for five years). In case of any symptoms, they were advised to follow-up any time. Abdominal ultrasound at six months and liver function tests (LFT) at three months were routinely done in all patients; any symptomatic patients (fever, abdominal pain, altered LFT, raised total leukocyte counts) were subjected to Hepatobiliary Iminodiacetic Acid (HIDA) scan to confirm anastomotic patency.

Data thus collected was analyzed.

Protocol for management of Choledochal cyst at our Institute

All patients who presented with a diagnosis of CDC on abdominal sonogram for the evaluation of symptoms like abdominal pain, jaundice, fever or lump abdomen were further evaluated by MRCP to elucidate the CDC type and guide further surgical management. Simultaneously, all routine laboratory tests like complete blood counts, LFT andrenal function tests were done. After pre-anesthetic evaluation and proper counseling, patients were taken up for surgery which included excision of CDC and restoration of bilio-enteric continuity by either a Roux-en-Y HJ or HD over a No. 10 Fr T-Tube, depending on the concerned unit in which they were admitted. While performing HD over T-Tube, complete Kocherization of the duodenum was done after cyst excision. Site of anastomosis was chosen distal to the normal location of ampulla in the second part of duodenum **Figure1.**

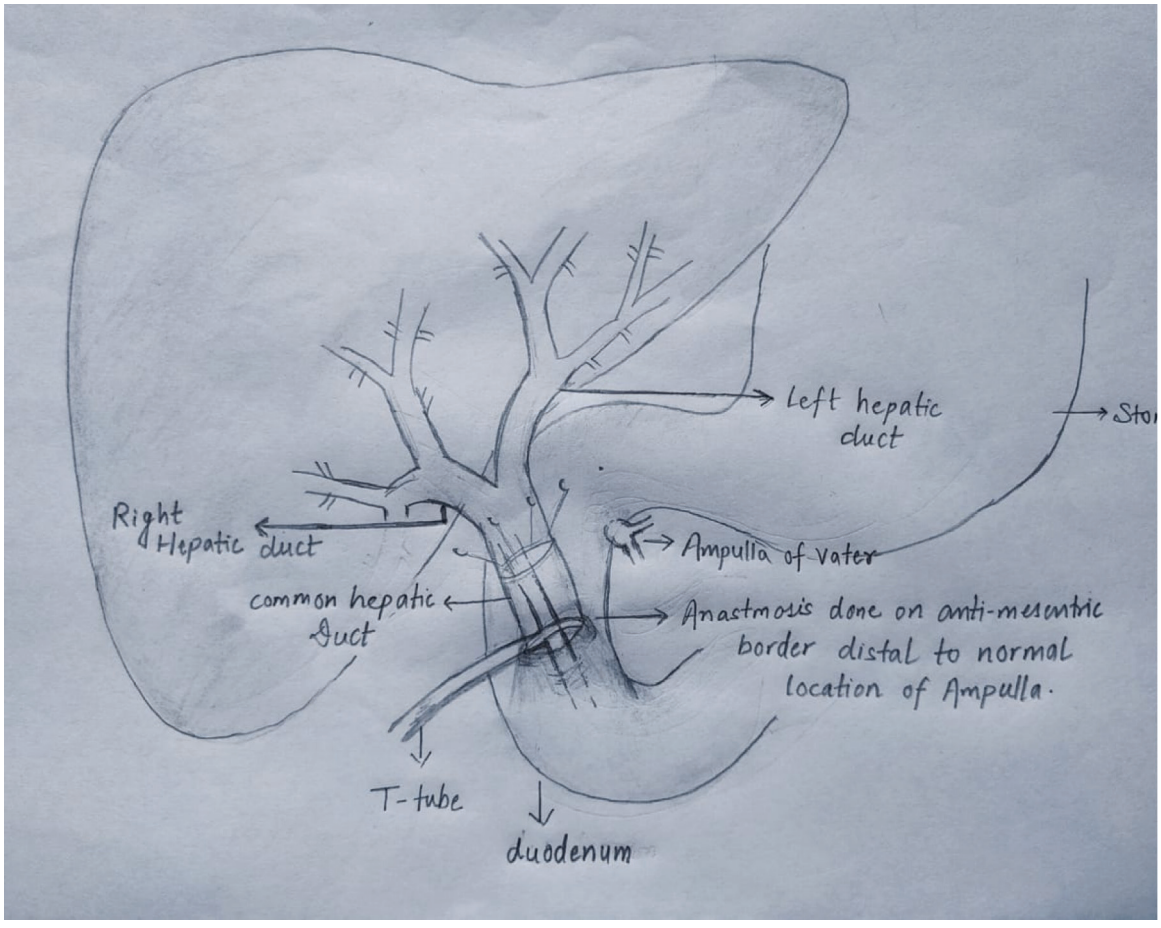

Figure 1: Sketch showing site and method used to fashion HD over T-Tube after excision of CDC

While dissection of the distal portion of the CDC, a small caliber cannula or feeding tube was inserted to flush the distal end and also to have an idea of the location of the ampulla in the duodenum. After taking the posterior sutures, T-tube was inserted with one limb in biliary channel and the other in the duodenum. Then, anterior layer was completed keeping T-Tube in dependent position **Figure 2**. Kocherization of the duodenum made this anastomosis easy and tension-free without any kink. A subhepatic drain and wide caliber nasogastric tube was introduced before closure of wound.

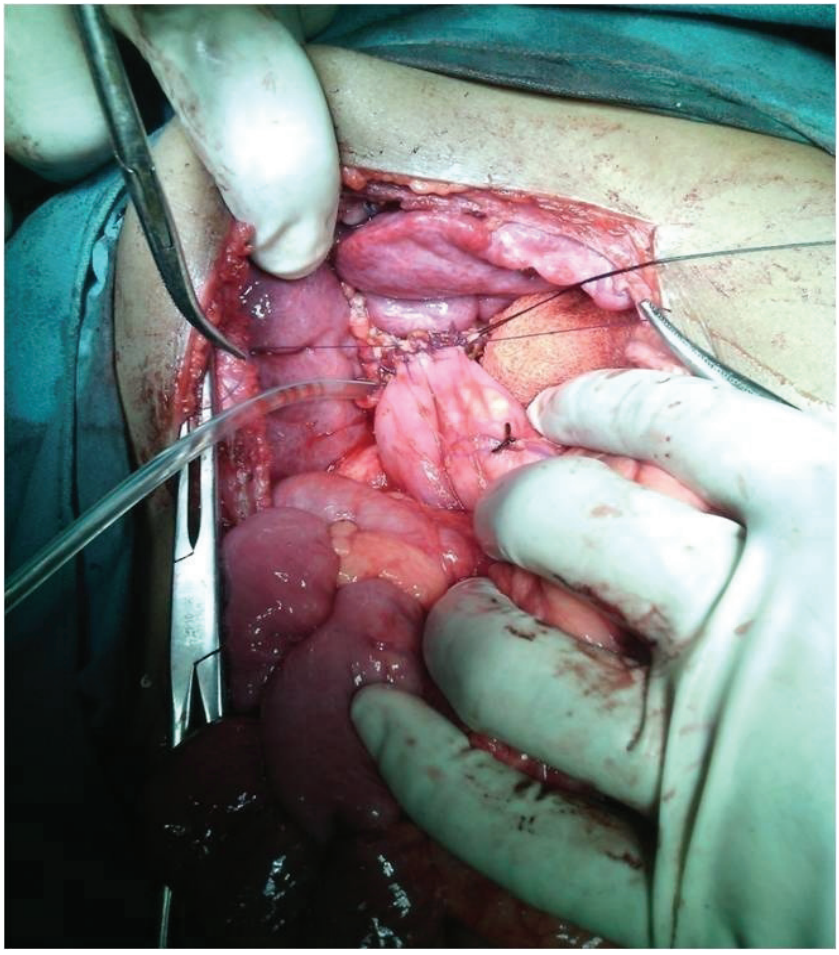

Figure 2: Intra-operative picture of completed HD over a T-Tube after excision of CDC

In the peri-operative period, patients were transfused blood products as per need and managed on intravenous (I.V.) fluids, antibiotics and analgesics. Patients were allowed oral intake when nasogastric tube output lessened and bowel movements normalized.

Patients with HD over T-tube needed clamping of the T-tube once bowel movements were normal and they

tolerated oral feeds. These patients were discharged with a clamped T-Tube coiled neatly underneath a strap dressing and on oral medications. T- Tube was removed after three weeks of surgery at the time of first follow-up. Thereafter, follow-up of these patients was done at three months interval during first year and six monthly thereafter for 2 years and yearly after that. Liver function tests were done after three months and hepato-biliary sonogram after six months of surgery. HIDA scans were done in selected patients who either had some abnormality in sonogram or liver function tests or had suspicion of cholangitis.

Summary statistics were presented in the table as mean quantitative variable and as n% for categorical outcome. Check for homogeneity of sample population and normality assumption was performed. Chi square test was used to compare proportions and t-test to compare the difference in averages. Statistical significance was based on p-value≤ 0.05.

Results

A total of 78 patients of CDC were operated during this period. Among these 31 belonged to Group 1 and had Roux-en-Y HJ after cyst excision; 47 belonged to Group 2 and had HD over T-Tube. **Table 1** gives the details of the patients presenting in these two groups.

Table 1: Demographic characteristics and clinical presentation of 78 patients who underwent excision of choledochal cyst and reconstruction.

Parameter	Group 1 (Roux-en-Y HJ) 31	Group 2 (HD over T-Tube) 47	P value	Mean Difference ±SE (95% CI)
No. of patients (78)	31	47		
Age (months)	33	38		
Gender **Male** **Female**	 12 19	 16 31	0.6741	Not applicable
Type of Cyst **1** **4**	 27 4	 43 4	.3916	Not applicable
Size of cyst in mm (Mean)	62.71 (range 36-124)	63.95 (range -32-122)	0.8013	-1.25 ± 4.95 (-11.11-8.61)
Operative time in min (Mean)	161.65 (range 129-190)	113.21 (range 96 -131)	P<.0001*	48.44± 3.130 (45.2056-54.675)
No. of sutures used for anastomosis(Mean)	Vicryl sutures - 5(range 4-6)	Vicryl -3(range 3-4)	<.0001*	1.66±0.147 (1.36-1.95)
Hospital Stay in days	9.26	11.87	<0.0001*	-2.61±0.349

(Mean)				(-3.306 to -1.91)
No. Of Post op. Days when i.v. fluids were given (mean)	7.41	7.49	0.7313*	-0.07±0.203 (-0.474to 0.33)

***t -Test significant at 5% level**

Patients in the two groups were comparable in age at the time of surgery and females outnumbered males in both these groups. Type 1 variety of CDC was the commonest CDC type operated upon during the period of the study. However, there was a great variability in the range of the age of the patients at presentation. Some of them presented as infants, while some presented as late as 12years. Youngest patient in Group 2 was 3.5months old; anastomosis (HD) over 10 Fr T-Tube could be easily done because we cut a linear segment of the T-Tube opposite to the long limb to ensure its easy insertion. Mean follow-up time of patients was 31months in our study.

Table 2 compares the intra-operative and post-operative results of 78 patients who underwent excision of CDC and reconstruction

Table 2: Intra-operative and Post-operative complications

Parameter	Group 1 (Roux-en-Y HJ) 31	Group 2 (HD over T-Tube) 47
Mortality	0	0
Morbidity		
Intra-operative major complication	Nil	Nil
Anastomotic leak	Nil	Nil
Post operative bilious nasogastric output	Nil	Significant in 9 (19.15%)
Post operative adhesion/obstruction	1	Nil
cholangitis	2(6.4%)	1 (2.1%)
Anastomotic stricture	Nil	Nil
Malnutrition (Anthropometric measurement & Serum Protein)	6 (All improved at one year after surgery)	8 (All improved at one year after surgery)

There was no significant difference in the postoperative complications like anastomotic leak, anastomotic stricture, adhesive obstruction or cholangitis. A significant incidence of increased nasogastric bilious output was seen in 9/47 (19.15%) of patients in HD over T-Tube group. These aspirates settled conservatively and on follow-up none of these nine patients have experienced any other symptoms suggestive of reflux gastritis. In 41/47 of these, anastomosis was distal to location of ampulla in distal second part of duodenum.

No T-Tube specific complication was observed in the HD over T-Tube group. T-Tubes could be easily pulled out after three weeks of surgery in all patients.

HIDA scan was not done in all patients due to cost constraints and was done only in selected patients (total seven in number), who had either suspicion of cholangitis (two in HJ group and one in HD group); altered LFT (raised alkaline phosphatase in three HD patients and in one HJ patient). In all seven patients, patency was confirmed on HIDA scan. On Ultrasound – abdomen, only two of these seven patients showed dilatation of intrahepatic biliary radicles with evidence of sludge in one patient and pneumobilia in another patient.

14/78 (17.95%) patients were malnourished at presentation (six in Group 1 and eight in Group 2) represented by weight below two standard deviations on WHO growth chart; all of these patients gained weight at one year follow-up after surgery.

Discussion

The choice between HJ and HD to restore bilio-enteric continuity after excision of CDC has been a matter of debate since a very long time.[1, 2]Several investigators have argued against HD claiming it to be associated with increased incidence of cholangitis and biliary gastritis and that such complication with traditional HJ is rare.[3, 4] On the other hand, HD appears to be more physiologic as the site of bilio-enteric anastomosis is the duodenum itself; unlike HJ, where a significant length of jejunum is lost in fashioning a Roux loop of adequate length to lessen the chances of cholangitis. Sufficient evidence now exists against the unnecessary anxiety about risks of cholangitis in HD patients.[1, 5 &6]Lesser operative time, fewer sutures and accessibility of the anastomotic site to endoscopy are a few other straightforward advantages of HD. [7]The performance of HD saw a significant rise after the advent of laparoscopic surgery for choledochal cysts. [7, 8] This was because of the obvious reasons of ease of performance in lesser time and with fewer expenses towards the fewer number of sutures needed and fewer chances of postoperative adhesions.[1, 8] However, two very important risks particularly possible in the cases of HD needed to be addressed to. First one is the remote but possible risk of anastomotic leak in the cases of HD, and the second is the matter of biliary reflux and associated gastritis in these cases. Our modification of technique of doing HD over a T-tube

makes the anastomotic site a low-pressure zone, reduces edema at the anastomotic site, and promotes healing thereby lessening further the chances of leak.[9, 10] This study is a comparison between HJ and HD over a T-tube done to achieve bilioenteric continuity after excision of CDC.

T-Tubes have been used for a long time in adult cases of surgery for stone in the common bile duct (CBD), and in those limited injuries of the common bile duct, where closure of the defect in CBD is done over a T-Tube. [11]While the clinical use of former has lessened after the advent of Endoscopic Retrograde Cholangiopancreatography (ERCP) with stenting and primary closure of the CBD after laparoscopic CBD exploration, the use of T-Tube in latter is still indicated as it has been a simple and effective option.[11, 12] In a study by Xiao et al., stented closure of CBD after laparoscopic CBD exploration was better than primary closure or closure over a T-Tube, when comparing the operative time, biliary complications and return of liver function tests to normal. [13]But, introducing a stent would subject the patient to another endoscopic procedure in the future. Although T-Tube has to be kept longer and as per our protocol is removed after 3 weeks of surgery, it has no other disadvantages and can be removed without any invasive procedure. Patients go home on full oral diet in the meantime. These advantages of T-tube in making the anastomotic region a low-pressure system by forming a controlled fistula ensures a complication–free healing process and can be used in HD after CDC excision depending on this principle.

In a study by Sharma et al., correlation between Cyst size, intra-cystic pressure, backpressure changes on hepatic histology, levels of amylase and lipase in intra-cystic fluid, and cyst wall histology was reported.[14] High pressure cysts tended to be smaller in size with comparatively more significant liver parenchymal changes on histology. By contrast, cysts with low intracystic pressures were comparatively larger and had higher levels of amylase and lipase to cause more damage to the cyst wall on histology.[14] Keeping this important observation in consideration, we emphasize on the importance of keeping low intra-cystic pressure in not only lessening the pressure on the anastomosis in HD but also in maintaining the physiological flow of bile from liver towards the bowel, thereby lessening the backpressure changes on the liver parenchyma. A simple T-Tube insertion at the anastomotic site ensures such pressure characteristics in the lumen and in the liver.

In this study, out of a total of 78 patients, 31 underwent HJ while 47 had HD over a T-Tube. This difference is only due to the fact that more patients presented to the outpatient's department of the unit practicing HD over a T-tube. Similar trends in the choice of surgery have been observed in a meta-analysis of pediatric patients of CDC who have undergone either HJ or HD; the number of patients with HD outnumbered those who had HJ.[1]Patients belonging to the two groups

were of comparable age. In both the groups, Type 1 was the commonest type of CDC followed by type 4 which were only a few in numbers. The mean size of the lesion in both the groups did not show any statistical difference and were therefore comparable. Size and anatomy of CDC has an important role in determining the duration of dissection (surgery). On comparing the overall time required to complete the surgery, HD took shorter time than HJ to be completed and this difference was found to be statistically significant. This finding can be explained by the extra time needed for jejuno-jejunostomy during Roux loop construction. This also is the reason for extra sutures and increased cost of surgery in HJ. These findings are in line with the observation of several other investigators.[2, 3 & 7]

An obvious advantage of HJ was reflected by the earlier discharge of these patients compared to the HD group. But we found that this was more due to the reluctance on the part of the surgeons in starting oral feeds early due to the presence of T-Tube in duodenum. We now routinely start oral feeds on 7th postoperative day in case of HD and discharge the patient as soon as they accept full oral diet after clamping T-Tube without any complications. None of our patients have had any problems after clamping the T-Tube and they go home with a clamped T-Tube neatly dressed and covered over the abdominal wall. There is no restriction in patient's mobility and none of the patients have pulled it accidentally. T-Tube is removed after 3 weeks of surgery as a day care procedure when the patient turns up in the outpatient clinic.

No anastomotic leak or any significant intra-operative complication was noted in either of the groups. In long term follow-up, two patients in the HJ group had cholangitis while this complication was found in only one patient of HD. So, we did not find any significant difference in the incidence of post-operative cholangitis in the follow-up period. All these patients were managed conservatively.

One patient in HJ group developed adhesive obstruction around three months after surgery. He had to be re-operated for adhesiolysis. Extensive bowel handling and an additional anastomosis increase the chances of such complications in HJ group. However, Shimotakahara et al.did not report any significant difference in the incidence of postoperative adhesive intestinal obstruction in the two groups.[6]

An important observation regarding post-operative nasogastric bilious output was made exclusively in the HD group. 19.15% of patients with HD had significant bilious output in the post-operative period which took time to settle down. In all these patients, there was a delay in starting oral feeds. This can be explained by the direct anastomosis of biliary channel with relatively straightened duodenum which has been Kocherized during surgery and is not guarded by any sphincter, like the naturally present sphincter of Oddi. We analyzed the reason behind not finding increased bilious output in other patients and inferred that an anastomosis distal to the normal biliary opening in a fully Kocherized duodenum

has lesser chances of such reflux. So, the anastomosis is never done proximal to the normal location of Ampulla of Vater and preferably done distal to it in the second part of the duodenum. Several other investigators have found the exclusive occurrence of reflux gastritis in HD patients and have therefore, favored Roux-en-Y HJ on this ground.[6, 15] In a meta-analysis by Narayanan et al., the incidence of reflux gastritis was the only difference between the HD and HJ group of patients; there was no difference in the incidence of post-operative cholangitis, bile leak, anastomotic stricture, bleeding, operative time, reoperation rate, and adhesive intestinal obstruction.[1, 5]

Chances of biliary reflux and associated gastritis in future have a strong correlation and such patients need to be counselled about the need for periodic endoscopy if patients show any symptoms. None of the nine patients with increased nasogastric bilious aspirates in our study have had any other symptoms in the follow-up period. However, they have been told about the need for endoscopic evaluation if such symptoms are noted.

We concluded that although Roux-en-Y HJ is a time-tested gold standard for bilio-enteric reconstruction and no definite advantage of HD is seen in our study with respect to complication rates; still HD is a prompt, feasible and cheap alternative with similar postoperative outcomes.

Conclusion

HD over T-Tube is as good a procedure as Roux-en-Y HJ to restore the bilio-enteric continuity. Being a more physiologic and prompter procedure, anastomosis over T-Tube lessens the intraluminal pressure and edema to further decrease the risks of post-operative leak. Although hazards of possible biliary gastritis in the long term with HD may need endoscopic evaluation, its incidence can definitely lessen by an anastomosis distal to the normal ampulla. A randomized study with more patients may further bring out stronger and conclusive evidence regarding this issue.

Ethical Consideration

This study was approved by Institute Ethics Committee of Indira Gandhi Institute of Medical Science Indira Gandhi Institute of Medical Sciences, Patna s:Sheikhpura: Patna-14

Acknowledgements

Not applicable

Funding/Support

Not applicable

Conflict of interests

There is no conflict of interest

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Comparison Outcomes of Divided End Loop Versus Separate Double Barrel Colostomy in Neonates with Imperforate Anus

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How to cite this article:
Askarpour SH, Peyvasteh M, Kazemnia KH, Javaherizadeh H. Comparison Outcomes of Divided End Loop Versus Separate Double Barrel Colostomy in Neonates with Imperforate Anus. Iranian Journal of Pediatric Surgery 2021; 7 (2): 81-89 .

DOI: https://doi.org/10.22037/irjps.v7i2.34639

Abstract

Introduction: the aim of current is to compare clinical outcomes between the divided end loop and separate double barrel colostomy in neonates with imperforate anus.

Materials and Methods: This is a retrospective study to review 184 patients who presented with imperforate anus and were managed with a divided end loop or separate double barrel colostomy between 2017 and 2020. Complications were compared in the two groups.

Results: There was more skin excoriation in separate double barrel colostomy compared to divided end loop colostomy (p=0.001). The mean of scar size in patients of divided end loop colostomy group was lower than that of separate double barrel colostomy significantly (p<0.0001).

Conclusion: Our results suggest that divided end loop colostomy may be more acceptable than separate double barrel colostomy for neonates with imperforate anus.

Keywords

- Imperforate anus
- Divided end loop
- Separate double barrel
- Colostomy

received: 22 April 2021
accepted: 2 May 2021
Published online: November 2021

Introduction

Anorectal malformation appears in one out of every 4000 to 5000 infants and it is a little more common in males.[1-3]

This risk for second child with imperforate anus is approximately1%.[4] The most frequent defect in males is imperforate anus with a recto urethral fistula. In females, it is a rectovestibular fistula.[5]

Imperforate anus without fistula occurs in about 5% of the anorectal malformations, and is associated with Down syndrome.[6]

Imperforate anus has been known for many years.[1–3] For years, physicians have tried to help these patients with creating orifice in their perineum.[7]

This malformation is divided in two types: low and high. Historically, those with a 'high' defect did not survive. Most of the low-type patients survive despite this defect.[8]

Imperforate anus is a major indication for colostomy in anewborn.[9,10] Most of these patients need a major operation to correct this defect in addition to colostomy. Two types of colostomy are performed: divided end loop and separate double barrel.

A divided end loop colostomy includes an opened intestinal loop that is brought out through an incision in the abdominal wall without complete transection of the colon. As cutting or electrocautery of the colon is not painful, it is divided on the rod 10 days after colostomy formation out of the operating room without the need for anesthesia. The remaining tissue of the colon between two colostomy openings is cut off and the rod is removed and this creates a divided loop colostomy with two holes next to each other. On the other hand, a separate double barrel colostomy needs complete division of the colon which makes a proximal colostomy and a distal mucous fistula that are fixed to the abdominal wall via separateincisions.[11-17]

Since the first time that colostomy was performed to treat for imperforate anus, it has always been a challenge to surgeons; type, site, complications and its management.[18] Several studies have shown the high morbidity associated with colostomies. As so, the current study was designed to compare clinical outcomes between the divided end loop and separate double barrel colostomies in neonates with imperforate anus.

Materials and Methods

A retrospective study was performed at Imam Khomeini & Abuzar hospitals in Ahvaz. The data of all patients who presented with imperforate anus, who were managed by a diverting colostomy (divided end loop or separate double barrel) between 2017 and 2020 was collected.

Children with incomplete medical records and those who passed away, were excluded from the study.

Information was gathered through admission, pre-operative, operation, post-operative, and follow-up clinic notes.

Demographic data included gender, gestational age and weight of birth. Type

and duration of the colostomies were determined via the operation notes.

For statistical purpose operation time, the start of colostomy function and nutrition, duration of hospitalization, wound infection and the duration of colostomy till closure in months were collected.

Presence of megarectum was rouled out by a distal colostogram contrast study.

Then we collected complications after colostomies; these included retraction, prolapse, parastomal hernia, stricture, necrotizing enterocolitis(NEC), recurrent recto-urinary or recto-genital fistula and need for revision. A more comprehensive work-up of patients who developed urinary tract infection (UTI) was also performed. Diagnosis of UTI was based on the clinical symptoms accompanied with a positive urinary culture (colony count > 10^5).[19, 20]

For those who had their colostomies closed at our hospitals, the duration of follow-up was1 year. We registered cases of wound infection, skin excoriation, wound dehiscence and scar size after closure of the colostomy.

Collected data were analyzed using Student's t-test for continuous variables and qualitative variables were compared using Fischer's exact test.

The results were of statistical significance if the calculated p -value was less than 0.05.

Results

In the three-year duration of our study, 196 neonates with imperforate anus were identified.

The mortality rate was 3.7%(7 patients with congenital heart disease who were excluded) and 5 patients were omitted due to incomplete records.

One hundred and two patients (55.4%) underwent a divided end loop colostomy, while separate double barrel colostomy was performed in82patients (44.6%).

The demographic data is shown in **Table 1**, including gender, gestational age, birth duration of hospitalization and time to close colostomy. Only the mean of operative time was found to be statistically lower in patients with divided end loop colostomy ($p<0.0001$). No other complication was of statistical significance between two groups.

Complications after colostomy formation are summarized in **Table 2** including prolapse, need for colostomy revision, megarectum, urinary tract infection, colostomy stricture and necrotizing enterocolitis.

There were no statistically meaningful differences between two groups. Urinary tract infection was detected in 33 patients (18 %) and was due to either Escherichia coli or Pseudomonas. The incidence of urinary tract infection was 16.7 % in patients with divided end loop colostomy compared with 18.2 %in patients with separate double barrel colostomy ($p=0.413$).

The Complications after colostomy closure including wound infection, skin dehiscence, skin excoriation and scar size are compared in **Table 3.**

There was more skin excoriation in separate double barrel colostomy in

comparison with divided end loop colostomies (p=0.001). The mean of scar size in divided end loop colostomy group was significantly lower than that of separate double barrel colostomy (p<0.0001). Rate of other complications demonstrated no significant differences between two groups.

Table1. Demographic characteristics.

variable		loop	Double Barrel	P-value
Gender	**Male N (%)**	67 (65.7%)	51 (62.2%)	0.741
	Female N (%)	35 (34.3%)	31 (37.8%)	
Gestational Age Mean ± SD (weak)		36.1 ± 1.8	34.6 ± 2.3	0.435
Birth Weight Mean ± SD (gr)		2950 ± 54	2830 ± 75	0.511
Fistula	**Perineal Fistula N (%)**	6 (5.9%)	7 (8%)	0.625
	Vestibular Fistula N (%)	20 (19.6%)	14 (15.9%)	
	Recto-urinary Fistula N (%)	22 (21.6%)	21 (23.8%)	
	Recto-Genitalia Fistula N (%)	9 (8.8%)	6 (6.8%)	
Operative Time Mean ± SD (min)		17.8 ± 5.3	48.3 + 12.6	0.001
Hospitalization (day)		3.5 ± 1.2	4.3 ± 1.8	0.451
Duration Feeding Start Mean ± SD (day)		2.4 ± 1.3	3.2 ± 1.6	0.214
Duration of colostomy Mean ± SD (month)		11.6 ± 4.2	12.1 ± 3.4	

Table2. Complication after colostomy formation.

Complication	loop	Double Barrel	P-value
Megarectum n (%)	7 (6.8)	5 (6.7)	0.711
Retraction n (%)	3 (2.9)	3 (3.4)	0.465
Prolapse n (%)	12 (11.8)	9 (10.2)	0.331
Parastomal Hernia n (%)	4 (3.8)	2 (2.3)	0.125

Stricture n (%)	5 (4.9)	6 (6.8)	0.274
Colostomy revision n (%)	9 (9.8)	7 (7.9)	0.241
UTI n (%)	17 (16.7)	16 (18.2)	0.413
NEC n (%)	3 (2.9)	1 (1.2)	0.621

Table3. Complications after colostomy closure.

Complication	loop	Double Barrel	P-value
Wound Infection N (%)	7 (6.8)	8 (9.1)	0.112
Skin dehiscence N (%)	7 (6.8)	9 (10.2)	0.061
Skin excoriation N (%)	12 (11.8)	19 (21.6)	0.001
Scar size Mean + SD (mm^2)	32.3 ± 11.4	95.4 ± 3.7	< 0.001

Discussion

Usually the formation of a colostomy is the first stage of the surgical treatment of a patient with imperforate anus.The colostomy allows the patient to continue to grow while awaiting the definitive corrective surgery. While the creation of inappropriate colostomy can lead to various complications and even life-threatening infections.[21-23]With the right decision about the type of colostomy one can reduce these serious complications.

Complications of colostomies are important factors that have caused some pediatric surgeons to support the primary correction of imperforate anus.[24]

It appears that sufficient distance between proximal and distal openings of the double barrel colostomy prevents the complications such as urinary tract infection, megarectum and wound infection. Separate double barrel colostomy is accompanied with lower incidence of colon prolapse in these patients.[25] On the other hand, using small incisions for creation of divided end loop colostomies allows easier closure and better cosmetic results in child.

Our results showed operative time was shorter in patients with divided end loop colostomy.

The operation time is one of the most important factors which determines patient overall outcome specially in emergency situations. Therefore it seems that divided end loop colostomy in patient with imperforated anus has better outcome than separate double barrel colostomy.

One study showed no difference between divided end loop and separate double barrel colostomy when the loop colostomy

was closed early (2-4 months).[26] Our results demonstrated no meaningful differences between the two groups in terms of duration of colostomy.

In this study, there was no meaningful difference in common complications of colostomy. This is similar to the findings of Patwardhan et al survey,[27] but in contrast to others who have reported an overall incidence of complications as 31%–63% with divided end loop colostomy versus 15%–45% with separated double barrel colostomies.[11,12]

Incidence of colostomy prolapse in our patients (divided end loop 7% vs. separate double barrel 9%) was a little lower than other reports of 15%–18% with divided end loop colostomies and similar to the 3%–6% reported for separate double barrel colostomy.[28]The reason for this result is the fact that we perform most of our colostomies at the first mobile part of the sigmoid colon, immediately distal to the descending colon, whether or not it was to be separated.

Imperforate anus and other anorectal malformations are associated with renal anomalies and it may influence the patient's outcome.[11-14]

The presence of a recto-vesical and recto-urethral fistulae leads to a raised risk of UTI caused by urine absorption in through colon. Study of Singh et al reported a high incidence (86%) of UTI in patients with high imperforate anus with fistula (recto-vesicular).[29]

Our results showed a 23% incidence of UTI in our patients. It agrees with Wiener and Kieswetter the who demonstrated that incidence of associated renal anomalies in the patients who had UTI was particularity high (51%).[12]However the number of patients in group of separate double barrel colostomy is small, incidence of UTI was similar after divided end loop (16.7%) or separate double barrel colostomy(18.2%).

In a survey of complications after colostomy closure the risk of wound infection was increased in separate double barrel colostomy. Incidence of wound infection or peri-ostomal skin breakdown in our study was very small and there was not a significant difference between two groups. Pena et al. did not report any wound infections in all 50 patients with separate double barrel colostomy.[30]

Although according to our results the incidence of skin dehiscence was higher in double barrel colostomy group, but we did not detect significant difference between two groups. Also, a similar finding was reported in previous studies.[21-25]

In this survey we have found that the incidence of skin excoriation between the divided end loop and separate double barrel colostomies is significantly different(divided end loop 11.8% vs. Separate double barrel 21.6%). Another study reported that the rate of skin excoriation was higher in the separate double barrel group compared to the divided end loop colostomy group.[10]

Finally, the extent of wound scar has not been studied in any of the previous studies but in this study we compared scar size one year after colostomy closure and it turned out that size of wound scar is bigger in patients with separate double barrel colostomy than the other group significantly.

Conclusion

The divided end loop colostomy has a shorter operative time and partly fewer complications including skin excoriation and wound scar size compared to the separate double barrel colostomy. According to the shorter operative time and more acceptable appearance after colostomy closure, our data suggests that divided end loop colostomy may be more favorable than separate double barrel colostomy for neonates with imperforate anus.

Ethical Consideration

This study was approved by Ethical Committee of Ahvaz Jundishapur University of Medical Sciences with code number IR.AJUMS.REC.1398.368.

Acknowledgements

Not applicable

Funding/Support

Not applicable

Conflict of interests

There is no conflict of interest.

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Outcome of Surgical Approach and Speech Therapy on Quality of Speech in Patients Suffering Cleft Palate

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How to cite this article:
Mohajerzadeh L,Harirforoosh I, Kazemi R, Khaleghnejad Tabari A, Rouzrokh M, Ghoroubi J, Sarafi M. Outcome of Surgical Approach and Speech Therapy on Quality of Speech in Patients Suffering Cleft Palate. Iranian Journal of Pediatric Surgery 2021; 7 (2): 90 - 97 .

DOI: https://doi.org/10.22037/irjps.v7i2.34973

Abstract

Introduction: A patient suffering from cleft palate has speech problems even after undergoing surgical procedures to correct it. These problems can be improved by some modality of speech therapy. In this study we aimed to evaluate the outcome of our surgical approach and also the impact of speech therapy on quality of speech in patients who suffered from cleft palate and had undergone surgical correction in Mofid hospital since2011 to 2015.

Materials and Methods: We evaluated the quality of speech in the patients suffering cleft palate, older than 3 years who had undergone surgical correction since 2011 to 2015 in our center. Parameters were evaluated in this study included hypernasality, audible nasal emission and disarticulation due to velopharyngeal insufficiency. This process was performed by our center's speech professionals and informed consent was obtained from the patient's parents.

received: 23 May 2021
accepted: 1 August 2021
Published online: November 2021

Results: We studied 202 children, 101 males and 101 females. The first surgical procedure was done in the average age of 16 months. Among the postoperative complications, 90.1% of the patients had hypernasality and 66% of the patients had velopharyngeal insufficiency. All these patients were referred to speech therapy and it was shown that there is a significant improvement in the quality of their speech. There was no significant relationship between gender and prevalence of postoperative complications orhypernasality as one of the speech quality elements (P value: 0.34) and there was no significant difference between the age of first surgical reconstructive surgery and speech quality outcomes, but the early reconstruction had strong relation with reduction in postoperative complications and overall final result (P value: 0.043). According to Kruskal-Wallis statistical analysis, there were no significant superiority on speech quality outcome among the three mentioned different surgical methods (P value: 0.203).Also there was a significant improvement in correcting hypernasality as one of the main complications after speech therapy courses (P value:0.0087).

Conclusion: In this study, supportive measures such as speech therapy have been shown to improve post-operative complications of cleft palate, such as hypernasality, nasal emission and disarticulation due to velopharyngeal insufficiency. Duration of speech therapy was also significantly effective on speech improvement.

Keywords

- Cleft palate
- Hypernasality
- Speech therapy
- Complications

Introduction

Cleft lip and palate are separations in the upper lip, the roof of the mouth (palate) or both. These problems occur when facial structures which develop in the embryo don't close and fuse completely. These are among the most common congenital defects and they are mostly isolated but may occur in relation with many inherited genetic conditions or syndromes.

Patients suffering cleft palate are at risk for speech and language developmental problems.[1]When the palate has these defects and velopharyngeal insufficiency (VPI) is present, feeding, hearing, speech and language problems often occur[2]. Many of the recent literature propose that early surgical cleft repair between 6 to 18 month in these patients has greater speech

and language developmental outcome and reduces hearing loss[3]. Velopalatine insufficiency remains as a common problem regardless of improvement in surgical palatoplasty techniques and its incidence is estimated to be 15-25% in different studies[4-5]. The main goal of surgical cleft palate correction is to achieve complete closure of the cleft and having an intact, smooth and hard palate with normal velopharyngeal function[6]. Facial deformity and speech impairment will not be completely corrected in every newborn receiving surgical correction and this will cause health care and familial burden of disease increment[7]. Speech and language therapists have the ability to decrease this gap and many of the patients will be improved by this approach post surgically. In this study we evaluated the improvement in postsurgical speech quality in patients with cleft palate.

Materials and Methods

In this retrospective study, 202 children were recruited who had undergone complete cleft palate palatoplasty between 2011 to 2015 in our tertiary center of Mofid children hospital. The inclusion criteria of this study was the age of 3 years and older with a history of primary or secondary surgical reconstruction of cleft palate. The age of having surgical reconstruction was12 month in most of the patients with the mean of 16 month in all of them. Genetic syndromes, cognitive delay, neurological syndrome or sensorineural hearing loss and postoperative fistula were the exclusion criteria of this study. All these patients were called back for speech quality evaluation using the available patient's documented information. This evaluation was done by our center's professional speech therapists. In addition to speech quality evaluation, patient's anatomical problems and also craniofacial abnormalities were examined. Data was collected and documented. Symptoms of speech problems include hypernasality, audible nasal emission, and disarticulation due to velopharyngeal insufficiency.Also the improvement in speech ability and quality was re-evaluated by the center's professional speech therapists after patients passing a course of speech therapy. More than these, all the patients passed a hearing screening test at the time of evaluation.

The items of speech quality assessment included nasal emission, articulation and hypernasality. Nasal emission evaluation was standardly done by a mirror and articulation development factors were development of simple sounds, words and connected sentences.

Statistical analyses were performed by using the Statistical Package for Social Sciences, version 20 (SPSS, Chicago, Illinois). Final results were demonstrated as frequency and percentage. We used Fisher's exact test for significance of percentage evaluation between two groups and MnNemar test was also used for comparison of the results before and after the course of speech therapy. P values of about <0.05 were considered as significant.

Results

202 children were enrolled in this study who had undergone cleft palate reconstructive surgery in our pediatric surgical tertiary center, Mofid hospital, Tehran, Iran during 2011 to 2015. One hundred and one of the patients (50%) were male and 101 (50%) were female. 34.2% of the patients resided in Tehran and the rest of the patients were from other cities. Mean age for first surgical reconstruction was about 16 month, but most of the patients underwent this first surgical reconstruction at age of 12 month and the mean duration of hospitalization was 4 days. Earliest age of surgery was 1.5 month and the latest one was 13 years. According to findings of this study, 58 (28.7%) patients had delay in starting to speak in comparison to their normal peers and 182 (90.1%) of patients had difficulty in pronouncing alphabets and 175 (86.6%) of the patients had nasal emission which could happen due to velophar yngeal insufficiency **Table 1**.

Table1: Complications of cleft palate in pronunciation of the words and sound production in the study patients

Complications of cleft palate in pronunciation of the words and sound production	Percentage
Hypernasality	90.1%
Late start to speak	28.7%
Difficulty in pronouncing or uttering alphabets	90.1%
Nasal emission	86.6%
Velopharyngeal insufficiency	66.2%

Of the patients who have been enrolled in this study, 18.3% had a history of recurrent middle ear infection that was caused by anatomical defects in these patients and 12.9% of the patients suffered from auditory problems and were referred for audiology consultation and proper intervention.

Regarding the reconstructive surgical techniques, about 70 patients (35.2%) were managed by the Vean-Ward-Kilner (VWK) pushback method, 128 patients (64.3%) by Von langenbeck and only one patient by Bardach two flap palatoplasty method **Table2.**

Table2: Reconstructive surgical techniques

Surgical technique	Prevalence(Absolute number)	Prevalence(Percentage)	P value
Vean-Ward-	70	35.2%	P value:

Kilner (VWK) pushback			0.203
Von langenbeck	128	64.3%	
Bardach two flap palatoplasty	1	0.5%	

147 patients suffered from velopharyngeal insufficiency as the complication of surgery and 67.8% (137 patients) underwent speech therapy courses. One of these patients underwent sphincter pharyngoplasty and one patient underwent pharyngeal flap imposition and for 4% (8 patients) repeat surgical cleft palate reconstruction was done. In follow up reevaluation, 182 patients (90.1%) had hypernasality and all of them were referred to a speech therapist.

By using mann-whitney statistical study, there was no significant relation between gender of the patients and prevalence of postoperative complications orhypernasality as one of the speech quality elements (P value: 0.34). There was no significant difference between the age of first surgical reconstructive surgery and speech quality outcomes, but the early reconstructive surgery had strong relation with reduction in postoperative complications and overall final result (P value: 0.043).According to Kruskal-Wallis statistical analysis, there were no significant superiority on speech quality outcome among the three mentioned different surgical methods (P value: 0.203).Also there was a significant improvement in correcting hypernasality as one of the main complications after speech therapy courses (P value:0.0087).

Cleft palate would happen in relation with other congenital abnormalities and in this study there was a significant relation between concurrent occurrence of cleft palate and cleft lip (in 88 of the patients: 43.6%) and also cardiovascular congenital disorders (18 of the patients: 9%) or neurological abnormalities (17 of the patients: 8.5%). Two cases of Pierre Robin sequence also occurred. With the aspect of anatomical distribution of the cleft palate, 18.8% of this patients population had only soft palate abnormalities and 80% had complete cleft palate. Also 1% of the patients had submucosal cleft palate. Three and a half percent of all had unilateral and 96.5% had bilateral cleft palate, but according to Mann-whitney study, this made no significant difference in speech quality after speech therapy courses (P value: 0.85).

Postoperative complications of the primary reconstructive surgery of cleft palate include: 42.8% (86 patients) had oral-nasal fistula which among them, 16.8% suffered from fistula in hard palate, 7.9% in soft palate and 17.8% had concurrent hard and soft palate fistulae. Ninety six and a half percent (83 patients) of this group underwent redo surgical reconstruction of fistula but unfortunately 3.5% (3 patients), did not receive the reconstructive surgery because of the poor

follow up. Among the patients in group of oral-nasal fistula, 59.3% (51 patients) needed only one-time fistula reconstruction but 23.3% (20 patients), 10.5% (9 patients), 3.5% (3 patients), 3.5% (3 patients) need two, three, four and five reconstructive surgeries for fistula management respectively. There was no significant relation between fistula formation as a complication of surgical reconstruction and hypernasality as a parameter of speech quality outcome (P value:0.45).

Other findings of this study included: the longest duration of speech therapy was 12 month and the mean was 8 month and there was a direct relation between duration of speech therapy and final speech quality result (P value: 0.0076).

Discussion

Cleft lip and palate are separations in the upper lip, the roof of the mouth (palate) or both. These problems are caused when facial structures which develops in the embryo don't close and fuse completely. These are among the most common congenital defects and they are mostly isolated but can occur in relation with many inherited genetic conditions or syndromes.

Patients with cleft palate are at risk of speech and language developmental problems.[1] When the palate is involved with this defects and velopharyngeal insufficiency (VPI) is present, feeding, hearing, speech and language problems often occur.[2] Many of the recent literature propose that early surgical cleft repair between 6 to 18 month has greater speech and language developmental outcome and reduces hearing loss.[3] Velopalatine insufficiency still remains as a common problem regardless of improvements in surgical palatoplasty techniques and its incidence is estimated to be 15-25% in different studies.[4-5] The main goal of surgical cleft palate correction is to achieve complete closure of the cleft and having an intact smooth and hard palate with normal velopharyngeal function.[6] Facial deformity and speech impairment will not be completely corrected in every newborn receiving surgical correction and this will cause health care and familial burden of disease increment.[7] Speech and language therapists have the ability to decrease this gap and many of the patients will be improved by this approach post surgically.

In this retrospective study we evaluated the improvement in postoperative speech quality in patients with cleft palate who underwent surgical reconstruction during 2011 to 2015. Finally, 202 patients who were older than 3 years, passed the inclusion criteria of the study and were evaluated for their quality of speech and speech problems by a standard protocol of speech evaluation by our center's speech therapy professionals. Thedatawas gathered and documented and finally statistical analysis was done.

In this study we showed that speech therapy is essential and necessary for management of speech quality in patients who underwent cleft palate reconstructive surgery and this approach obviously reduced the velopharyngeal insufficiency

and hypernasality problems in these patients.

Also, the mean duration of speech therapy was 8 month and there was a direct relation between the age of surgery and final speech quality result.

Haapanen et al's study which was done on 108 patients showed that the best and most effective age for surgical reconstruction of cleft palate is between 12 and 18 months old and Cronin modification technique was superior to Push back technique of surgical reconstruction.[8-10] In this study it has been revealed that there is no significant difference between different techniques of surgery in the management of these patients and none of them were superior to the others.

Davari et al estimated that the prevalence of hypernasality, post reconstructive operation is about 70.9% and there was no significant difference between different genders[11].Also Magee et al in their study showed that interventional reconstruction of these defects as the aspect of general health cost burden and cost effectiveness is absolutely effective and it can prominently reduce the disability caused by this defect.[12]The findings of of Mary Hardin-Jones[13-14] indicated a large degree of variability in opinions of speech-language pathologists(SLP),who responded regarding assessment and treatment of children with cleft lip and palate. In the present study the hypernasality prevalence was about 90.1% and we also found that there is no significant difference between different gender groups.

Conclusion

Finally, we showed that post reconstructive operation speech therapy will be useful on improving the speech quality outcome of patients suffering from congenital cleft palate (P value:0.0078). This improvement is in complications such as hypernasality, nasal emission and difficulty in pronunciation of the words due to velopharyngeal insufficiency. Also the duration of speech therapy has a direct relation with final speech quality result. Finally, by the findings of this study, we strongly recommend that surgeons use speech therapy as one of the most important aspects of their patients' cleft palate management**.**

Ethical Consideration

This study was approved by Research Institute of Children Health - Shahid Beheshti University of Medical Sciences with code number IR.SBMU.RICH.REC.1400.016

Acknowledgements

Not applicable

Funding/Support

Not applicable

Conflict of interests

There is no conflict of interest.

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Evaluation of Congenital Diaphragmatic Hernia Repair with Thoracoscope in Neonates

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How to cite this article:
Mohajerzadeh L, Aghajani M, Rouzrokh M, Sarafi M, Fallahi M, Borhani S. Evaluation of Congenital Diaphragmatic Hernia Repair with Thoracoscope in Neonates. Iranian Journal of Pediatric Surgery 2021; 7 (2): 98 - 106 .

DOI: https://doi.org/10.22037/irjps.v7i2.34978

Abstract

Introduction: congenital diaphragmatic hernia (CDH) is one of the most common congenital anomalies with significant morbidity and mortality. The most common form of CDH is Bochdaleck type of the anomaly, with the defect located in poster lateral part of the diaphragm. The aim of this retrospective study was to compare the outcome of open repair (OR) with thoracoscopic repair (TR) for Bochdaleck CDH at Mofid Children Hospital.

Materials and Methods: neonates with Bochdaleck CDH at Mofid Children Hospital from 2015 to 2018 were studied. After meeting the criteria, the information during and after surgery was recorded in the questionnaire. We compared the data with independent T- test and Chi-square.

received: 24 May 2021
accepted: 5 August 2021
Published online: November 2021

Results: a total number of 18 patients were included, of whom 9 underwent thoracoscopic repair and 9 underwent open repair. Mean age at the operation was 5.3 days in TR group vs. 3.89days in OR group(P= 0.9). In TRgroup 37.5% of the patients were female and 62.5% male, versus OR group with 22.2% female and 77.8% male (P= 0.62). Gestational age at birth in TR group was 37.2 weeks vs 37.89 weeks in OR group (P= 0.85).Birth weight in TR group was 2904.22 g vs. 2988.88g in OR group (P= 0.96). Delivery type in TR group was NVD in 22.2% and C/S in 77.8%;vs. 33.3% NVD and 66.7% C/S in OR group (P = 1). In TR group none had prenatal diagnosis, while in OR group 11.1% had and 88.9% didn't have prenatal diagnosis (P= 1). Entrance status to Mofid Children Hospital: all of them were nonintubated, with no family history of CDH. CPR Hx: TR one vs. OR none (P= 1), recurrence: TR 0vs. 1 OR (P= 0.3), Echo abnormal results: TR 44.4 % vs. OR 66.7 (P=0.63), brain abnormal US: TR 0 vs. OR 1 (P= 0.38), skeletal anomaly: 1in TR (P= 1), post-op admission: TR 18d vs. OR 25d (P= 0.04),commencement of enteral feeding: TR 8.7d vs. OR 5.7d (P= 0.78), complications: 1 in OR (P =0.3), survival: 1 death in TR & 1 in OR (P= 1), death age: TR at 270d vs. OR 81d (P= 1).

Conclusion: thoracoscopic repair appears to be as effective as open repair for treating cardiopulmonary stable patients. Thoracoscopic repair is in some way much better than open repair such as wound cosmetic and hospital stay.

Keywords

- Congenital diaphragmatic hernia
- Bochdaleck diaphragmatic hernia
- Bochdaleck diaphragmatic hernia

Introduction

One of the relatively common neonatal abnormalities is congenital diaphragmatic hernia (CDH).[1] It can be associated with significant mortality and morbidity. Determining the factors involved in prognosis and selecting appropriate surgical method can play an important role for increasing the chances of survival in these patients.

Mofid Children Hospital, as a referral center from different parts of the country, accepts a significant number of patients annually who undergo reconstructive surgery. In this study, the effective factors

in choosing the type of surgery by thoracoscopic and open methods were investigated in order to reduce morbidity and mortality. This study is a retrospective one that has been evaluated and reviewed on the files of the hospital. Our study is a comparison of the treatment outcomes of Bucholadalek diaphragmatic hernia, referred to Mofid Children Hospital, with both open and thoracoscopic methods. During this study, the effects of various factors on the results were investigated.

The specific purpose of this study is to identify appropriate preoperative criteria to increase the chances of success of one of these two methods.

Materials and Methods

In this study, cases of congenital diaphragmatic hernia admitted to neonatal Intensive Care Unit in 2015 until 2018were included. Hemodynamically stable, non-intubated patients were chosen for study after explaining both types of thoracoscopic and open surgery and the possible benefits and complications of each technique for the parents and obtaining written consent from them, then they were randomly operated on by thoracoscopic or open surgery. The process of carrying out the study has been approved by the ethics committee of Shahid Beheshti University of Medical Sciences.

In the study, demographic information, information about the disease and type of hernia, length of hospital stay, number of surgical cases, postoperative complications (such as recurrence, mortality, incisional hernia, and wound infection), duration of antibiotic treatment, duration of chest tube in place, duration of ventilator dependency, and need for readmission were assessed.

Input criteria: All stable and non-intubated patients who were diagnosed with congenital diaphragmatic hernia referred to Mofid Children Hospital in Tehran, with the desire and consent of their parents for each of these two surgical methods were included in the study.

Output criteria:

1. Dissatisfaction of the patient's parents to participate in the project
2. Patients who did not stay in the study until the end of the study

After selecting the patients to enter the study, during and after treatment, the information was entered into a questionnaire. The information was entered in the SPSS statistical software version 17 and the two treatment groups were compared using statistical analysis.

Quantitative data were described by means and variance and compared with t-test.

Qualitative data were compared with Chi-square test with frequency and percentage of description.

Ethical considerations**:** Sufficient explanation was given to the patient's parents about both types of surgery and the possible benefits and complications of each technique. Written consent was obtained from the patients and no additional costs were imposed on the patients.

Results

Among the 18 neonates studied in this study, 22% were male and 77% were female in the open surgery group; and 37.5% were female and 62.5% were male in the thoracoscopic group. There was no significant difference between sex and type of surgery (P= 0.62).

The mean gestational age of neonates born in the open surgery group was 37.89 weeks, and in the thoracoscopic group was 37.2 weeks. In terms of treatment results, there was no significant difference between gestational age and type of surgery (P=0.85).

The mean age of surgery in the open surgery group was 3.89 days, and in the thoracoscopic group was 5.3 days. There was no significant difference between the age of surgery and the type of surgery (P=0.9).

Birth weight was 2988 g in open surgery group and 2904 g in thoracoscopic group. There was no significant difference between birth weight and type of surgery in terms of treatment results (P=0.96).

Regarding the type of delivery in the open surgery group, 33.3% were vaginal delivery and 66.7% were Cesarean Section. In thoracoscopy, they were 22.2% and 77.8%, respectively. In terms of treatment results, there was no significant difference between the type of delivery and the surgical method (P=1.0).

In terms of prenatal diagnosis in the open surgery group, 11.9% were diagnosed prenatally and in the thoracoscopic surgery group, all cases were diagnosed after birth. There was no significant difference between prenatal diagnosis and type of surgery in terms of treatment results (P=1.0).

In terms of onset conditions, all 18 cases were non-intubated, of which 9 neonates were in open group and 9 cases in thoracoscopic group.

There was no family history of hernias in either groups.

Regarding the history of consanguineous marriage, in the open surgery group, 44.4% were unattached and 55.6% were attributed. In the thoracoscopic surgery group, 77.8% were unassigned and 22.2% were attributed. In terms of treatment results, there was no significant difference between the history of consanguineous marriage and the type of surgery (P=0.335).

In open surgery group, 16.7% were born in the Capital and 83.3% in some other cities. In the thoracoscopic group, it was 22.2% and 77.8%, respectively. In terms of treatment results, there was no significant difference between the place of birth and the type of surgery (P=1.0).

66.7% of patients who underwent open surgery and 44.4% of patients who underwent thoracoscopy had abnormal echocardiographic results. There was no significant difference between surgical method and echocardiographic results (P= 0.63).

In the study for other anomalies, 2 patients had accompanying other anomalies; who underwent open surgery. There was no significant difference between other anomalies and surgical method in terms of treatment results (P=0.47).

In terms of type of organ in the thorax; there was small intestine in 12 people,

large intestine in 13, and stomach in 4, and spleen in 5. In terms of treatment results, there was no significant difference between the organ in the thorax and the type of surgery (P=0.15).

None of the patients had any gastrointestinal abnormalities during the operation. Patients' Characteristics are showed in **Table 1**.

Table 1. **Patients' Characteristics (n = 18)**	**Open Group (9 cases)**	**Thoracoscopic group**	**P value**
Gender (female)	77%	37.5%	0.62
mean gestational age (weeks)	37.89	37.2	0.85
mean age of surgery (days)	3.89	5.3	0.9
Birth weight(g)	2988	2904	0.96
type of delivery (cesarean)	66.7%	77.8%	1
prenatal diagnosis	11.9%	0%	1
consanguineous marriage (attributed)	55.6%	22.2%	0.335
Cardiac abnormality	66.7%	44.4%	0.63

Hernia's side in all cases was on the left.

One patient in open group recurred. There was no significant difference between surgery and recurrence (P=0.47).

The mean duration of postoperative hospitalization was 25 days in the open group and 18 days in the thoracoscopic group. In terms of treatment results, there was significant difference between the duration of hospitalization and surgery (P=0.04).

The mean period of postoperative feeding was 8.7 days in the open group and 5.7 days in the thoracoscopic group. There was no significant difference between the surgical method and the age of onset of feeding (P=0.78).

Postoperative complications were seen only in one patient with open surgery. In terms of treatment results, there was no significant difference between postoperative recurrence and surgical procedure (P=0.3).

For survival, one died in the surgical open group and one died in thoracoscopy. In terms of treatment results, there is no significant difference between survival and surgical method (P=1).

The age of death was 81 days in open surgery and 270 days in thoracoscopy group. There was no significant relationship between age of death and surgical method in terms of treatment results (P=1). Post operative results are shown in **Table 2**.

Table 2: Postoperative results

Postoperative data	Open Group	Thoracoscopic group	P value
-Hernia's side(left)	100%	100%	
-recurrence	11%(1)	0%	0.47
-mean duration of postoperative hospitalization(days)	25	18	0.04
-mean of postoperative feeding(days)	8.7	5.7	0.78
Mortality(N)	1(81 days)	1(270 days)	1

Discussion

Considering that Mofid Children Hospital is a referral center for surgery of newborns with diaphragmatic hernia and due to the high prevalence of this anomaly, this study is used to compare thoracoscopic and open methods for repair and also to evaluate the effective factors in choosing the method. In one study, most of the infants were male and the sex of the infant had no effect on determining the outcome of treatment.

In terms of birth weight, Apgar score, type of delivery by natural methods or Cesarean Section, prenatal diagnosis, conditions of the birthplace of the infants, type of organ in the thorax, echocardiography and brain sonography results, there were no significant correlations between the two methods.

Gourley[2] and colleagues performed study on congenital diaphragmatic hernia onneonates. They reported successful thoracoscopic surgery is expected in infants with good pulmonary condition (the more stable the baby was). In this study the mean age of surgery was 3.89 days in the open group and 5.7 days in the

thoracoscopic group, and all cases were in good condition.

Gander JW[3] et al. reported thoracoscopy is associated with lower morbidity and faster recovery but has an increase in recurrence or complications compared to the open surgery. They concluded that early recurrence in thoracoscopic method is more than open surgery. In unstable patients, thoracoscopic repair is not recommended due to further recurrence. In this study, there was no significant difference between recurrence and surgical method.

Costerus S[4] studied on 108 neonates with appropriate criteria for repair of CDH, 75 neonates underwent thoracoscopy and 34 neonates underwent open surgery. The results of thoracoscopic and open repair were almost similar, but the recurrence rate was just higher in the thoracoscopic group. In the current study, postoperative complications were seen only in one patient in open surgery group. In terms of treatment results, there is no significant difference between postoperative recurrence and surgical procedure (P value= 0.3).

Gomes Ferreira[5], performed thoracoscopy in 40 neonates. Significant statistical differences were found between results of repair secondary to postnatal PaCO2 >60 mmHg, intrathoracic position of the stomach, signs of pulmonary hypertension on the postnatal cardiac ultrasound, and preoperative O2 saturation. They reported that an oxygenation index>3.0 was the only suggested accompanying factor of conversion to open surgery, and considered it as a main post-operative complication of thoracoscopic surgery. Congenital diaphragmatic hernia is able to be repaired securely in the neonates by thoracoscopic surgery. The limiting factor for repair by the thoracoscope is persistent pulmonary hypertension of the newborn. The best preoperative indicator for persistent pulmonary hypertension of the newborn is oxygenation index. Prospective studies are essential to show the efficiency of these risk factors for use as assistant criteria for planning type of surgical protocols in neonates with CDH. In the current study, stable non-intubated patients were chosen for study after explaining both types of thoracoscopic and open surgery details to the parents.

Takaki Tanaka[6] studied on 24 neonates with congenital diaphragmatic hernia, identified prenatally or within 6 h after birth, in a 10-year-period with mild pulmonary hypertension and without the use of inhaled nitric oxide. Open operation was performed routinely until 2006 (n = 14) and thoracoscopic repair became routine in 2007 (n = 10). They concluded that thoracoscopic repair seems to be as effective as OR for management of carefully chosen cases of CDH in neonates, with superior wound cosmesis.

Schneider A et al.[7] reported that thoracoscopy can be successfully used for the operation of CDH in selected cases; but pulmonary hypoplasia is a major restrictive factor. Some cases should be omitted secondary to higher probable requirement for patch repair because of technical difficulties and prolonged operative duration. The success is related

to close teamwork activity among pediatric surgeon, neonatologist and pediatric anesthetist.

The essential problem during repair of diaphragmatic hernia is presence of intraoperative acidosis and hypercapnia unrelated to the method used. Nevertheless, this would be extremely severe during thoracoscopic repair.[8-10] The consequence of intraoperative hypercapnia and acidosis on the neonate's neuro developmental status is unidentified. So, selection criteria for the cases of thoracoscopic repair are obligatory.[11,12,13] Putnam LRetal[14] reviewed a total of 3,067 congenital diaphragmatic hernia patients who underwent open (n = 2,579; 84%) or minimally invasive surgery (n = 488; 16%). Neonates experiencing open approach were more probable to be identified prenatally, be premature, had lower 5-minute Apgar scores, and had main cardiac anomalies. After risk assessment of CDH patients, an MIS method was autonomously accompanying with diminished interval of hospital stay and adhesive small bowel obstruction, but more recurrence rates.

Conclusion

In conclusion, diaphragmatic hernia is one of the most common anomalies in infancy. The study on the effective factors determining the appropriate surgical method and the results of these methods can have a significant impact on the better outcome of these infants. The thoracoscopic method seems to be as effective as the traditional open surgery in neonates with stable cardio-respiratory system, and this method can be used in patients with stable condition, with aesthetically much better results.

Ethical Consideration

This study was approved by Research Ethics Committees of Shahid Beheshti University of Medical Sciences with code number IR.SBMU.RETECH.REC.1396.174.

Acknowledgements

Not applicable

Funding/Support

Not applicable

Conflict of interests

There is no conflict of interest.

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Early Detection and Aggressive Management Is a Key to Success in Management of Childhood Constipation

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How to cite this article:
Kalavant B A, Swathy A R, Shetty SP, Annigeri V. Early Detection and Aggressive Management Is a Key to Success in Management of Childhood Constipation. Iranian Journal of Pediatric Surgery 2021; 7 (2): 107 – 120.

DOI: https://doi.org/10.22037/irjps.v7i2.35457

Abstract

Introduction: Functional constipation is an emerging problem in childhood of Indian subcontinent. If diagnosed early and managed aggressively along with lifestyle modification the success rate of disease treatment improves.

Objectives:

- To assess whether the pain abdomen is the early feature of constipation
- To determine the relationship between good follow ups and the outcome
- To determine the minimum duration of laxative therapy in management of constipation

Materials and Methods: A retrospective analytical study of 150 children with constipation. Various parameters were considered namely demographic

received: 11 July 2021
accepted: 14 August 2021
Published online: November 2021

details, eating habits, clinical examination, abdominal radiographs and abdominal ultrasonography. Patients were divided into two groups based on the symptoms at the presentation. The first comparison was between constipation group vs pain abdomen group. Second comparison was between the patients with good follow-ups and bad follow-ups. The finally the duration of laxative therapy was assessed. Data was entered into Microsoft Excel and analysed using SPSS version 25. The categorical variables are presented using frequency and percentages. Chi square tests were used for statistical analysis. $p<0.05$ is considered as significant value for interpretation of results.

Results: All the 150 patients were treated for constipation 70% showed improvement. Eighty percent of pain abdomen group patients also showed improvement in their symptoms suggesting that they might be the subset of larger constipation group. 97.1% patient with good follow-up followed the dietary advice. The study reveals that there is an association between following dietary advice and final improvement. 70.5% who followed dietary advice had good outcome irrespective of the use of regular laxatives. Most of the patients (78 %) showed the improvement with 3- 6 months of laxative therapy.

Conclusion: Pain abdomen with stool stasis must be considered as early feature of constipation. Patients with better follow and good dietary habits have significant improvement. Aggressive therapy with two laxatives for 3-6 months can increase the success.

Keywords

- Childhood constipation
- Functional constipation
- Nonspecific pain abdomen

Introduction

The worldwide prevalence of childhood constipation varies between 0.7% and 29.6%.[1] Constipation is not an infrequent problem in the Indian subcontinent, in contrast to what was previously assumed. Various articles suggest that the constipation prevalence is rising due to changing lifestyle. An early diagnose of

constipation and aggressive management along with the life style modifications can increase the success rate of the disease treatment.[2] The organic causes of constipation are not more than 5%, majority of the cause of constipation beyond the neonatal period is functional in nature, also known as idiopathic constipation.[3] Childhood functional constipation is often regarded as a trivial symptom that would gradually disappear, but it is not true. Various studies show that overall success rate in constipation treatment is 50-60% at the end of 1 year of intensive therapy and goes up to 68% by 5 years. Among them 80% of the children treated early in the course of disease recovered completely compared to 32 % of patient where initiation of the treatment was delayed.[4] Effective treatment within 3 months of symptoms leads to better outcome. The common cause of intractable constipation is inadequately treated constipation and irregular follow-ups. If not taken care of effectively one third of the children continue this problem to adolescence.[5]

Materials and Methods

This is a retrospective analytical study, in which150 patients aged between 2 and 14 years who presented to pediatric surgery out-patient department (OPD) since June 2017 till May 2020 with various symptoms strongly suspicious of constipation were reviewed. These patients were followed up for a minimum of 1 year before making a conclusion. Follow-up parameters were recorded to assess the outcome such as: age and gender of the child, duration of symptoms, different modes of presentation, compliance to the treatment in terms of following the dietary advice and duration of treatment, grading of stool retention in abdominal radiograph, nature and quantity of consumption of junk food, consumption of quantity of fluid per day, final outcome of the treatment The following patients were included in the study

A) Patients presenting with the clinical feature suggestive of constipation based on ROME-3 criteria and were classified as "constipation group"
B) Children who presented with non-specific abdominal pain as the only symptom along with stool retention, identified by clinical examination or abdominal radiographs not matching the ROME -3 criterion were classified as "pain abdomen group"

Following patients were excluded from the study.

A) Patients who had organic causes of constipation; diagnosed initially or Later in the course of management, eg.Hirschsprungs disease and anorectal malformation.
B) Mentally retarded individuals
C) The patients with incomplete medical records and irregular follow-ups.

These patients were assessed with thorough clinical examinations (including digital rectal examination), abdominal radiographs andultrasonography. All were treated for constipation, as the stool stasis was demonstrated in them.

Blethyn's classification was used to grade stool retention in abdominal radiograph, namely:[6]

Grade 0- Normal study, minimal stool noted in rectum and cecum

Grade 1- Minimal stool scattered in the region of the colon other than normal areas

Grade 2- Significant amount of stool loaded in all the segments of colon allowing gas in between.

Grade 3- Assigned when whole of the colon is filled with stool with dilated colon and impacted stool in rectum.

Patient information was collected about the stool pattern and frequency based on Bristol stool charting and Rome -3 criteria

The parents (preferably the mother) were counselled for 35-34 minutes regarding the dietary advice and other management.

The patients who had impacted stools as seen in abdominal radiograph were admitted for rectal wash with 20ml/kg of normal saline (NS) 2-3 time a day. Non responders were offered bowel cleansing using NS, 30ml/kg/hour for 3-4 hours administered per orally or through nasogastric tube.[4]

Patients who were less severely affected were subjected to laxative therapy. Two laxatives were used; polyethylene glycol and liquid paraffin.

Patients were called for review on 15^{th}, $30^{th}$and 90^{th}days for titration of the dose.

The decision on tapering the laxative therapy was considered after 3 months of therapy, if there was a significant improvement. In the absence of improvement, the laxative therapy would be continued.

The patients were divided into different groups. Initially comparisons were made based on the symptoms at presentation- Constipation group vs. pain abdomen group to estimate whether pain abdomen group was a subset of constipation group.

Another comparison grouping was to see the relationship between the follow up and compliance for the dietary advice.

Patients were divided into good follow-up and bad follow-up groups. If the patients visited to our OPD at regular interval as advised, they were considered to have good follow-ups. Their compliance for the diet was assessed in these patients. Assessment of final improvement was also made in these patients.

Third comparison was made between patients taking laxatives for at least 3 months as advised and the other group being the one who had discontinued laxatives before three months (against the medical advice) to know the minimum period of laxative therapy. The final out-come was assessed in both these groups.

Statistical analysis

Data was entered into Microsoft Excel and analysed using SPSS version 25. The categorical variables are presented using frequency and percentages. Chi square tests were used for statistical analysis. $p<0.05$ was considered as significant value for interpretation of results.

Results

Out of 150 patients, 94 patients were diagnosed with constipation based on the Rome 3 criteria. Fifty-six patients were mostly referred to us for exclusive abdominal pain with occasional other

nonspecific symptoms. Seventy-five percent of the patients belonged to the age group of 2-10 years. The profile of the patient are noted in **Table 1.**

Table1: Distribution of study participants(n=150) according to different study variables

Sl No	Variables	Classification	N=150	%
1	Age (in years)	2-5	57	38.0
		6-10	56	37.3
		11-14	37	24.7
2	Gender	Male	96	64
3	Constipation (As per ROME -III criteria)		94	62.6
4	Patient presenting with exclusive pain abdomen		56	37.3
5	Hard stools as presenting complaints		68	45.3
6	Foul smelling stool		18	12
7	Abdominal distension		56	37.3
8	Encopresis		10	6.6
9	Stool with holding manoeuvre		27	18
10	Bleeding per rectum		26	17.3
11	Genital handling		8	5.3
12	Rectal mucosal prolapse		3	2
13	Vomiting as the only presenting symptoms		4	2.6
14	Pain abdomen (Overlapping with other symptoms)		132	88
		Central or generalized pain abdomen	91/132	68.9
		Right iliac fossa	14/132	10.6
		Hypochondrium	2/132	1.5
		Left iliac fossa	12/132	9
		Flank on either side or both	10/132	7
		Epigastric pain	4/132	3
15	Urinary symptoms		45	14.8
		Day time Incontinence	14/45	31.1
		Increased frequencyof passing urine	18/45	40
		Enuresis	3/45	6
		LUTS	6/45	13.3
		Documented UTI	4/45	8
16	Abdominal radiograph	Blethyn's classification		
Missing =22 (14.7%)		0	2/128	1.5
		1	37/128	28.9
		2	43/128	33.5
		3	46/128	35.9
17	Loaded rectum	Yes	114	76
18	Referred to us as appendicitis	None of them were operated on first visit	8	5.3

	based on USG report			
19	Dietary compliance	Following diet as prescribed	95	63
20	Follow up	Coming for regular follow up	68	45.3
21	Laxative therapy	Duration		
		<3months	62	41
		3-6 months	76	50.6
		>6months	12	8
22	Need for enema	Yes	63	42
23	Need for gut irrigation	Yes	18	12
24	Final overall outcome	Improvement noted in	105	70

USG of abdomen of forty-five patients in pain abdomen group showed mesenteric lymphadenitis in 26, sub-acute appendicitis in 8, intussusception in 4, ileal thickening in 1, 4 patients had cholelithiasis and 16 had upper tract stones. These patients had significant stool retention in abdominal radiograph (Blethyn's grade type 2 and 3) and loaded rectum in 66% each respectively. Only 3 patients required appendicectomy, one needed cholecystectomy and four required surgical intervention for urinary calculi later in the follow-up.

Comparison was done between the constipation group and pain abdomen groups **Table 2**, which showed that the majority of children in age group 11-14 years complained of pain (64.9%) and majority of the children in age group 2-5years presented with constipation (78.9%). The study also found that stool retention in the rectum was a predominant feature noted in constipation group (82.9%). Both groups were comparable in terms of gender, need for enema and findings in the abdominal X-ray. There was a significant improvement in the pain abdomen group after they were treated for constipation.

Table 2: Comparison between the constipation group and pain abdomen group.

Characteristics		Constipation		Chi square value	p value
		Presenting as constipation (%)	Presenting as pain (%)		
Age group (In years)	2-5	45 (78.9)	12(21.1)	18.508	<0.001*
	6-10	36(64.3)	20(35.7)		
	11-14	13(35.1)	24(64.9)		

Sex	Female	35(64.8)	19(35.2)	0.166	0.683
	Male	59(61.5)	37(38.5)		
loaded colon	No	16(44.4)	20(55.6)	6.723	0.010*
	yes	78(68.4)	36(31.6)		
Enema	No	50(57.5)	37(42.5)	2.39	0.122
	Yes	44(69.8)	19(30.2)		
X ray #	1	20(54.1)	17(45.9)	0.693	0.707
	2	25(58.1)	18(41.9)		
	3	29(63.0)	17(37.0)		
Improvement	No	34(75.6)	11(24.4)	4.565	0.033*
	Yes	60(57.1)	45(42.9)		

*p value <0.05 is considered statistically significant
Xray#: No records were found for 22 patients and 2 records were found to be normal

The final improvement and regular follow-up attitude of patient's guardian were compared with the dietary compliance of the patients to look for any correlation between them in **Table 3**.
Study shows that 97.1%of patients with good follow-up followed the dietary advice whereas only 35.36% of patients among bad follow-up group were complaint to the dietary advice. The study reveals that there is an association between following dietary advice and final improvement. Among the patients who followed the diet, 70.5% had good outcome irrespective of the duration of therapy.

Table 3: Association of following dietary advice with follow up and final improvement

Characteristics		Following dietary advice		Chi square value	p value
		No	Yes		
Follow-up	Poor Follow-up	53(35.3)	29(19.3)	60.925#	<0.001*
	Good Follow-up	2(1.3)	66(44.0)		
Final improvement	No	24(16.0)	21(14)	7.69	0.006*
	Yes	31(20.7)	74(49.3)		

*p value <0.05 is considered statistically significant
Fisher's Exact test is used
Note: total %'S are reported

The Table 4 shows the impact of duration of therapy on the outcome of constipation.

Table 4: Association of final improvement and duration of treatment

Duration of treatment(in months)	Final improvement		Chi square value	p value
	No	Yes		
≤3	28(45.2)	34(54.8)	11.568	0.001*
>3	17(19.3)	71(80.7)		

*p value <0.05 is considered statistically significant

Patient who had laxative therapy for more than three months had 80.7% improvement in their symptoms. Majority of these patients had good follow-ups, were complaint to the medication and dietary advice. Patients who had discontinued the treatment before 3 months noted initial improvement in 54.8% patients, without long-term follow-ups. Seventeen of our patients (19%) didn't respondto our treatment despite regular follow-ups and laxative therapy. Among them 4 patients didn't follow the given dietary advice well. Nine patients recovered with 1 year of laxative therapy but had waxing and waning symptoms hence were not considered cured. Six patients who were not relieved of symptoms, 2 were laxative dependant with more than 18 months of therapy. Three patients were subjected for barium enema which were normal. All the three children, subjected for nuclear scintigraphy had normal colon transit timewith obstructive pattern at the rectum. Among the seventy-one patients were relieved of their symptoms after good follow up and more than 3 months of laxative therapy Table 4 seven patient required laxative therapy ranging from 8 months to 2 years.

Discussion

Constipation is perceived differently by the patients, parents and by treating physicians. There were many criteria for diagnosis such as Lowa system, Loening-Baucke criteria, etc. used in the past as it was not well defined. In 2006, Paris consensus led by concerned specialists, developed new diagnostic criteria which is now popular, known as ROME-3 consensus.[7] The North American Society for Pediatric Gastroenterology, Hepatology, and Nutrition (NASPGHAN) and European Society for Pediatric Gastroenterology, Hepatology, and Nutrition (ESPGHAN) have further modified the criteria by lowering the

duration of symptoms, to avoid unnecessary delay in diagnosis of constipation.[7] The above-mentioned criteria is still considered restrictive by some of the authors as these are formulated based on the western standards. Bowel movement and frequency of defecation are highly influenced by geographical situation, staple diet of the communities and diverse living conditions. Forinstance, average stool frequency in Indian subcontinent is 7 time a week. Colon transit time (CTT) is faster in Asians (16.6 hours). The upper limit of CTT is 31. 8 hours in Indian population compared to European study which is 63 hours.[8] Encopresis is seen in up to 75-90% and are more common in the boysin western literatures.[9, 10] It was not common in our study but was limited to 10 patients (6.6%), among which six patients were male and two patients were younger than 4years. These factors suggest that the disease pattern and severity is different in the Asians.

Patients or parents often complain about pain in the abdomen as a presenting symptoms noted in up to 33% -55% of the cases in one of the studies.[11] About 123 (82%) patients presented with abdominal pain in our series. Fifty-six among them presented with exclusive pain without any other symptoms. These patients had significant stool retention in abdominal radiograph (Blethyn's grade type 2 and 3) and loaded rectum in 66%.Mesenteric lymph nodes in abdominal ultrasonography were noted in 47 %. We thus suggest that a triad of nonspecific abdominal pain, essentially normal USG abdomen with mesenteric lymphadenitis and features suggestive of stool retention in abdominal radiograph is indicative of constipation and would avoid unnecessary delay in the initiating treatment for constipation **Figure 1**. Other vague symptoms are noted in the **Table 1**. Most of the patients are treated for nonspecific symptoms with recurrence of pain agonising the patient's family. The patient may present with urinary incontinence and frequent urinary tract infection and rectal bleed.[12]In our study 14.3% and 17.3 % of patients presented with bladder and bowel symptoms respectively. The other rare symptoms, with which patient may present is recurrent vomiting and upper abdominal distension. This could be attributed to reversal of gastrocolic reflux, where the individual develops gastric paresis due to filled rectum[13,14] We too had 4 patient (3 male and 1 female) who presented without any symptoms other than vomiting who had been thoroughly worked up for the cause. Significant stool retention was the only positive findings in imaging studies. All of them had shown improvement after bowel cleansing therapy and continuing laxative therapy. The precipitating factor for the disease might be low socioeconomic status, over-crowding, inadequate hydration, non-availability of toilets, intercurrent illness and dietary practices in the communities.[5]Eight of our patients were staying in hostels who presented with recurrent symptoms to us. They couldn't follow the medical and dietary advices, who later lost to the follow-ups. Three adolescences; one male and two females, with the constipation had

problem of open-air defecation due to lack of sanitary facilities who also didn't show the improvement and lost for follow-ups. Rushing to school in the morning was noted in 31 of our patients, which may also have been precipitating factor in constipation.

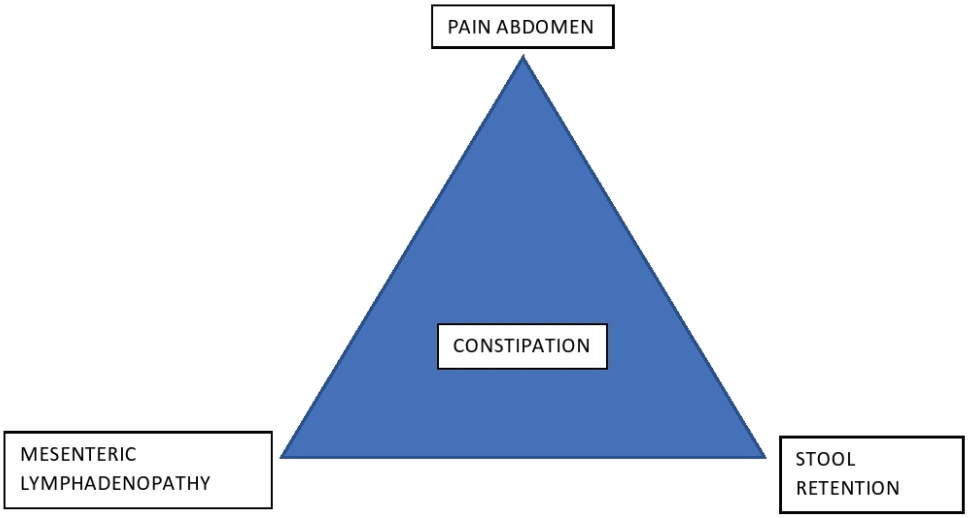

Figure 1: Triad for early detection of constipation

The prevalence of the constipation is not known in our country, but it is on raise in Asian countries as reported from Taiwan (32%), Hongkong (29.3%), Saudi Arabia (22.5%), Srilanka (15.4%) and China (12.2%).[15] There are articles which suggests no difference in incidence between the genders, but we have noted that the boys presented more frequently in our study. This is similar to the findings noted by van der Plaset al.[16] Three fourth of the constipation cases were noted in the age group below 10 years. Similar trends are also noted by authors in the west. In our study we found that the children less than 5 years predominantly presented with constipation and children above 10 years had predominantly pain in the abdomen Treatment of constipation starts from the disimpaction of the stools, studded in distal colon. It can be identified by either palpating abdominal lump, by feeling hard stool in digital rectal examination or with the help of abdominal x ray. The abdominal radiograph is not considered to be a sensitive tool in assessment of constipation according to various studies as the sensitivity and specificity of the investigation was not uniform when done by other authors.[17] But we are of the opinion that stool stasis can be identified in patients presenting with the subtle symptoms on radiograph. There are different grading systems designed by Leech, Barr and Blythen. Latter is by far more simplistic and is used by us in our study.[6]

Poly-ethylene glycol is unequivocally the initial laxative of choice in the management.[18] Patient not responding to the above management would be treated by normal saline enemas. We use normal

saline enema for bowel cleansing when enema fails. Only one of our patients had hypokalemia after the bowel cleansing. Oral disimpaction and enemas are accepted first line of disimpaction therapy.[19] Rectal disimpacition has been more successful treatment compared to more acceptable oral therapy.[20] We have lower tolerance in using rectal disimpaction at the first visit in case of severe stool retention. Strict dietary follow-ups in the treatment of constipation and it had important bearing in long term maintenance of bowel movement. Diet or laxative alone cannot be effective in improving the outcome, but the combined therapy can give us better results. Family education also has an important role in the achieving the success of the treatment. Our average consultation time with each patient in their each visit is 35- 45 minutes.

There is no consensus regarding the duration of laxative therapy in treatment of constipation. Many clinicians prescribe it for more than a year. When we followed the policy of two laxative approaches **Table 4**, 78% of the individuals following diet and taking laxatives for 3-6 months were relieved from the symptoms and remained so within 2 years of follow up. We recommend the constipation to be treated by two laxatives for at least 3 to 6 months with good diet and follow up. In our experience the patients having severe perianal pain and tight anal sphincter on digital examination, local application of combination of muscle relaxant and local anaesthetic agent or occasional use of systemic nonsteroidal anti-inflammatory agents have been beneficial in 6 of our cases to break the chain of painful defection and constipation. One of our patients had perineal abscess which didn't recur after treating constipation.

Need for elimination of cattle milk from the diet is debatable. There are literatures in support of the elimination of the cow's milk.[21]We also advise to avoid milk in the diet.[22]

Role of fibre in the diet is immense.[23]But, only administration of dietary fibres should not be relied upon in the management of childhood constipation. The fibres can reduce the flatulence and abdominal pain in patients with chronic constipation. It also reduces the fecal incontinence rate and increases overall treatment success.[24]

Many of our patients had unhealthy diet, such as the junk food available consisting of lot of oil, food cooked using refined flour, cookies, etc. Excessive junk food consumption was noted in 47 of our patients and exclusive biscuit consumption was seen in 15 of our patients. The parents informed that the symptoms improved after stopping the consumption of such foods. Water consumption as advised in the guidelines given by American dietary recommendation, institute of medicine, was not met in 96 percent of our patients.[18]

In the refractory disease, various pharmacological treatment alternatives have been tried. Erythromycin showed some significant results in a group of 14 children and can be an option in constipation.[25]Lubiprostone, Linaclotide, and Prucalopride are other novel drugs

that have been found to be effective in constipated adults. To date, no randomized studies have been published in children.[4] Malone anterograde enema and sigmoidectomy are also advocated in retractable constipation.[26] We have not used any of these medications nor performed any surgical interventions.

Hypothyroidism was evaluated in 232 patients who presented with constipation to our OPD, 3 patients had subclinical hypothyroidism and 2 had clinical feature of hypothyroidism. This is similar to the study done by William et al, where they noticed hypothyroidism in 56 patients out of the 873 patients, out of which, 9 patients had clinical hypothyroidism. NASPGHAN does not recommend routine thyroid assessment except in cases of severe and refractory constipation.[27]

Conclusion

A total number of 150 children were followed up for management of constipation. Ninety-four (62.7%) patients presented with constipation, whereas the rest of them presented with non-specific symptoms. Children presenting with pain in the abdomen and stool stasis have to be considered as early feature of constipation and need early and aggressive therapy. Thyroid function test is not essential in work up of constipation, but abdominal radiograph should not be underestimated in assessment of constipation.

Laxative therapy along with dietary advice has better outcome in our study compared to either of them alone.

Aggressive therapy with two laxatives for 3-6 months can relieve symptoms in about 78% of patients provided physicians devote sufficient time for counselling for diet and regular follow ups. Bowel cleansing and enemas form an important part of aggressive management.

Ethical Consideration

This study was approved by the institutional ethical committee vide the no. SDMCDS IEC. No. 2021/Medical/ Pediatric/S/05 dated 15-06-2021.

Acknowledgements

Not applicable

Funding/Support

Not applicable

Conflict of interests

There is no conflict of interest.

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Pediatric Patients Undergoing Surgery with Peroperative SARS-Cov-2 Infection: An Iranian Case Brief Report

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How to cite this article:
AskarpourSh, Yousofzadeh M, Khoshkhabar M, Ilkhanipak H, Rahim F, KazemniaKh. Pediatric Patients Undergoing Surgery with Peroperative SARS-Cov-2 Infection: An Iranian Case Brief Report. Iranian Journal of Pediatric Surgery 2021; 7 (2): 121 – 125.

DOI: https://doi.org/10.22037/irjps.v7i2.34959

Abstract

Introduction: The present study aimed to assess COVID-19 disease complications andits related 30-day mortality in pediatric patients with perioperative SARS-CoV-2 infection who had surgery.

Materials and Methods: A multi-center, prospective, brief report of pediatricpatients who had surgery at 6 hospitals in 4 cities of Khuzestan Province, South-western Iran, one of the provinces with the highest prevalenceand death rates due to COVID-19 disease. COVID-19 status and its effect on the course and outcome of the patients was investigated.

Results: 246 pediatric patients who had surgery between Jan 20 and Jun 01, 2020 with a 30-day follow-up period enrolled in the study. Four (1.62%) of the 246 patients

received: 21 May 2021
accepted: 10 August 2021
Published online: November 2021

who underwent surgery had perioperative COVID-19 infection. The most common symptoms included dyspnea, fever, and cough. Surgical procedures included total gastrectomy and esophagojejunostomy, bilateral pleural effusions tap, catheter placement for dialysis, and CV-line placement.
Three patients had comorbidities including congestive heart failure (CHF), end-stage renal disease (ESRD), and diabetes.

Conclusion: Based on our results, it can be said that the prevalence of this disease in children is lower than the average of the society; and the outcome in younger patients seems to be better. Though it seems that COVID-19 disease is a low risk and somehow benign condition in children undergoing surgery, but due to the unpredictable nature of the disease, public health recommendations at both general and special levels have been made by the World Health Organization (WHO) to prevent the disease. Further studies with larger samples are necessary to confirm our findings and to clarify which age groups are at increased risk for developing severe COVID-19 infection and its related morbidities and death.

Keywords

- COVID-19 infection
- 30-Day mortality
- Comorbidities

Introduction

Since the end of December 2019, severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) has spread in Wuhan, China, and It has caused the loss of life of 462,500 people so far.[1] COVID-19 related symptoms include fever and sometimes breathing problems, such as shortness of breath, sore throat, and runny nose.[2] The first patients with the disease were present at the seafood market in Wuhan; however, due to the spread of the disease in people who have not been in contact with animals, the World Health Organization (WHO) has also considered human-to-human transmission. Most countries in the world have started serious prevention programs to combat the disease. Although many attempts have been made to produce the vaccine, there is still no vaccine or antiviral drug to eradicate COVID-19 disease. Besides, there is no definitive cure, prevention or treatment for COVID-19 infections in general.

Preoperative evaluation is an essential part of the professional work of any anesthesiologist, surgeon, and intensive care specialist to ensure a low-risk and safe anesthesia, and operation as well.

Surgical patients are a vulnerable group at risk for SARS-CoV-2 infection, due toexposure to the hospital and subsequent pulmonary complications, they are also particularly sensitive to the inflammatory response of cytokines and the suppressive response of the immune system to surgery.[3] Recently, an international, multi-center, cohort study was conducted at 235 hospitals in 24 countries which included all patients undergoing surgery who had SARS-CoV-2 infection, and reported that postoperative pulmonary complications occur in half of these patients, and those surgeries are associated with high mortality rates.[4] Given the importance of postoperative pulmonary complications and mortality that affect the surgeon's evidence-based decisions making, instructions and guideline have been publishedfor management of surgical patients during the COVID-19 pandemic.

According to the scope of COVID-19 disease and considering that our country is one of the regions with conflicting statistics, it is essential for the surgeonto consider optimal use of facilities with priority to treat the associated diseases and performing necessary interventions during the outbreak. To maintain the safety of colleagues, patients and their companions, developing and following precise instructions for surgical interventions during the COVID-19 pandemic is urgently needed.

Materials and Methods

The current study is a multi-center, prospective, brief report of pediatric patients with SARS-CoV-2 infection, who had surgery at 6 hospitals in 4 cities of Khuzestan province, Southwestern Iran, one of the provinces with the highest prevalence and death due to COVID-19 disease. The study was approved by Ahvaz Jundishapur University of Medical Sciences (AJUMS) independent ethics committee or institutional review board (IRB), and all participants and their parent(s) or legal guardian signed the informed consent prior to participation.

Children with SARS-CoV-2 infectiondiagnosed within 7 days before or 30 days after surgery, undergoing any procedure performed by a surgeon under general, regional, or local anesthesiain an operating theater, were included.

COVID-19 diagnosis was confirmed using reverse transcription polymerase chain reaction (RT-PCR) testing of nasal swabs or broncho-alveolar lavage, followed by diagnosis of lesions through computed tomography (CT) scan of the chest according to the national guidelines.[5,6]

Results

30-day follow-up had been reached for 246 pediatric patients who had surgery between Jan 20 and Jun 01, 2020. Four (1.62%) of 246 patients who had undergone surgery were COVID-19 infected **Table 1.**

Table1: Baseline and demographic characteristics, comorbities, diagnostic and clinical data, and operation details

Patients	Case 1	Case2	Case3	Case4
Birth Place	Mahshahr	Sosangerd	Abadan	Ahvaz
Age (years)	12	8	9	6
Sex	Male	Female	Female	Male
Symptoms	Dyspnea	Dyspnea and Fever	Cough	Diarrhea and Fever
Comorbidities	Negative	CHF	ESRD	Diabetes
COVID-19 in Family	Negative	Negative	Positive	Negative
PCR	Positive	Positive	Positive	Positive
WBC($\times10^9$/L)	1500	4800	3200	10600
Lymph($\times10^9$/L)	6.4%	8.6%	7.5%	4.7%
Hb(g/dL)	10.8	11.3	9.6	9.3
PLT(10^3)	746	443	321	452
D-dimer	346	395	215	179
CT scan report	Right ground glass opacity	Bilateral ground glass opacity	Left ground glass opacity	Left ground glass opacity
CRP	2+	3+	1+	1+
Procedure	Total gastrectomy and Esophagojejunostomy	Bilateral pleural effusion tap	Catheter placement for dialysis	CV-line placement
Outcome	**Discharge**	**Discharge**	**Discharge**	**Discharge**

CHF, Congestive heart failure; ESRD, End-stage renal disease; PCR, Polymerase chain reaction.

Discussion

Based on the results of our study, 1.6% of the pediatric patients undergoing any surgical procedure were reported COVID-19-positive in terms of symptoms, PCR and chest CT scan. Considering that this study was conducted at the peak of COVID prevalence in Iran, the results are promising.

All the four COVID-19-positive patients had underlying chronic diseases and also one of them underwent a major surgical procedure which was total gastrectomy. Eventually, all the 4 patients were discharged safely. So, despite the patients' underlying chronic diseases and different kinds of surgical procedures performed, these patients were able to overcome the infection and discharged from the hospital. Based on our results, it can be said that the prevalence of this disease in children is lower than the average of the society and also its outcomes look good in young ages. The equal prevalence of COVID-19 infection according to gender in this study is also considerable.

Ethical Consideration

This article was approved as a research project on the 31/10/2020 by the Ethics Committee of Ahvaz Jundishapur University of Medical Sciences.

Acknowledgment

Not applicable

Funding/Support

Not applicable

Conflict of interests

There is no conflict of interest

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Death Due to Late Onset Diaphragmatic Hernia

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How to cite this article:
Dandeniya Arachchige Harshani S, Ruwanpura PR. Death Due to Late Onset Diaphragmatic Hernia.Iranian Journal of Pediatric Surgery 2021; 7(2): 126 – 132.

DOI: https://doi.org/10.22037/irjps.v7i2.27473

Abstract

Diaphragmatic hernia is usually congenital, nevertheless it can also be acquired, particularly following trauma. Most of late onset hernias are acquired and reported cases of congenital type are rare. This case explicates a late onset congenital diaphragmatic hernia (CDH) where the diagnosis was made at the time of autopsy. Autopsy revealed the stomach and the proximal small intestines to be present in the left hemi thorax through a 4.5cm X 5cm defect in left hemi-diaphragm. Left lung was found to be collapsed and 60 g in weight. Histology revealed pulmonary hypoplasia. The Cause of Death was declared as CDH complicated with lung hypoplasia.

Keywords

- Diaphragmatic hernia
- Congenital defect
- Lung atelectasis

Introduction

In 1679, Riverius registered the first noted case of a congenital diaphragmatic hernia (CDH). It was diagnosed after death of a 24-year-old man.[1] The first effort to operate on a CDH was by Nauman in 1888. Laparotomy was accomplished in a 19-year-old man with acute respiratory distress and acute abdomen. In 1889, O'Dwyer attempted the first repair of CDH during infancy.

Diaphragmatic hernia is a congenital defect of the diaphragm which results

received: 21 July 2020
accepted: 10 October 2020
Published online: November 2021

from incomplete closure of the pleuroperitoneal canal or septum transversum during fetal development. Four to ten percent of all infant deaths are due to congenital anomalies caused by congenital diaphragmatic hernia (CDH).[1] It is usually diagnosed immediately after birth or within the neonatal period [2,3] with the presence of severe respiratory distress. The term 'Late onset CDH' refers when the symptoms appear after the neonatal period and they account for 10%-13% of all CDH cases.[4, 5]

This case explicates where the diagnosis was made at the autopsy of an eleven-year-old boy.

Case Presentation

The deceased patient was an 11-year-old boy who was living in a children's home since two months of age. According to the matron's statement he was not diagnosed to have any major medical ailment, except for intermittent mild to moderate respiratory infections for one year. He has had complained of abdominal pain and nausea for 3 days and he had been taken twice to a General Practitioner. He was treated for gastritis and gastroenteritis on both occasions. Despite the treatment given, the symptoms did not subside completely. He developed vomiting and diarrhea night before his death.

However, he had fallen asleep after dinner, after having two biscuits as the discomfort subsided to a certain extent with vomiting. He was found unresponsive on the following morning and the body was cold at that time. The matron had noticed the presence of vomitus around his pillow and on the floor. He was immediately brought to the hospital. He was pronounced dead on admission at the outpatient department. The patient did not give any history of blunt or penetrating trauma to chest or abdomen. An inquest was ordered. A post mortem autopsy was ordered by the inquirer into sudden deaths. The autopsy was done on 17th April 2017, after about six hours of confirmation of death.

Autopsy Findings

The body was that of a slim child with a brown skin complexion. There were no signs of any dysmorphic features or congenital abnormalities. There were no external injuries. On opening the thoracic cavity, it was noted, that the stomach and the proximal part of the small intestines to be present in the left hemithorax **Figure 1**. On further dissection a 4.5X5cm defect was noted in the posterior part of the left hemi-diaphragm **Figure 2**. The stomach contained whitish undigested rice meal. Bowels did not show any necrosis, malrotation or malfixation throughout its entire length.

Left lung was found to be collapsed **Figure 3** and was 60 g in weight. The right lung was normal in size and weighing 110g. Features of pulmonary hypertension were not seen. Histological examination of the lung revealed pulmonary hypoplasia of the left lung whereas all other organs showed normal histology. Cause of death was given as late onset diaphragmatic hernia, complicated with lung hyperplasia.

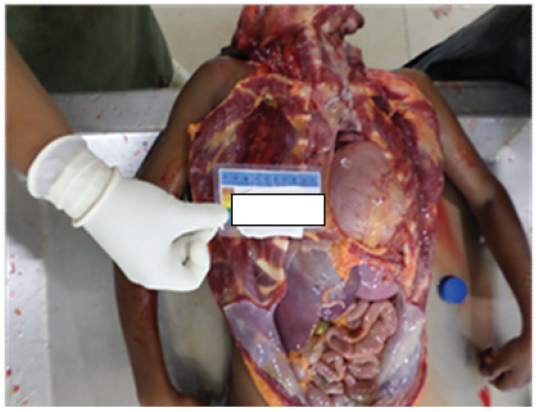

Figure 1: Position of the organs in the chest and abdominal cavity

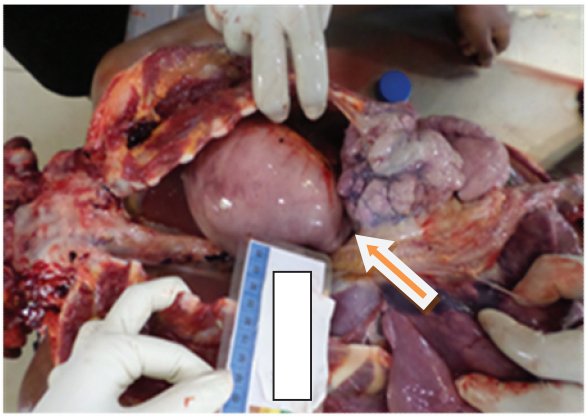

Figure 2: Defect in the diaphragm

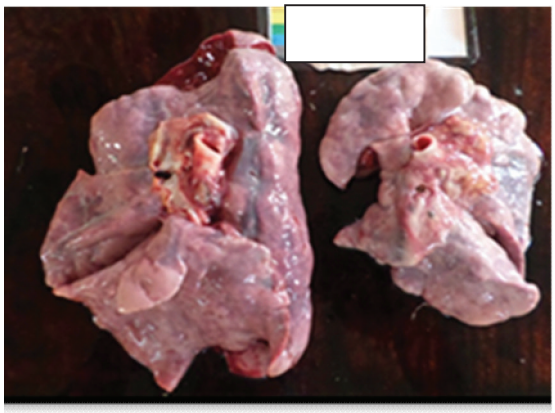

Figure 3: Normal right lung with hypoplastic left lung

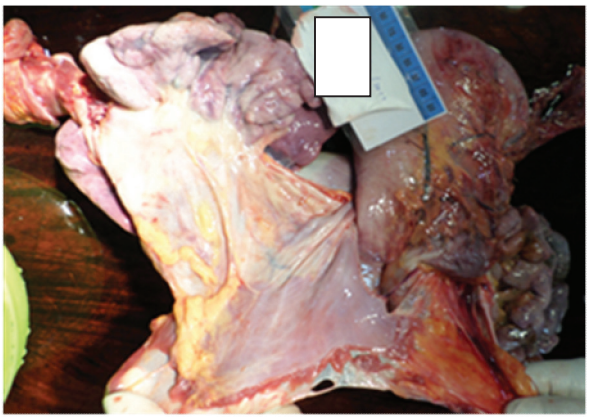

Figure 4: Hernation through the diaphragm

Discussion

Diaphragmatic hernia is usually congenital. Nevertheless, it can be of acquired type, particularly following trauma. Most of the time, late onset hernia are acquired in nature [6,7,8] which occurs following blunt trauma as it happens in falls or blows, or penetrating injuries (stab or firearm injuries). Rarely, it could be an iatrogenic due to an inadvertent injury during surgery.[9]

In this case the patient happened to be a previously healthy child. There was a history of only intermittent attack of chest infections. A plain radiograph which had taken four years ago is said to be apparently normal. There was no history of trauma or any surgery. Postmortem features showed herniation of bowels into the left hemi thorax through a diaphragmatic defect with pulmonary

hypoplasia. This indicates a long-standing problem rather than an acute event. Recurrent lung infections as given in the history also indicate a chronic event. It is probable that interference with the lung movements and function particularly on the left side could have been the reason for these recurrent episodes of lung infections. In this case the history together with the post-mortem features indicates a diagnosis of congenital diaphragmatic hernia excluding acquired defect of the diaphragm.CDH occurs in about 1 in 3300 live births[10] and it is usually diagnosed before or just after birth. Prenatal diagnosis of CDH is possible in majority of cases. Earlier, 80% of patients with CDH used to die in their neonatal period in spite of optimal treatment.[11] Recent studies have shown that there is an immense impact on the outcome of the child with CDH with advent of prenatal diagnosis and successful treatment has reached to 80 %.[12] Recurrence rate of CDH is 2% in subsequent pregnancies on a rough estimate.[12]

Three types of CDH are identified. 'Bochdalek' Hernia is the most common type. The defect is confined to the posterior or lateral aspect of the diaphragm. It is more common in left than right side of the diaphragm and carries a high rate of mortality. 'Morgagni' Hernia is rare, and the defect of the diaphragm is located in the anterior part of the diaphragm. 'Diaphragm event ration' the third type is used to describe when there is elevation in an intact diaphragm. In this patient the defect was found in the posterior part of the left dome of the diaphragm which goes along with the classical picture of 'Bochdalek' Hernia.

Incomplete closure of the diaphragm during fetal development, herniation of abdominal viscera to the chest and lung hypoplasia are the three main pathologies which are identified in CDH. If hernia formation precedes the development of lung, pulmonary hypoplasia sometimes occurs with severe respiratory symptoms at birth. In adulthood, it is not common, the development of lungs in most instances is normal and therefore symptoms are rare. [13] In this case histology revealed lung hypoplasia which indicates that the herniation of abdominal contents could have been there since birth. It is difficult to explain the absence of any significant symptoms and signs for such a long period. Both Lurie and Enns et al. have published remarkable papers of chromosomal anomalies in association with CDH.[14, 15] There are many other congenital chromosomal abnormalities believed to be associated with pathogenesis of congenital hernia i.e., genes COUP-TFII, FOG2, GATA4, WT1, and SLIT3.[16]

In the older age groups there are two clinical scenarios are common: an incidental finding on plain radiographs performed for reasons other than the hernia [17] or when symptoms develop due to incarceration, strangulation and visceral rupture inside the chest cavity. In this case neither of them was present but in author's opinion the history is a little unreliable as he was inmate of a children's home and possibly absence of close supervision could have missed certain features of

CDH. The diagnosis is made by a simple chest radiograph, computerized axial tomography or an MRI. Although it was revealed that an X-Ray was taken four years prior to his death, it was not available for observation. This could have been an important piece of evidence to come to a conclusion whether this to be an acute or chronic event.

In this case most of the features present were common with some other pathologies like gastritis and gastroenteritis. In a case of late presenting type there is usually a lesser degree of herniation of small part of bowel since birth, which could be misdiagnosed due to non-specific symptoms of vague nature.

It is said that the presentation, management and prognosis are somewhat different in congenital and acquired forms of diaphragmatic hernia. In neonatal type, presentation is mainly with the respiratory distress compared to late onset CDH. In the latter type those respiratory symptoms together with recurrent respiratory infections are common mainly with right sided CDH. Left sided CDH are mainly presented with gastrointestinal symptoms which matches the findings in this case. The deceased here also had abdominal pain, vomiting and loose stools which again mimics the common clinical picture. According to the history he had not had any significant history of those symptoms before.

Prognosis of late onset CDH is excellent with early diagnosis and surgical repair than the neonatal type, but the morbidity is high. [2, 3] Another important fact which has direct impact on the prognosis is the association of other malformations with late onset diaphragmatic hernia. Main co-existing anomalies identified are gut malrotation or malfixation, pulmonary hypoplasia, pulmonary sequestration, umbilical hernia, atrial septal defect, ventricular septal defect, polysplenia, type 1 diabetes mellitus. Gut malrotation and the pulmonary hypoplasia are the most two common associations.[2] However, 50 to 60 percent of cases of congenital diaphragmatic hernia are isolated, which means that patients have no other major anomalies. In this case, the presence of pulmonary hypoplasia was confirmed by the histology.

Conclusion

Congenital diaphragmatic hernia is relatively uncommon in the older age group. Severe defects usually present soon after birth. Nevertheless, less severe ones can present later as in this case. As children find it difficult to explain their problems, it might lead to missed or wrong diagnoses in some instances. The insight to this anatomic defect is crucial to identify and manage patients of older age groups, as it should be surgically corrected to avoid complications or to deal with them if they are already present.

This case highlights the importance of higher clinical vigilance when an early teenage patient presents with the vague gastro-intestinal and respiratory symptoms as timely medical intervention could save his life.

Ethical Considerations

The material contained in this article refers to coronial autopsy report which is publicly available according to Srilankan law. However, authors have observed anonymity and other general guidelines biomedical research in preparation of the paper.

Acknowledgment

Not Applicable

Funding/Support

Not Applicable

Conflict of interests

There are no conflicts of interest.

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Simultaneous Antegrade and Retrograde Intussusceptions in a Child: A Rare Condition and Literature Review

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How to cite this article:

Yaokreh JB, Moufidath SS, Thomas HA, Kouamé YGS, Kouamé BD, Ouattara O. Simultaneous Antegrade and Retrograde Intussusceptions in a Child: A Rare Condition and Literature Review. Iranian Journal of Pediatric Surgery 2021; 7(2): 133 - 138 .

DOI: https://doi.org/10.22037/irjps.v7i2.29583

Abstract

Intussusception is one of the major abdominal emergencies in children under two years of age. Double intussusceptions are rare, and the simultaneous ante grade and retrograde occurrence is even rarer. Fewer than 10 such cases have been reported in the pediatric population.

An 8-month-old girl was presented to a peripheral health center with paroxysmal abdominal pain for two days, as well as postprandial vomiting and four mucous stools. She was diagnosed with amoebic dysentery. Faced with the appearance of glairy-bloody stools, she was admitted to our pediatric emergency unit. After examination and abdominal ultrasound, the diagnosis of intussusceptions was made. On the fourth day, operative findings showed a double ante grade and retrograde pattern. A manual reduction followed by an appendectomy was performed. The postoperative course was uneventful.

received: 19 March 2020
accepted: 9 August 2020
Published online: November 2021

Keywords

- Children
- Double intussusceptions
- Retrograde intussusceptions

Continuing education of peripheral practitioners will allow early diagnosis and appropriate treatment as this misdiagnosis could have compromised the vascular reserve of the bowel, resulting in intestinal ischemia and possibly perforation.

Introduction

Intussusception is defined as the telescoping of a segment of the bowel into an adjacent point. It is considered among the most common causes of acute abdominal pain in children less than two years of age.[1] Double intussusception is rare, and fewer than 20 cases have been reported in the English literature.[1,2] However, simultaneous antegrade and retrograde intussusception (SARI) is rarer in children, and most of these are reported in adult patients with a pathological lead point.[2,3] To our knowledge, this is the seventh case of SARI in children.[2,4-6] Here, we are reporting a first case of SARI in an Ivorian child because of the rarity of this condition. We will then discuss the difficulty of preoperative diagnosis and review the literature.

Case Presentation

An eight-month-old girl with no specific medical history was presented with paroxysmic abdominal pain for two days. She had six episodes of postprandial vomiting and four mucous stools with fever. The diagnosis of amoebic dysentery was made at the local hospital and the treatment consisted of metronidazole®, and paracetamol® and an oral dehydration solution. Faced with the appearance of glairy-bloody stools on the second day, her mother came to our emergency room. On admission, the infant was conscious and reactive. Her temperature was 38.2°C and she weighed 9kg. She was moderately dehydrated. The abdomen was breathing well, it was soft, compressible and painless. Abdominal palpation showed the mass in the umbilical quadrant. Digital rectal examination showed an empty rectum, and the finger of the glove was stained with blood. Cardiovascular and neurological examination was normal. The diagnosis of acute intestinal intussusception was suspected and confirmed by abdominal ultrasound **Figure 1**. Laboratory results show a hemoglobin level of 10.4g/dL and a prothrombin level of 97%. The blood ionogram was moderately disturbed. She was scheduled for emergency laparotomy. After the hydro-electrolyte rebalancing, a transverse laparotomy under the umbilical tissue was performed the next day. The operative findings revealed an antegrade ileocolic boudin through the ascending colon with multiple mesenteric adenopathies. During exploration, a second retrograde colocolic intussusception **Figure 2** was noted on the

transverse colon about 15 cm downstream of the first. Manual reduction was performed for both boudins, and the bowel appeared to be well perfused. There was no pathological lead point. An additional appendectomy was performed. The postoperative outcome was uneventful, and she was discharged home on the sixth postoperative day. Ten months later, there were no adverse events in the follow-up.

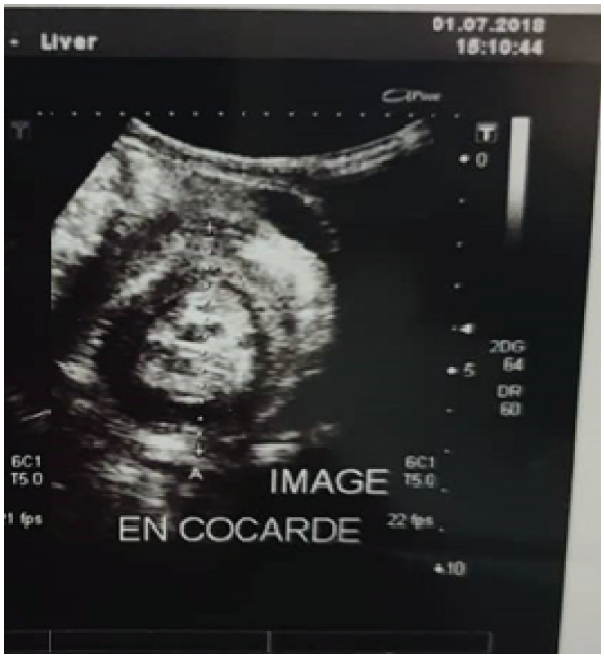

Figure 1. Abdominal ultrasonography show the classic target sign

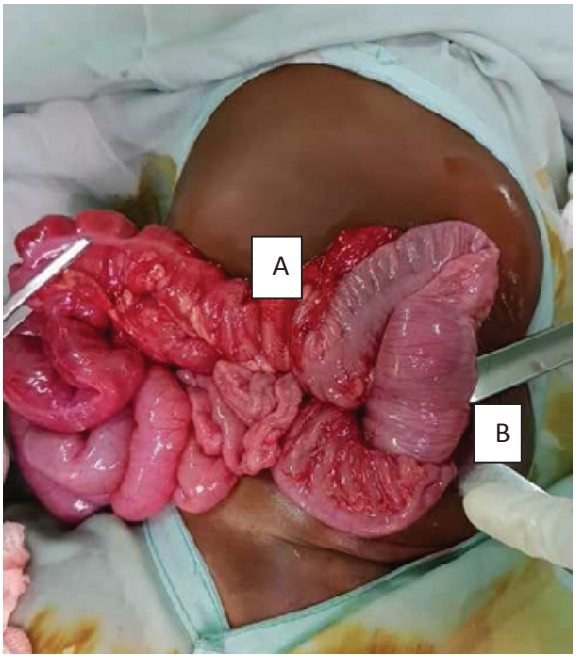

Figure 2. Operative findings shows double antegrade (A) and retrograde (B)

Discussion

Intussusception is one of the most common causes of bowel obstruction in children under 2 years of age, and antegrade ileocolic intussusception is the most common pattern.[5,7,8] Few cases of double intussusception have been reported. Among cases of double intussusception, SARI is a rare condition in children **Table 1**. A review of the literature over the past 30 years has identified fewer than 10 cases of SARI.[2,4-6,9] Our case was certainly the seventh case of SARI. This entity involving two distinct points in the bowel differs from compound intussusception which has been defined as double, triple or quadruple superimposed intussusception occurring as a single mass.[3-5,10] The term "retrograde intussusception" describes the intussusception of an intussusceptum in a proximal direction, but the exact mechanism of this condition remains unknown. Most retrograde forms are diagnosed in adults with a pathological lead point,[11] and the preferred site was the sigmoid colon. To date, apart from the known causes, the explanation of idiopathic retrograde forms is based only

on theories. The most common advanced theory was initiation by antiperistaltic waves in the left colon. In our case, the site of retrograde intussusception was the transverse colon which was free without attachment. We are unable to give an explanation for this condition.

Table 1. Reported cases of simultaneous antegrade and retrograde intussusception in children

Authors	Age / Gender	Type	Cause	Treatment
Shekhawat [7]	7 months ; M	Ileocolic and retrograde colocolic	Idiopathic	Manual reduction
Arnold [5]	5 months ; F	Double compound: ileocolic and retrograde sigmoido-colic	Idiopathic	Manual reduction
Lukong [6]	10 months ; M	Ileocolonic and retrograde colocolonic	idiopathic	Manual reduction
Egbuchulem [8]	11 months ; M	Double compound: ileocolic and retrograde colocolic	Idiopathic	Resection and anastomosis
Seyi-Olajide [3]	11 months ; M	Ileocolic and retrograde colocolic	Idiopathic	Manual reduction
Randimbinirina [9]	11 mois ; F	Ileocecocolic and retrograde sigmoidocolic	Idiopathic	Manual reduction
Our case	8 months ; F	Ileocolic and retrograde colocolic	Idiopathic	Manual reduction

Most authors have reported difficulties in the preoperative diagnosis of double intussusception. The clinical features in our case do not differ from those of classical intussusception (paroxysmal abdominal pain, palpable abdominal mass and redcurrant jelly stools). In our practice, amoebic dysentery is the most common diagnosis reported by most practitioners in peripheral health centers. This misdiagnosis could compromise the vascular supply of the bowel, resulting in intestinal ischemia and possibly perforation. In most cases of double intussusception such as ours, a single mass was palpated on abdominal examination, and ultrasound revealed the classic target sign. The second intussusception is not detected, either because there is too much gas distension or because the sonographer convinces himself of the diagnosis by finding the prominent intussusception and not looking for the others. None of the reported cases had two palpable masses on abdominal palpation, and less than half of them were diagnosed preoperatively by ultrasound or CT scan showing two target signs or a triple circle.[1,12-14]

To date, there is no medical treatment available for the preoperative diagnosis of double or retrograde invagination. The failure of the enema is undeniable. We

have systematically performed a laparotomy. On the other hand, laparoscopy was performed.[4] This approach limits parietal morbidity and the length of hospitalization. The operative results indicate a second intussusception with no pathological lead point. None of the previously reported SARIs had a pathological lead point.[1,6,9] Faced with the possibility of a double or compound or multiple intussusceptions, we agreed to always perform a careful evaluation of the entire bowel in order to identify the multiple distant segments that could be involved.

Conclusion

Simultaneous anterograde and retrograde intussusception is a rare entity in the pediatric population. Because of the potential for this occurrence, the radiologist should be referred by indicating this on the ultrasound bulletin. Coexistence of double or multiple intussusceptions requires careful examination of the entire bowel peroperatively.

Ethical Considerations

Written informed consent was obtained from the parents for publication of this case report and any accompanying images.

Acknowledgment

Not Applicable

Funding/Support

Not Applicable

Conflict of interests

There are no conflicts of interest.

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Lip Commissure Reconstruction with the Facial Artery Muscular-Mucosal (FAMM) Flap: A Case Report of a Child with Electrical Commissural Injury

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How to cite this article:
Yousefi J, Tabrizian Namini F, Raisolsadat SMA. Lip Commissure Reconstruction with the Facial Artery Muscular-Mucosal (FAMM) Flap: A Case Report of a Child with Electrical Commissural Injury. Iranian Journal of Pediatric Surgery 2021; 7(2):139-145.

DOI: https://doi.org/10.22037/irjps.v7i2.27876

Abstract

Electrical burn damages constitutes of 4% of all burn trauma. Approximately 20% of total electrical burn injuries occur in children. Most electrical injuries in children occur at home. When the child bites or chews an electrical cord, it results in oral and commissural injury. It is also noted that male children are predominantly affected with electric current injuries than female children. We introduce an immigrant 3-year-old boy admitted in the hospital one week after an electrical injury of the oral cavity due to biting a television cord. He had a necrotic lesion on his right oral commissure. There is controversy about the time and relevant management of mouth commissure injury. In this case, because of lack of parents' consent, we did the reconstruction of his oral commissure very late without any molding intervention.

received: 15 December 2020
accepted: 9 March 2021
Published online: November 2021

Keywords

- Electric injury
- Mouth and lip
- Flap repair

There is more than 200 techniques in the management of lip and oral commissural injury; we used a Facial Artery Mucosal-Muscular (FAMM) flap for the repair with good result. Although with the improvement of burn care and its management outcomes, prevention is still the best way of minimizing the prevalence and severity of electric current burn injuries.

Introduction

Most electrical burns occur before four years old, and injury to oral commissures, lips, mouth, and tongue occur disproportionately in injured kids presenting with electrical traumas. The oral cavity is the most common place of damage, and most patients are under4 years of age (93%). There are two phenomena that usually explain the mechanism of electrical trauma: the electrical arc and the contact injury.[1] There are several classification systems for the commissural burn but none of them gained widespread acceptance.

Surgical reconstruction interventions sometimes are needed to restore normal structures, functions, and aesthetics of injured parts. There is controversy about the regulations and relevant management of electrical trauma to the mouth commissure. Interventions can be categorized based on the time that has been passed: primary management; which is done within several days of trauma, Intermediate management; which is done when the margin of necrotic tissue is distinguished from normal appearing part (usually 1-4 weeks), and postponement technique; when the entire of trauma site is healed (usually after a few months).[2] The favorite management plan depends on the gap between the initial injury and the final repair, amount, and harshness of trauma. It is also based on the surgeon's choice.[2] The Facial Artery Muscular-Mucosal (FAMM) flap, is a composite flap that is harvested from lateral inner buccal muscular-mucosal tissue. However, the material of this flap is not similar to the oral commissure, but it has some features that would be a suitable substitution for mouth commissure restoration.[3] Herein, we report an electrical commissure burning case, involving the reconstruction of the labial commissure using a FAMM flap that produced favorable outcomes.

Case Presentation

A three-year-old immigrant boy was admitted to our hospital (22 Bahman Hospital, Mashhad, Iran) one week after an electrical trauma to the mouth commissure due to biting a television cord.

At first the boy was examined and managed in the accident department of another hospital abroad and then his

parents transferred him to our hospital for more assessment. The child was active, alert, and in no distress, with severe inflammation and necrosis of right-side labial commissure when seen **Figure 1**. His parents claimed when the child has been playing in home, he suddenly screamed and had fallen near a television without any convulsion or loss of consciousness. After that, a pale lesion appears on the right side of his lips angle and redness of his right cheek. Clinical examination revealed a more than 2 cm circular, severe inflammation and necrosis of right side of labial commissure. His teeth were intact with normal occlusion. The parents and first physician have not found any bleeding from the injury site.

In the former department, the boy had taken normal saline for mouth wash along with tetracycline ointment and cephalexin syrup. When we saw him, we performed minimal debridement and then referred him to the dentistry clinic for oral molding. Unfortunately, he left the country without oral molding. He came back 4 months later with severe drooling, commissural contracture, and swallowing problems, then we used a Facial Artery Muscular-Mucosal (FAMM) Flap for repair. We designed a 20 mm (width) × 30-35 mm (length) Musculo-mucosal flap in the right cheek which had an inferior pedicle, where the facial artery consists of its base **Figure 2, 3**. After the incising the mucosa on the marking of the flap, a further dissection was made in the buccinators muscle to achieve free movement of the flap. The flap was included a branch of the facial artery deep inside the muscle. The flap was rotated and sutured to lip commissural defect. To obtain consistency and movement of the flap, the muscular part of the flap was sutured to the upper orbicularis oris muscles. The donation site of the flap was closed primarily. Unfortunately, the patient and his family left the country three days after surgery. Three weeks later, the hospital social worker contacted the family with the phone call and reported that he had a good condition without swallowing problems **Figure 4**.

Figure 1: severe inflammation and necrosis at first visit

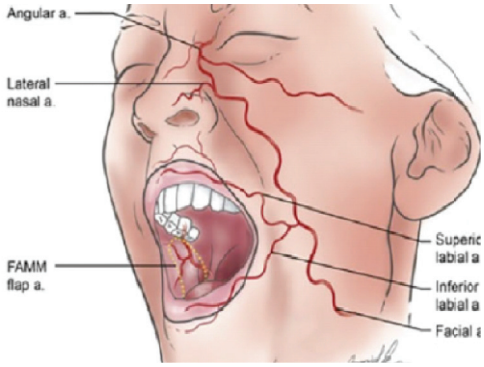

Figure 2: vascular anatomy of FAMM flap

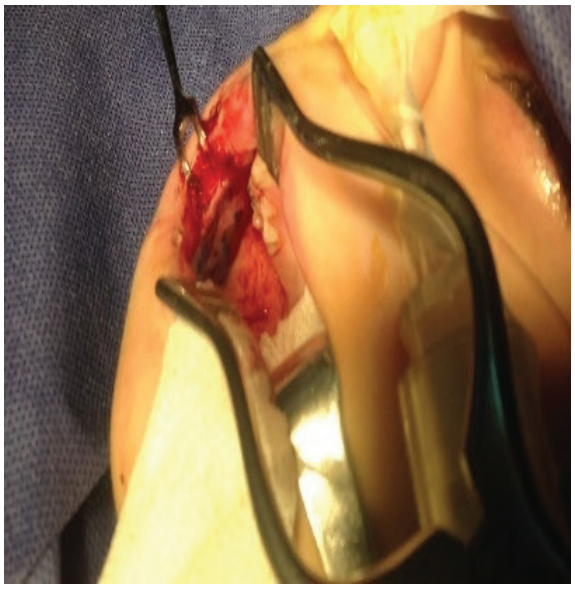

Figure 3: An inferior-based FAMM flap marking on right facial artery and vein

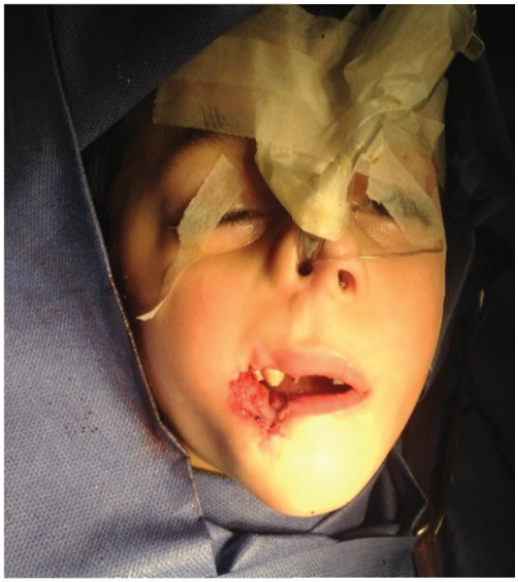

Figure 4: After flap repair

Discussion

Lips, mouth, and mouth angles are places that electrical injury falls out due to an electrical arc mechanism. Initially, these injuries happen in mucosa due to wetness by saliva and low resistant mucosal membranes. These traumas usually occur because of biting or sucking a power cord by infants.[1] Labial commissure destruction causes difficulty in swallowing and drooling, which results not only in interruption of lips integrity and swallowing function and decrease in quality of life, but also damages aesthetics.[4, 5] It also has many negative effects on self-confidence and psychological status.[6, 7]

The objective of oral commissure restoration is to build up normal structures to their normal positions and create a thin, mobile lip part that moves dynamically and uniformly with facial mimics.[1] Lips and commissure defects restoration is tuft. Because they are complicated structures, and consist of several parts[3], more than 200 different methods have been proposed by various scientists to reconstruct this area for many purposes like cancers, vascular anomalies, and trauma.

"Schulten (1894) used a double-pedicle flap and Lexer described a tongue flap in 1909. Friedlander Spira, Stal, Kolhe and Leonard described other types of local advancement flaps. A cross-lip mucosal flap was described by Mazzola and Lupo in 1984 for superficial resurfacing of the vermilion and then again by Standoli in 1994. This flap borrows labial mucosa from the inner surface of one lip to resurface the vermilion of the other lip. Sakai et al in 1988 described an interesting technique, and then Iwahira et al again described in 1998, which involved the bilateral island vermilion flap or the sliding-door flap. Uni-pedicle and bi-pedicle cross-lip vermilion flaps were described by Kawamoto in 1979 and Lew et al in 1987".[1, 8]

Reconstruction of the oral commissure with Estlander flap, is the most common method used in oral defects. The Fries technique suites for horizontally oriented defects. Platz and Wepner method, the Brusati method, and Zisser flap are other techniques to reconstruct oral commissure.[2, 9, 10]

In commissural restoration procedures, it is best to use identical or the same tissues, which results in favorite function outcomes in texture, and color.[5]Pribaz et al [11] in 1992, introduced the FAMM flap, that was somehow different from the mucosal buccinators or buccal flaps indirection and vascular supply. The FAMM flap consists of mucosa, submucosa, and a small amount of buccinator muscle, the deeper plane of the Orbicularis Oris muscle, and the facial artery and venous plexus. One of the prominent advantages of this flap is being completed in one stage, as compared with tongue flaps and cross-lip flaps, which require a next procedure for stalk excision and inset of the flap. Since then, new modifications were developed with subsequent publications, and the FAMM flap became more versatile for use.[3, 12]

Varied algorithms depend on the range of the defect have been explained.[5]Hence, we selected the FAMM flap, because of the features and advantages of FAMM flap over the other methods. Reliability, ease for harvest, proximity to the defect, and the same mucosal coverage, absence of external scar, and lower rate of significant complications are the advantages of this flap. Therefore, this flap is an optimal donor site for labial commissure reconstruction.[3,13] In addition to local flaps, mucosal-free grafts have also been described by Ahuja[14] that used labia minor grafts for vermilion reconstruction. However, we used FAMM flap in this patient, and both aesthetically and functionally, satisfactory outcomes have been achieved successfully three weeks after the procedure.

Conclusion

Oral commissure electrical burns management in infants and children remains controversial. In most circumstances, a postponed repair is accepted to give time for the initial wound being healed by itself. The use of oral commissure splinting, although challenging in practice, has been shown to help prevent the need for surgical reconstruction of the oral commissure in the future. If operational restoration is inevitable, the objectives include both normal function and cosmesis. Different procedures have been recommended for the reconstruction of the oral commissure; each has particular advantages and disadvantages, and FAMM flap is the one we found to have some good results.

Ethical Considerations

This study was approved by Islamic Azad University-Mashhad Branch with code number IR.IAU.MSHD.REC.1399.184. Written informed consent was obtained for operation.

Acknowledgment

Not Applicable

Funding/Support

Not Applicable

Conflict of interests

There are no conflicts of interest.

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