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A TRIBUTE



Dr. Banamali Nath

Dr. Banamali Nath, an alumnus of Gauhati Medical College was born at Goalpara on 15.07.51. After starting his schooling from primary level at Goalpara, he passed HSSLC in 1967 from P R H S School in the same town and then joined Gauhati Medical College from where he graduated. He later on obtained his MS in General Surgery from his Alma Mater Gauhati Medical College in 1978. A brilliant student, Banamali Nath, also obtained MNAMS degree in 1979 and later on got his Fellowship (FAIS) of Association of Surgeons of India too.

After his post graduation, Dr Nath joined State Health Service at Morigaon Civil Hospital in 1979 and retired from the same hospital as SDMO in 2009. After retirement, he joined as Senior Consultant Surgeon at Catholic Hospital, Borgang in Sonitpur district and after serving there from 2010 to 2011, he served as Assistant Professor of Surgery in, Manipal College of Medical Science, Pokhara (Nepal) from 2011 to 2014 and lastly as Assistant Professor of Surgery at RD Gardi Medical College, Ujjain from 2014 till he breathed his last.

Dr. Nath was an academician, active member of ASI & ASA and presented several study papers in state in national level conferences and published several papers in Association's journals. He was also recipient of the prestigious Dr. Arthur D'esa Travelling Fellowship in 1994, Ethicon Travelling Fellowship Award Rural Surgery, ASI, Chennai, in 2002, & Ethicon Visiting Professorship Award ASI, Ahmedabad, 2013.

He was also recommended for Padmashree award by the Ministry of Home Affairs.

Following a massive stroke on 9th July, 2016 at the same Medical college where he was working at Ujjain (M P) he breathed his last. He is survived by his wife Dr Santana, son Chiranjeeb and daughter Jayashree.

We pay our respectful homage to him.

Editorial

Thoughts for the 'days'.

Celebrations are part of social activities. Commemorative days for health awareness are celebrated all over the world. July first happen to be 'Doctor's Day' in India. On this day, one of the worthy sons of India, Bharat Ratna Dr. B C Roy was born. This day is an opportunity to refresh our commitment as a member of the noble medical profession for the roles & responsibilities that we undertake in the society. This is also a great opportunity of medical professionals to get closure to the society. In recent times there have been severe trust deficit between the doctors and the society. An introspection from both sides to address these misgivings is the need of the time. It is important if we have to think of improving the dismal health care delivery system in our country in general and Assam in particular. An environment of trust and goodwill among the various stakeholders will help in creating an ideal atmosphere so that the best possible health care could be provided to the people. Proposed National Medical Commission Bill to be brought in shortly has created a sense of anxiety and uncertainty among the medical professionals. A regulatory body needs to be independent one with sufficient number of representatives from concerned disciplines as well as representative from the civil society to bring in credibility and transparency to the body.

World Population Day is celebrated on 11th July all over the world. It is celebrated to raise awareness among public regarding some important and urgent issues related to increasing world population. When global population crossed five billion the UNDP in 1989 started this to pay attention to reproductive health and family planning. Population explosion is a major issue particularly in the developing world. In Indian perspective, this problem is going out of hand day by day. This country with 2.4% of world land mass is home to 1.23 billion people. The present situation was anticipated way back in 1952. It became the first country in the world to accept family planning program as a national policy. Lack of education especially of women, superstitions, early marriages, preference for male child are few contributing factors. Males are sole decision maker



in a family. He is always reluctant to accept any kind of contraceptive methods many a times due to lack of awareness. Male reproductive service is neglected in India. Male participation in family planning program has to be increased for its successful implementation. Medical professionals should use their resources to make awareness about population explosion amongst the masses.

The environment is directly affected by the people inhabiting it. To make people aware of green and healthy environment, the UN observes World Environment Day on 5th June since 1973. Medical profession has tremendous obligation in Protection of the mother earth and the environment. Reduction of population pressure has direct positive impact on it. As responsible member of the society, medical professionals' whole hearted participation in all plan and program of family planning is warranted for the better future of mankind.

K. Bhuyan
Guest Editor



Original Article

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Stentless pyeloplasty in children: experience of 68 pyeloplasties in 60 patients

ABSTRACT

Objective : Open Anderson-Hyne's pyeloplasty is the standard procedure for pelvi-ureteric junction obstruction in children but few surgeons do this operation by laparoscopy also. A trans-anastomotic stent is generally used by both the groups. Aim of present study is to evaluate the safety and efficacy of pediatric pyeloplasty without a stent.

Materials and methods: From March, 2009 to Feb, 2015, sixty-eight pyeloplasties in 60 consecutive patients were performed without a stent. Kidney was explored through an anterior extraperitoneal flank approach. Anderson-Hyne's pyeloplasty was done in all cases and anastomosis was done by 6-0 polyglycactin. A modified nephrostomy was done in initial 20 cases where pelvic reduction was done and a no.10 infant feeding tube was placed below the operated kidney as drain in all the cases. Nephrostomy clamping was started after 48 hours and nephrostomy tube was removed after 7 days. Nephrostogram was not routinely done. Follow up ultrasonography was done at 3 months and 6 months and DTPA scan at 6 months.

Result: Age of the patients ranged from 2 months to 9 years. There were 38 male and 22 female patients. Left side was affected in 31 cases and right side in 19 cases; 8 patients had bilateral hydronephrosis. Nephrostomy was done in 20 cases and nephrostogram was done in initial 16 cases and it showed flow of contrast along the ureter. Follow up ultrasonography showed residual dilated collecting system but the DTPA scan showed improvement of function and relieve of obstruction.

Conclusion: Avoidance of stent did not adversely affect the results; rather it eliminated the possible complications related to stent. It also eliminated the cost of the stent and another operating session for removal of an internalized stent. We conclude that pediatric pyeloplasties can safely be done without a stent.

Key Words : Hydronephrosis; pelvi-ureteric junction obstruction; pediatric pyeloplasty; stent.

Introduction :

Pyeloplasty is the most common procedure performed in Pediatric Urology with minimum complication rates [1]. There are many operations and approaches available for pediatric pyeloplasty. But open pyeloplasty is the standard procedure and Anderson-Hyne's pyeloplasty is the most commonly employed operation. Main aim of the operation is to achieve free drainage of urine from the renal pelvis into the ureter. Differences of opinion exist regarding use of a nephrostomy and trans-anastomotic stent. Authors performed 68 routine pyeloplasties in 60 consecutive patients without using any kind of stent with satisfactory results.

MATERIALS AND METHODS :

This prospective study was carried out in the department of paediatric surgery, Gauhati Medical College Hospital, Guwahati. From March 2009 to February 2015, sixty-eight pyeloplasties were performed in 60 patients. All the fresh cases of hydronephrosis due to pelvi-ureteric junction obstruction (PUJO) were included in the study. Cases with previous failed pyeloplasty and with initial nephrostomy were excluded from the study group. Initial diagnoses were made by ultrasonography and cases were further evaluated by DTPA renal scan and intra-venous urography (IVU) and occasionally by CT-IVU. Micturating cystourethrogram (MCU) was done in bilateral cases and in cases where ureter was prominent in IVU. All the operations were done by a single surgeon through anterior extra-peritoneal flank approach and in one case transperitoneal approach was used because of ectopic kidney. Anderson-Hyne's pyeloplasty was done in all the cases. When the pelvis was too large, it was aspirated partially by a 20-G cannula for ease of handling through a small incision. Then the pelvi-ureteric junction was pulled out and extent of the narrow segment was defined. When the pelvis was grossly dilated (>5 cm), pelvic reduction was done. Ureter was transected just below the visible narrow segment. Then the ureter was split longitudinally towards the ipsilateral renal pelvis till normal caliber ureter was obtained. A No. 5 feeding tube was then passed into the ureter and flushed with normal saline to ensure distal patency and free flow. Pyeloplasty was then done by suturing the spatulated ureter with dependant part of the renal pelvis by 6-0 polyglycactyn sutures - posterior layer continuous and anterior layer interrupted. Upper part of the pelvis was closed by 6-0 polyglycactyn running sutures. Initially, we used upper tract drainage routinely in all the cases where pelvic reduction was (initial 20 cases). But instead of nephrostomy we did pyelostomy, by inserting a 6 Fr/8 Fr Foley catheter into the pelvis through the pelvis itself rather than through the renal parenchyma. A perinephric drain (No.10 feeding tube) was used in all the cases and this tube was brought out along with the pyelostomy tube through a single skin incision (Fig. 1). Perinephric drain was removed as soon as output subsided (usually 24 to 48 hours). Intermittent clamping of the nephrostomy tube was started after 48 hours, for increasing period of time. Initially we did nephrostogram in all cases with nephrostomy on 6th or 7th postoperative day. Whenever flow of contrast was seen into bladder the tube was removed. Later on with experience and refinement of operation we stopped

doing nephrostogram routinely and did not encounter any problem like urinoma formation and persistent urine leakage after removal of the pyelostomy tube. In all cases we used a perinephric drain. Urinary catheter was not used. Prophylactic antibiotic was given for 6 weeks post operatively. Cases were followed up regularly for post operative results; ultrasonography was done at 3 months and 6 months and 1 year and, DTPA scanning was done at 6 months.

Table 1: Showing age distribution

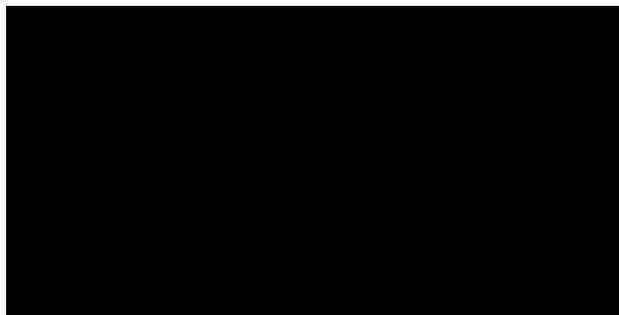


Table 2 : Sex and side distribution of the cases.

Sex	No. of cases	Right side	Left side	Bilateral
Male	32	10	17	5(10 kidneys)
Female	18	6	11	1(2 kidneys)
Total	56 kidneys	16	28	12

RESULTS :

Total 68 pyeloplasties were done in 60 patients. Youngest patient was 2 months old and oldest was 9 years old at the time of surgery. Maximum numbers of patients were operated below 2 year of age. Mean age was 9.4 months. Age distribution is shown in table 1. There were 38 male patients and 22 female patients. Left side was involved in 31 cases and right side in 19 cases and 8 cases had bilateral involvement. Out of eight bilateral cases 6 were male and two were female. Side and sex distribution is shown in table 2. Seven bilateral cases were operated at the same time and in one case each side was operated at intervals. Most common presentations of the cases were abdominal lump in 31 cases, recurrent pain abdomen in 14 patients, urinary tract infection (UTI) in 8 cases, incidental in 6 cases; 10 cases were diagnosed in the antenatal period. Indications for operation in our cases were palpable lump (31 cases), USG and IVU showing hydronephrosis and DTPA scanning showing less than 40% function on affected side, progressive hydronephrosis on USG and IVU even when there was no significant reduction of function on DTPA scan. MCU was done in 10 cases and there was no case of vesico-ureteral reflux (VUR).

Mean operative time was 78 minutes (range 55 min -115 minutes). Pyelostomy was done in 20 cases and contrast study was done in 16 cases and 14 cases showed free flow of dye into bladder. In two cases no passage of dye was observed on initial study. They were observed by intermitted clamping and repeat nephrostogram was done after 14 days and it showed flow of contrast into bladder. In rest 4 cases where nephrostogram was not done, there was no problem related to urine leakage like urinoma formation or persistent urine leak after removal of the tube. Mean hospital stay for patients with nephrostomy was 8.3 days (range 7-21 days); and without nephrostomy was 5.5 days (range 5 to 7 days).

Ultrasonography was done after 3 months and six months and DTPA scan after 6 months. Ultrasonography showed persistent residual dilatation in 90% of the cases but DTPA scan showed relieve of obstruction in all cases and improvement of function in 88% of the cases. Improvement of function varied from 0% to 32%. Increase in glomerular filtration rate and renal perfusion of individual kidneys were observed in bilateral cases. Post operative complications included paralytic ileus with hyponatremia and seizure in one case, prolonged bloody discharge through drain in one case. Two cases had edema at the anastomotic site and temporary blockage upto 3 weeks after operation. Post operative UTI was observed in two cases.

Fig. 1 : Schematic diagram of the procedure

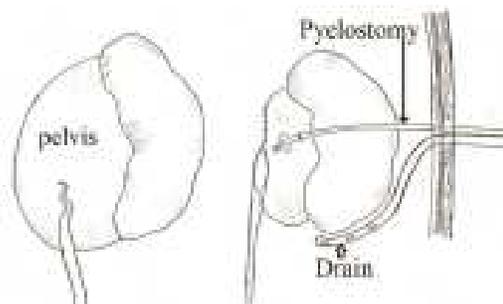


Fig. 2 : A case of left side pyeloplasty showing drain and pyelostomy tubes together



DISCUSSION :

Though many types of procedures are available for pediatric pyeloplasty, Anderson-Hynes pyeloplasty by open surgery remains the standard operation. There are various approaches for this operation viz; extra peritoneal flank approach, anterior transperitoneal approach, lumbotomy approach and laparoscopy. Main objective of all these operations is to widen the uretero-pelvic junction. Majority of the surgeons prefer either an internal or an exteriorized stent during operation [1,2]. But unanimity still exists regarding reduction of pelvis, upper tract drainage (nephrostomy), use of trans-anastomotic stent and perinephric drain. On one extreme some authors use all - nephrostomy, stent and a drain; on the other extreme some other surgeons use none.

When all three tubes are used, drain is removed as soon as there is no output (usually 24 to 48 hours), stent is removed before nephrostomy so that a nephrostogram may be done if deemed necessary. On the other hand some authors have done away with all tubes and tried to discharge patients on first post operative day with good result [3,4].

Proponents of nephrostomy as well as stent feel that urinary tract needs to be drained to protect the suture line, avoid invisible urinary leakage and to achieve fibrosis-free healing at the anastomosis and this does not necessitate longer hospital stay [1,5]. Nephrostomy is very safe, with few complications [6]. Nephrostomy tube dampens the high pelvic pressure if there is transient edema at the anastomosis and also provides access for radiographic study before removal of the tube. But we feel that when nephrostomy alone is used, free drainage through the nephrostomy tube may prevent urine to percolate across the anastomosis and may invite synechia formation. Therefore, we start clamping the nephrostomy (when used) tube after 48 hours. Question has been raised about renal scarring and nephron loss at the site of nephrostomy. We have avoided this by simple modification of putting the tube into the pelvis through the pelvis itself (pyelostomy) instead of through renal parenchyma. This tube can be passed very easily and without bleeding and thereby avoiding blood clot formation in the pelvis in the post operative period.

Antegrade placement of double 'J' stent is hazardous particularly in infants [7]. It is safer to put a small feeding tube several centimeters below the anastomosis. But these tubes may damage the ureterovesical junction and may raise a mucosal flap that can lead to a permanent obstruction requiring redo

surgery [8]. Other complications of stents include blockage / non-drainage, difficult retrieval, urine leak, urinoma, and post-operative infection [1]. A higher incidence of UTI has been observed in case of stented pyeloplasties compared to non-stented pyeloplasty [5,9]. We encountered UTI only in two cases during follow up period. We also feel that ureteral stent, especially in small infant, in whom the ureteric caliber is sometime too narrow, can cause mucosal damage and subsequent inflammation and synechia formation after removal of the stent. But Chandrasekharam has reported a large series of 116 laparoscopic pyeloplasties in infants, where he successfully placed a double "J" stent in 109 cases with satisfactory result and enabled him early discharge of the patients [10].

A routine dismembered pyeloplasty with minimum tissue handling may not warrant the placement of either nephrostomy tube or internal stent [6]. Smith et al analyzed 9 previous studies comparing a total of 339 stented with 494 nonstented repairs [5]. Overall the number of complications were almost equal (12% versus 14%) but the stented group had more infections, whereas more leaks occurred in the nonstented group. Therefore, when stent is not used the placement of a perinephric drain seems logical to

drain any leakage of urine that may cause urinoma formation. We used a no.10 feeding tube below the kidney and brought it out along with the pyelostomy tube (whenever used). These two tubes behave as a single tube outside the patient (Fig. 2). But role of stent is unequivocal for solitary or poorly functioning kidney, redo cases, concomitant presence of VUR, stone, and residual pyelonephritis [11]. We have used no.5 feeding tube as stent in two cases only, who had gross hydronephrosis with poor renal function (<10% on DTPA scanning), where temporary nephrostomy was done before pyeloplasty. These cases usually have chronic pyelonephritis and have chance of significant edema and obstruction. These two cases are not included in this study.

So, we conclude that routine dismembered pyeloplasty can be done satisfactorily without the use of any kind of stent. It avoids the possible complications associated with stent and eliminates another operating session for removal of an internalized stent. Upper tract drainage (pyelostomy) may be reserved for gross dilatation of pelvis where pelvic reduction is done. A perinephric drain for 24 to 48 hours is helpful in preventing urinoma formation and it does not add any morbidity to the patients.

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Original Article

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"A Comparative Study of T Tube drainage versus choledocho-duodenostomy in Choledocholithiasis"

ABSTRACT

Objective: Common bile duct (CBD) stones may occur in up to 3%-14.7% of all patients undergoing cholecystectomy operation. In spite of the modern era of 'minimally invasive approach', the open approach is still in practice in many centres. The aim of this study was to compare both short and midterm complications of T-tube drainage vis-a-vis choledocho-duodenostomy.

Materials and Methods: This is a prospective study carried out between July, 2013 and December, 2015. 100 patients underwent choledochotomy procedure done during this period which constitute about 8.26% of the total cholecystectomy operations. Cases were followed up following surgery for a period of 12 to 18 months. Morbidity, mortality and potential factors influencing the complication following T Tube insertion and choledochoduodenostomy were recorded.

Results: 1224 cases of cholecystectomy were done during the period and out of this 100 cases had CBD stones. T-tube was inserted after choledochotomy in 82 patients and Choledochoduodenostomy was done in 18 patients. Post operative complications occurred in 13 patients in T-tube drainage and 3 patients in Choledochoduodenostomy group. Most of the patients were discharged on 7th postoperative day in Group 2 and in group -1 on 15th postoperative day.

Conclusion: Choledochoduodenostomy is an easy, effective and definitive method of decompression, especially when there are multiple stones in a dilated CBD. A high rate of complications was found associated with T-tube drainage.

Key Words: Choledocholithiasis; Choledochotomy; T tube drainage; choledochoduo-denostomy; T-tube Cholangiography.

Introduction :

In 1889 Robert Abbe of New York performed the first documented successful choledocholithotomy. However the credit for popularizing choledochotomy as a routine procedure goes to Hans Kehr, who introduced the T tube for intraductal drainage which is still in use today. The first successful choledochoduodenostomy was performed by Sprengel in 1913 and ever since has been accepted as an easy and effective measure to drain the CBD.

Materials and Methods

All patients with Common bile duct stones undergoing treatment between July, 2013 and December, 2015 were included in this study. The study was also approved by Institutional

Ethical Committee. Patients were allocated to two groups,

Group-1: T tube was inserted following choledocholithotomy and in Group-2 Choledochoduodenostomy was performed. The CBD was opened through a supra-duodenal vertical incision between stay sutures. Stones were taken out and the CBD was flushed with normal saline to ensure patency. A T-tube was inserted after that in group 1. In Group-2 patients, a 2 cm long side to side anastomosis of the CBD and duodenum was done, if the diameter of the CBD was >15 mm, had multiple calculi and in elderly persons. The anastomosis was done in the most distal portion of CBD to avoid sump syndrome. A sub-hepatic drain was placed in all patients. T-tube Cholangiography was done on the 7th postoperative day in Group-1 patients. Once patency was confirmed, intermittent clamping of T-tube was done for few days and the tube was removed usually on 14th postoperative day. Most of the Patients were discharged on 7th postoperative day in Group 2. Cases were analysed prospectively for a period of 12 to 18 months. Morbidity, mortality and potential factors influencing the complication rate after T Tube insertion and choledochoduodenostomy were recorded.

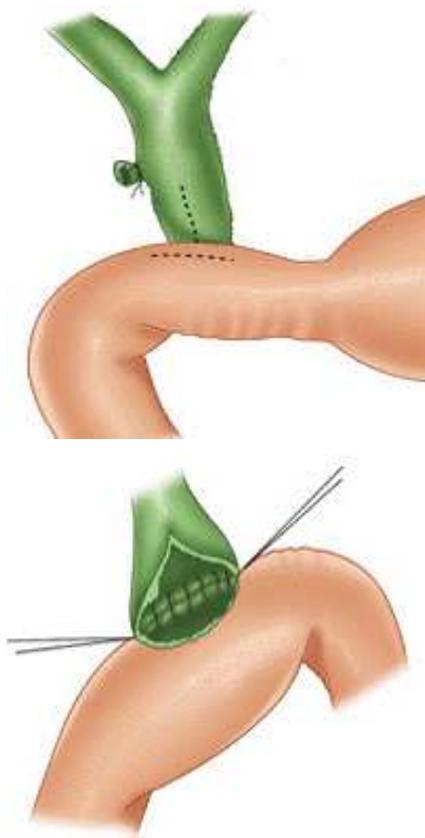


Fig.1: Diagrammatic representation of choledochoduodenostomy (source- Schwatz's Principles of Surgery, 9th edition)

Results :

1224 cases of cholecystectomy was done during the period out of which, 100 cases of choledochotomy was done. Age ranged from 20 years and to 80 years, with maximum cases (44) noted in the age group of 21 to 40 years, mean age being 50 years.

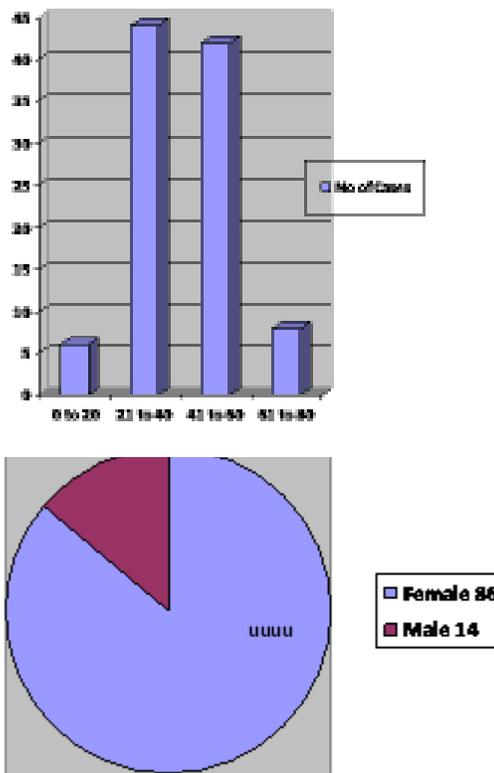


Fig.2: Showing age & sex distribution in the series

Females were predominantly involved (86%) with a male to female ratio of 1: 6.1. 92 patients presented with biliary colic with or without history of jaundice. The most common symptoms were right upper quadrant pain and jaundice and the most frequent physical signs were right upper quadrant tenderness and icterus.

Laboratory and imaging findings were recorded. T-tube was inserted in 82 patients (82%, 72 female) in group I and Choledochoduodenostomy was done in 18 patients (18%, 14 female) in group 2. Choledochoduodenostomy was done in elderly persons where CBD was more than 15mm in diameter with multiple calculi in CBD. Post operative complications were noted in 13 patients in group I and 3 patients in group 2. In group I, 4 had biliary leakage, 4 had retained calculus, 2 had wound infection, 1 had sepsis and difficulty in removal of T

Tube was noted in 2 cases. In group 2, biliary leakage, sepsis and wound infection were recorded in one patient each (Table 1). All patients who had biliary leakage were managed on conservative treatment with closed external drainage and antibiotics. Two patients, one in each group needed exploratory laparotomy when they did not respond to conservative treatment. Four patients with retained stone in group 1 required endoscopic sphincterotomy and removal of calculus by ERCP in another centre. Two patients developed severe pain after removal of T Tube, but did not require any intervention. 'Sump syndrome' was not observed in any of the cases in group 2. The median postoperative hospital stay in group-1 was 15 days while in Group-2 was 7 days. There was no mortality in either group.

Table-1: Postoperative complications in the two groups

	Group -1 (n=82)	Group -2 (n=18)
Biliary leakage	4 (4.87%)	1 (5.88%)
Sepsis	1 (1.20%)	1 (5.88%)
Wound infection	2 (2.40%)	1 (5.88%)
Retained Calculus	4 (4.81%)	0
Difficulty in removal of TTube	2	0



Fig. 1: T-tube cholangiography with retained calculus



Fig. 2: T-tube cholangiography with normal study

Discussion :

CBD stones may occur in 3%-14.7% of all patients undergoing cholecystectomy operation [1]. In the present study incidence of CBD stone was recorded in 8.26% of the patients. About 500,000 gallbladders are removed in the United States annually. The CBD is explored in nearly one quarter of them, and stones are removed in about two-third of these explorations [2]. In the present study most of the cases were in the age group of 21 to 40 years. Several studies reported commonest age for gall stone disease as more than 50 years [3-5]. 86% of the patients in the present study were females, which was similar to other series reported earlier [6,7]. 60% of our patients had multiple calculi in the CBD. 54.5% of the patients in the T tube group had multiple stones in the CBD, with 18% in the intrahepatic ducts. The indications for choledochoduodenostomy in the present series were large size of the CBD, multiple calculi with sludge, and elderly persons. Other indications, such as, ampullary stenosis, retained or residual stones, hepatic stones, distal CBD stricture, recurrent common duct stone, impacted ampullary stone, primary common duct stones etc were not found in any of our cases. Some authors prefer choledochoduodenostomy when CBD diameter is greater than 1.5cm in order to create a large opening between the bile duct and duodenum [8]. Many experienced surgeons traditionally choose open biliary surgery and perform this technique with excellent outcome [9]. The routine use of Intraoperative Cholangiography (IOC) is still controversial. Some authors support routine IOC, while others favour selective IOC and others report no advantages in IOC with respect to missed CBD stones [10-12]. However, it

can be a useful tool to identify choledochal stones [13]. IOC was not performed in any of our cases.

In the present series, biliary leakage and bacteraemia were the two main problems encountered after T-tube removal. Out of 33 patients in this group, one patient developed biliary peritonitis after removal of the T tube . The leak from the CBD was managed by endoscopic stenting with antibiotic cover. One patient had a retained stone.

Choledochoduodenostomy, although an easy and safe procedure, was abandoned for the fear of sump syndrome [14]. However long term follow up of patients with this procedure could not substantiate the fear [15] . The term sump syndrome signifies variety of symptoms caused by stone, sludge or food residue stagnating in a blind pouch of CBD distal to the anastomosis. Making an adequate size stoma is the key to prevent sump syndrome. Various studies showed that the immediate postoperative complications were significantly less following choledochoduodenostomy as compared to T tube drainage [16]. The need for re-exploration for recurrent choledocholithiasis was reported to be high for the T-tube group in another study [17].

Conventionally, T-tubes are removed between 14th and 16th post-operative day, provided the T-tube cholangiogram shows normal finding [18]. However, some other authors preferred to leave latex T-tubes in for 21 days [19]. Current practice favours latex as the material of choice for T-tubes considering its inflammatory reaction, rate of development of a fibrous tract and absence of bile precipitation in the lumen [20]. However, prospective trials involving latex T-tubes have also shown cases of bile peritonitis after their removal [21]. It is suggested that if T-tubes are to be used broad spectrum antibiotic cover should be employed at the time of removal [14]. Major causes of bile leakage after T-tube removal are rupture of the T-tube tract and outflow obstruction [22]. There is also possibility of the CBD being traumatized at the time of removal with possible fibrosis and stricture formation [7,23]. Migration of the T-tube into the duodenum diagnosed during T-tube cholangiogram is also reported [24].

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Table 2 : Recent series showing complications following T Tube & choledocholithotomy

Table 3 : Series showing complications following choledochoduodenostomy

Conclusion :

With the availability of sophisticated gadgets and minimally invasive surgery, CBD stones are now managed with minimal postoperative complication and better long term outcome and low recurrence rate. However, choledochoduodenostomy is a valuable and safe procedure, have stood the test of time and can be more useful in centres where per-operative cholangiography is not available.

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Original Article

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Evaluation of Modified Alvarado Score in diagnosis of Acute Appendicitis

ABSTRACT

Objective: The aim is to evaluate Modified Alvarado Scoring System (MASS) in diagnosing acute appendicitis and its value as an aid in surgical decision making.

Materials & Methods: A prospective observational study, period-June,2014 to May,2015, in department of surgery of a teaching institute. Patients admitted with acute appendicitis were included. Parameters of MASS were measured and scores assigned to each patient. Operated appendix specimens were sent for histopathological examinations. MASS score was correlated with histopathological diagnosis. Sensitivity, specificity, PPV, NPV, accuracy, odds ratio and likelihood ratio of the MASS at cutoff point of 7 were evaluated.

Results: Total patients-160 (69-males, 91-females), 146 patients operated. 111 patients had histopathologically confirmed diagnosis. 117 patients were in score 7-9, 34 patients in 5-6 and 9 patients in 1-4. True positive were 89.74%(group7-9), 20.68% (group 5-6). Score 5-6 has higher false positive rate (79.31%) than score 7-9 (p-0.0001)

Estimated prevalence was 0.76(95% CI - 0.681 to 0.82). Overall sensitivity of MASS in score 7-9 was 94.59%, specificity-65.71%, PPV-89.74% and NPV-79.31%. The odds ratio was 33.54, likelihood ratio-2.77(conventional), prevalence weighted likelihood ratio-8.75 and diagnostic accuracy-87.67%. Negative appendicectomy rate was less in score 7-9. Rate was higher in females.

Conclusion: MASS is useful in diagnosis of acute appendicitis. It is sensitive, non invasive and provides good accuracy. Study showed significant decrease in negative appendicectomy in score7-9. A larger multicentre study is advocated to reinforce the findings.

Key Words : Modified Alvarado Scoring System; Acute appendicitis; Negative appendicectomy. Abbreviations: MASS - Modified Alvarado Scoring System

INTRODUCTION :

Acute appendicitis is a very common cause of acute abdominal pain. The incidence of the disease is 110 per 1,00,000 population over a life time period and the life time risk of the disease is 7%[1]. The rate of misdiagnosis of the disease is as high as 40% in certain populations [2-4]. The negative appendicectomy rate ranges from 15% to 30% [5,6]. Apart from clinical diagnosis many modern modalities like ultrasonography, laparoscopy, MDCT, MRI are used for diagnosis. But, the diagnostic accuracy remains between 80-90 percent[7-10]. The accuracy of clinical diagnosis ranges from 70-97% depending upon the clinical experience of the surgeon[10-13]. Traditionally surgeons have accepted a negative appendicectomy rate ranging from ten to twenty percent rather than a delay in treatment [14].



To improve the result of clinical diagnosis various scoring systems have been designed. But, most of these scoring systems are complex and many a times not feasible in an emergency setting[15-17]. The Alvarado scoring system was designed in 1986 was used to improve clinical diagnosis of acute appendicitis [18]. The scoring is based on some specific symptoms, signs and laboratory findings and they are allotted some values. The symptoms are migratory right iliac fossa pain (value-1), anorexia (value-1) and nausea and/or vomiting (value-1). The signs are tenderness in right iliac fossa (value-2), rebound tenderness (value-1) and elevated temperature (value-1). The laboratory findings are leucocytosis (value-2) and shift to left (value-1). The leucocytosis is defined as white cell count in excess of 10,000 per cubic mm and shift to left means left shift of neutrophil maturation (percentage of segmented immature neutrophils with total WBC count) [18].

Patients with a total score of 1 to 4 are considered as not likely to have acute appendicitis. Patients with a score of 5 to 6 are considered to have a possible diagnosis of acute appendicitis, but the score does not warrant immediate surgery and needs further review. Patients with a score of 7 to 8 are considered to have probable acute appendicitis. Patients with of 9 to 10 are considered to have an almost definite diagnosis of acute appendicitis and they are subjected to surgery.

The Alvarado score was modified by M. Kalan, D. Talbat, W.J. Cunliffe and A.J. Righ in 1994. The laboratory criteria of 'shift to the left of neutrophils' (value-1) was excluded from the scoring system. Because the facility for this test was not routinely available in the laboratory they used during the study. So, in Modified Alvarado Scoring System the patients were scored out of 9 rather than 10[19].

The aim of this study is to evaluate the Modified Alvarado Scoring System (MASS) in the diagnosis of acute appendicitis. The aim also includes evaluation of feasibility and value of Modified Alvarado Scoring System as an aid in surgical decision making so that negative laparotomies can be minimized.

Materials and methods:

The study was conducted in the department of surgery of a teaching institute on patients who were admitted in different surgical units with a provisional diagnosis of acute appendicitis. Approval of the institutional ethics committee was taken prior to the study.

The patients who developed appendicular lump or appendicular abscess, generalized peritonitis were

excluded from the study. Pregnant women with a provisional diagnosis of acute appendicitis were also excluded from the study. The patients included in the study were explained about the study procedure in their mother tongue and written consents were taken.

It was a prospective observational study. The null hypothesis was that Modified Alvarado Scoring System is not sensitive in the diagnosis of acute appendicitis. The study period was one year starting from July,2014 to June,2015.

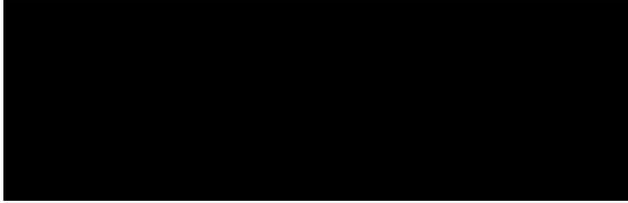
A detailed history and thorough clinical examination was done in each patient to diagnose acute appendicitis clinically and records were put in a predesigned proforma. On separate sheets scores were assigned to each patient from 0 to 9 based on Modified Alvarado Scoring System.

The observed value in each parameter were added and the total value (MASS score) was assigned to the patient. If the MASS score is from 7 to 9, acute appendicitis is likely and these patients underwent appendicectomy. If the score is from 5 to 6 the diagnosis is equivocal. These patients were observed for next 24 hours for frequent assessment of scoring. If score becomes more than 6 or the surgeon found the clinical condition of the patient highly suspicious of acute appendicitis they were operated upon. If the MASS score is from 1 to 4, then acute appendicitis is an unlikely diagnosis. These patients were observed and managed conservatively and reassessed at regular intervals. If the treating surgeon considered the condition highly suspicious of acute appendicitis then the patient was operated.

The specimens of appendix of the patients who were operated were carefully examined. Both the serosal and mucosal surfaces were examined for macroscopic features of acute appendicitis. Specimens were labeled and sent for histopathological confirmation of the diagnosis.

The clinical findings and scoring of the patients were correlated with histopathological findings and the Modified Alvarado Score was evaluated in term of its diagnostic value.

Appropriate statistical tests were used to evaluate the sensitivity, specificity, PPV and NPV of the Modified Alvarado Score with cutoff point of 7. Evaluation and case categorization were done as follows-



Sensitivity = $TP / (TP + FN) \times 100\%$
 Specificity = $TN / (TN + FP) \times 100\%$
 PPV = $TP / (TP + FP) \times 100\%$
 NPV = $TN / (FN + TN) \times 100\%$
 Accuracy = $(TP + TN) / (TP + FP + TN + FN) \times 100\%$
 Odds ratio = $(TP \times TN) / (FP \times FN)$
 Likelihood ratio = $Sensitivity / (1 - Specificity)$

Statistical significance of the study is done by using student 't' test for comparison of mean value, whereas for the comparison of proportion, p value is calculated using Chi square with or without Yates correction and Fisher's exact test using 2 tailed p value whichever applicable depending on the sample size using IBM SPSS statistical software v 2.1, MS Excel 2010 and GraphPad Prism version 6.05.

Results and observations:

A total of 160 patients were admitted with a provisional diagnosis of acute appendicitis. Out of these 69 were male and 91 were female. One hundred and forty six patients were operated, 64 males and 82 females. Number of patients who had histopathologically confirmed acute appendicitis in the operative specimen was 111. The estimated prevalence of appendicitis in our study population is 76.02% (95% CI 68.1% to 82.52%).

There were 117 patients (Male-51, Female-57, Children-9) in the score group 7-9, 34 patients (Male-13, Female-21) in the score group of 5-6 and 9 patients in the score group of below 5. Out of 9 children 3 were male and 6 were female (Table I, Figure-1).

All patients in the study group had pain abdomen. But, migratory right iliac fosse pain was seen in 67.64% (score group 5-6) and 66.66% (score group 7-9) of patients. Anorexia and nausea and/or vomiting were more common in 7-9 group. Tenderness in right iliac fossa was present in 99.14% (7-9 score group) and 79.31% (5-6 score group). Rebound tenderness and pyrexia was higher in 7-9 score group than in 5-6 score group. Leukocytosis was also seen more in 7-9 score group. (Table II, Figure 2)

The estimated prevalence of acute appendicitis in the study population was 0.76(95% CI - 0.681 to 0.82).

Taking a score of 7 or more as diagnostic in Modified Alvarado score system, it was found that overall sensitivity of MASS was 94.59%, specificity was 65.71%, PPV was 89.74% and NPV was 79.31%. The odds ratio was 33.54, likelihood ratio was 2.77 (conventional), prevalence weighted likelihood ratio was 8.75 and diagnostic accuracy was 87.67%. It has been seen that modified Alvarado score system has higher sensitivity, specificity, PPV, NPV and diagnostic accuracy in male. (Table III)

The proportion of true positive in score group of 7-9 was 89.74% (Male-94.11%, Female-84.2%, Children-100%). (Table IV) The proportion of true positive in score group of 5-6 was 20.68% (Male-20%, Female-21.05%). The score group of 5-6 has a higher false positive rate (79.31%) than the score group 7-9 (p-0.0001) (Table V)

It was seen that the negative appendectomy rate was less in the score group of 7-9 than those in the score group of 5-6. (10.25% vs. 23.97%, p- 0.018) The rate was higher in females than in males. (29.26% vs. 17.18%, p-0.0408) This rate was higher in females both in 7-9 and 5-6 score groups (Table VI, Figure 3).

Table I: Distribution of male, female and children in different Modified Alvarado Score group

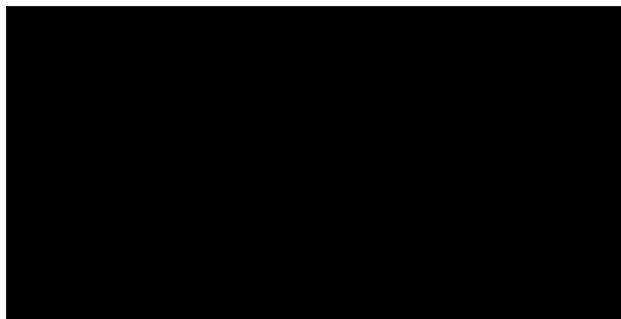


Figure 1: Distribution of male, female and children in different Modified Alvarado Score groups

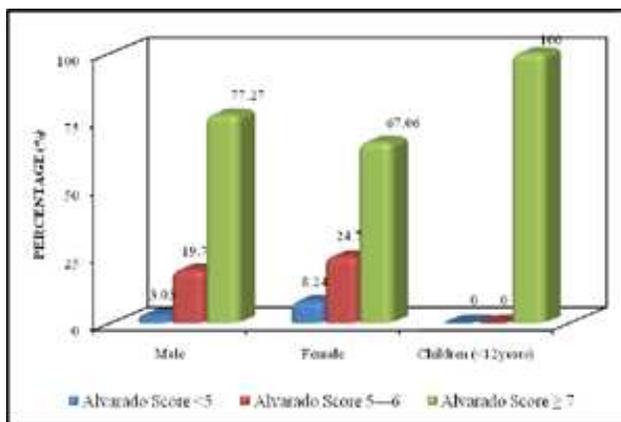


Table II: Distribution of signs and symptoms in different Modified Alvarado Score group

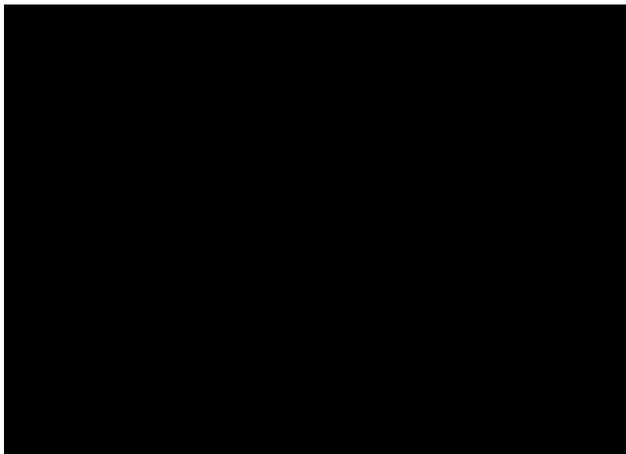


Table IV: Histopathological findings of the appendix specimen in Modified Alvarado Score 7-9 group

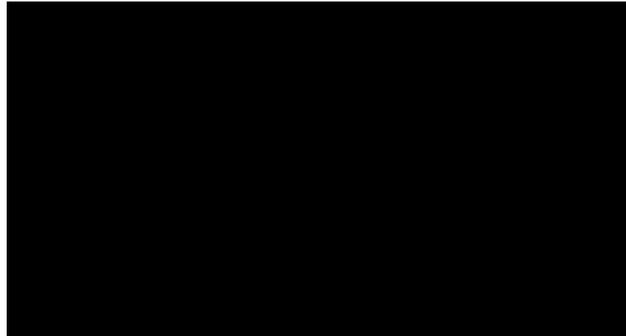


Figure 2: Distribution of signs and symptoms in different Modified Alvarado Score group

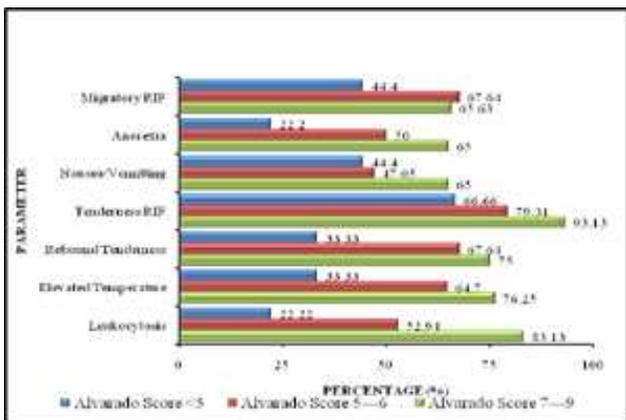
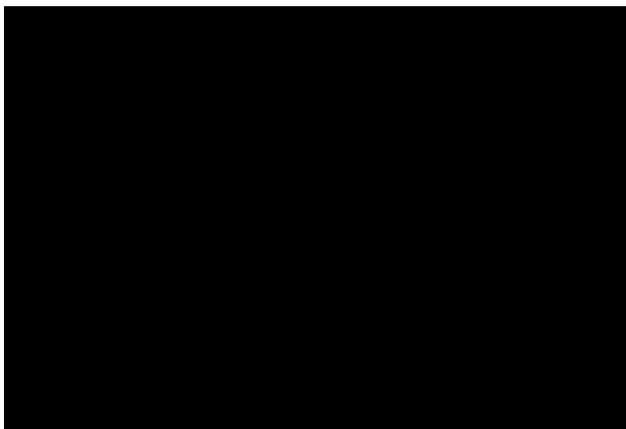
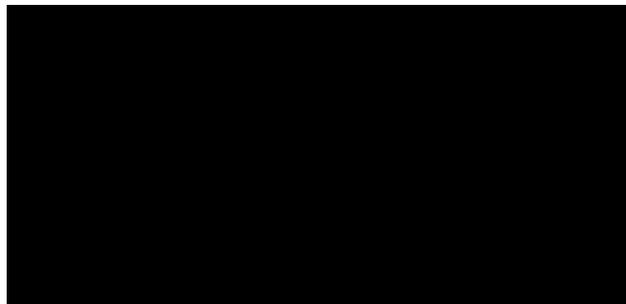


Table III: Sensitivity, specificity, PPV, NPV and Diagnostic accuracy of Modified Alvarado Score (Cutoff value 7 or >7)



N.B.: Number in parentheses indicate children (<12years)

Table V: Histopathological findings of the appendix specimen in Modified Alvarado Score 5-6 group



Discussion:

Modified Alvarado score system is a good supporting tool for diagnosis of acute appendicitis. It is simple, easy to use, noninvasive in nature and requires no special equipment. Many studies have shown its validity in diagnosing acute appendicitis [19-24]. But, at times it may be inadequate as a single diagnostic tool [7]. In some studies Alvarado score has shown poor results in women, children and elderly patients [25-27]. The score can also be used to identify the patients who need imaging studies for diagnosis [28].

There has been a shift in the paradigm with advancement of laboratory facilities and the imaging studies. Modern texts say that investigative studies give an additional edge in improving the diagnostic accuracy of acute appendicitis [29,30].

According to Modified Alvarado scoring system patients with a score below 5 are not considered to have acute appendicitis [19]. We had 9 patients who had a score below 5. All these patients recovered following conservative treatment and had no symptom thereafter. No patient was operated in this group. As the study was based on histopathological confirmation of the

disease in the resected appendix specimen, this group could not be taken for evaluation of the scoring system.

In evaluation of 5-6 group, 29 patients were operated. It was found that out of them only 6 patients showed evidence of acute appendicitis in the operative specimen, true positive being 20.68%. It is significantly lower than 7-9 group. ($p=0.481$). True positive was slightly higher in females than in males in this group. (21.05% vs 20%, $p=0.327$)

In this study histopathological diagnosis of the operated specimen was taken as the final diagnosis and modified Alvarado score was correlated with this diagnosis. These specific parameters could not be evaluated separately in the children as there was no patient in the paediatric age group who underwent operation and had a score below 7.

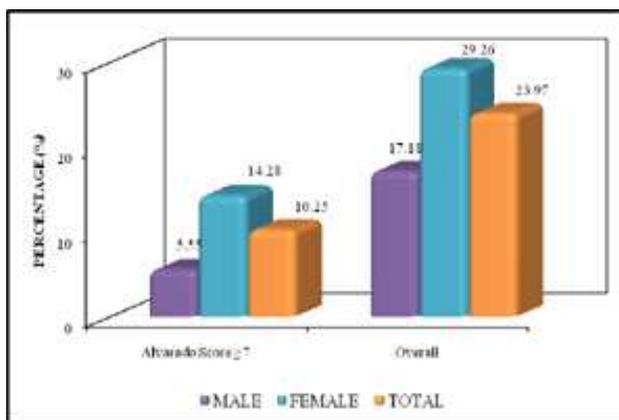
In the score group 7-9, out of 117 specimens 105 specimens had evidence of acute appendicitis and 12 specimens were normal. Proportion of true positive in this group is 89.74%. This is similar to findings of Ramchandra et al, Ijeri and Jadhav and Kanumba et al [20,31,32]. The PPV of MASS at this cut off point comparable to PPV obtained by CT [33].

The proportion of true positives in score group 7-9 was more in males than females (94.11% vs. 84.21%, $p=0.331$) Out of the female patients who were operated on basis of modified Alvarado score one had right sided ovarian cyst torsion and another had bilateral salpingitis with pelvic inflammatory disease. Dey et al also found female patients with twisted ovarian cyst and another with salpingitis in 7-9 score group in a study of 92 patients [34]. This group also included 9 children who had evidence of acute appendicitis in all specimens yielding a PPV of 100% ($p=0.1435$).

The PPV in our study is 89.74% in 7-9 group. The scores for PPV in this group ranges from 71.79% to 88.63% [19,28, 31,32]. From the observed sensitivity and PPV it can be commented that there is high likelihood that acute appendicitis can be diagnosed in this group by using the scoring system.

Table VI: Negative appendectomy rate in different Modified Alvarado Score groups

Figure 3: Negative appendectomy rate in Modified Alvarado Score group 7-9 and overall negative appendectomy rate



In our study the overall PPV in 5-6 group was 20.68%. Different studies show that the PPV in 5-6 group ranges from 34.04% to 63.63% [19,28,31,32]. It is evident that low and variable predictive value of MASS in 5-6 score group leaves significant lacunae to unanimously advocate its usage as a single diagnostic tool for acute appendicitis. It remains 'equivocal' [19-21]. This group will benefit if additional diagnostic tools are applied to increase the diagnostic accuracy.

The overall sensitivity of modified Alvarado score in the diagnosis is 94.59% when the cut off level of the score is 7 in our study. Higher sensitivity was found in males (96.22%) than in females (93.10%) ($P=0.0147$). The literature shows that the overall sensitivity ranges from 82.7% to 99% [19,22,31,33,35,36]. It means that if the cut off value MASS is taken as 7, more than 90% of the patients can be diagnosed as acute appendicitis. If the score is below 7, the chance of diagnosing acute appendicitis is less.

The study results showed low specificity (65.71%). It was lower (62.50%) in females than in males (72.72%) ($p=0.79$). Test with high specificity can correctly identify individuals who don't have the disease (true negative). The specificity of MASS is found to be suboptimal in the literature [22,24,27, 31-35,37-47]. It implies that MASS alone may not be sufficient to rule in the diagnosis of acute appendicitis. The lowest specificity in the literature is found to be 37.5% at cut off point 7[41].

It is seen that even at cut off point 7, there is a small chance of diagnosing the disease when in fact some other pathology is causing similar symptoms. This is more likely in females. In males and children, if specificity is combined with PPV at scores 7-9, MASS can be considered as an effective tool in diagnosis of acute appendicitis. To use MASS effectively as a 'rule in' test it needs to complement with a test that is highly specific.

The accuracy of diagnosis entitles recognition and removal of inflamed appendix prior to its perforation. It also minimizes number of negative appendicectomies. In our study, the diagnostic accuracy was 92.8% in males and 84.4% in females ($p=0.707$) at cut off value of 7. The overall diagnostic accuracy was 87.67%. This is more or less similar to study done by Kanumba et al (92.9%)[35]. It can be said that if MASS done correctly in this group, then approximately nine out of ten patients can be accurately diagnosed. In a developing country acute appendicitis is a common emergency and it causes a significant burden on health service [48]. MASS may be helpful in such situations.

The likelihood ratio is of importance in determining value of clinical tests. The positive likelihood ratio is 2.77 in our study at the cutoff point of 7. When estimated prevalence of acute appendicitis was weighed in the likelihood ratio was 8.75. According to clinical guideline this positive likelihood ratio indicates the increased probability of acute

appendicitis in patients with a MASS score of 7 or more. But, the likelihood ratio is not large enough for a conclusive diagnosis of acute appendicitis[49]. The likelihood ratio is of significance when it is above 10 or below 0.1[49]. Some of the similar observed likelihood ratio is seen in studies done by Dey et al (9.5), Macklin et al (6.81) and Chan et al (6.71) [35,47,50].

At the cut off value of 7, the negative appendicectomy rate in our study was 10.25%. More or less similar observations were seen in the studies of Ramchandra et al(11.36%), Ijeri et al (8%) and Dey et al (13%)[20,32,35]. In our study the females had higher percentage of negative appendicectomy in score group 7-9(17.18%) and also in the whole study group (29.26%). So it can be assumed that diagnosis of acute appendicitis should not be assumed entirely on clinical ground in females, particularly in reproductive age group. Some complementary tests should be done to rule out diseases with similar symptom complex.

The common histopathological diagnosis of the operated specimen of appendix was acute suppurative appendicitis(39.04%). In 7-9 group acute suppurative appendicitis was 47.86%. All specimens showing complicated appendicitis (12.31%, gangrenous and perforative) had MASS score of 7 or above. In a study of 127 patients, Kanumba et al found complicated appendicitis in 14.1% of patients. The perforation rate in their study group was 9.4% whereas in our study group it was 5.47%. In their study all these patients had a score of 7 or more ($p=0.106$)[36]. It seems that high MASS score has a correlation with complicated appendicitis.

It can be emphasized that modified Alvarado scoring system has a definite value in diagnosis of acute appendicitis. However, the study was conducted in a single centre for a limited period. A multicentre study over a longer period of time with large sample size will shed more light in this regard.

Summary and Conclusion:

The modified Alvarado scoring system is a sensitive and useful aid in the diagnosis of acute appendicitis. It is non invasive, easily reproducible and demonstrates a good accuracy in diagnosis at a cutoff score value 7 or more. It is a feasible clinical diagnostic tool. The study also finds a significant decrease in negative appendicectomy when score is 7 or more. The negative appendicectomy was higher in female than male in the study group. In females specially in reproductive age group with equivocal scores,

additional diagnostic test should be used to enhance the diagnostic accuracy. A large multicentre study is advocated to reinforce the usefulness of modified Alvarado scoring system in diagnosing acute appendicitis.

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Original Article

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Potency Outcome in Ischemic Priapism Presenting after 24 hours - Our Experience and Review of Literature

Objective :

Priapism is prolonged and persistent penile erection, unassociated with sexual interest or stimulation, lasting longer than 4 hours. Treatment comprises of aspiration/irrigation with sympathomimetic injections, surgical shunts, and rarely, penile prosthesis implantation. We report 12 cases of ischemic priapism treated at our centre with evaluation of the status of their potency post treatment .

Materials and method :

12 cases of ischemic priapism with median age of 37 years (range 24-50 years) were included in the study. Duration of symptoms ranged from 24 hours to more than 3 days. The patients were treated with corporal aspiration and injection of phenylephrine or distal shunting. The patients were evaluated at 1 month and 6 months post-treatment for their potency status.

Result :

25% of the patients presented after 24 hours, 50% presented after 48 hours and 25% presented after 72 hours. At 1 month, 4 patients (33.33 %) had morning erection. At 6 months, 5 patients (41.66 %) had morning erection. Average international index of erectile function (IIEF5) questionnaire score was 22.8 in the group with morning erection.

Conclusion :

Ischemic priapism constitutes a true Urological emergency that should be treated in a time bound manner. Priapism alone is a risk factor for erectile dysfunction (ED). Any shunt procedure may not modify this risk. Early correction of priapism will play a great role in preserving the erection. Penile prosthesis is the last resort in those who do not show improvement with other modalities of treatment.

Key Words : Priapism; potency; aspiration; shunting.

Introduction :

Priapism is defined as a prolonged and persistent penile erection, not associated with sexual interest or stimulation, lasting longer than 4 hours. It was first described in the literature in 1616 and named after Greek God Priapus[1]. Incidence rate in general population is 1.5 per 100,000 person-years [2]. In men 40 years of age and above it is 2.9 per 100,000 person-years. The incidence is much higher in high risk group of people like men using intracorporal injection therapy for erectile dysfunction (ED), where it ranges between 1% -17% [3]. This condition is exclusive to men and typically involves the paired corpora cavernosa. Rare instances of corpus spongiosum involvement with sparing of the cavernosal spaces have also been reported. Malignant priapism is an uncommon condition that is caused by metastasis of solid tumors to the penis, most frequently from the bladder and prostate



(32% and 28%, respectively), the kidney (17%), and the gastrointestinal tract (8%). These tumors rarely arise from primary sources in the testes, lung, liver, bone, or from sarcomas [4]. Here we report 12 cases of ischemic priapism who were treated in our department and status of their post treatment potency on follow up.

Material and methods :

12 cases of ischemic priapism patients who presented to our centre, a tertiary care hospital between 2010 and 2015 were studied. Non-ischemic priapism and shuttering priapism patients were excluded from the study. Patients were evaluated with history, physical examination and laboratory investigations like complete blood count, platelet count, coagulation profile ,aspirated corporal blood gas analysis for pH, pO₂,pCO₂, and hemoglobin electrophoresis (Figure 1).

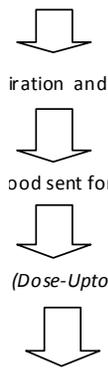


Figure 1 : Management protocol we followed in our hospital.

The patients were managed with intravenous fluid, intravenous antibiotic, proper analgesic and corporal aspiration with or without injection of phenylephrine or distal shunting under intravenous sedation (Figure 2). The patients were evaluated at 1 month and 6 months for their potency status.

Results :

The patient's age ranged from 24 years to 50 years (Median age 37 years). All patients presented with persistant painfull erection of the penis. On examination, the penis was rigid and tender in all of them. Chronic myelocytic leukemia was the cause of priapism in one patient whereas in the other 11 patients it was idiopathic. 3 patients presented between 24-48 hours, 6 patients between 48-72 hours and the other 3 patients after 72 hours of onset of penile erection. None of them presented before 24 hours. All patients had

failed to respond to the first line therapy of corporal aspiration and irrigation with inj. Phenylephrin (upto 200 µgm/ml, maximum 1 mg). After proper informed counselling all patients underwent shunt procedure. Distal T-shunt was performed in 8 patients and Ebbehoj shunt was performed in 4 patients (Randomly selected, as both procedures have similar outcome). 80% detumescence was achieved on day one.



Figure 2 : Distal shunt procedure.

All patients required repeated procedures with an average of 2 procedures per patient to achieve complete detumescence. Patients were discharged from the hospital on an average 8 days following admission. We followed the patients at 1 month and 6 months. Only 5 patients had morning erection and none had voiding dysfunction. On Colour Doppler ultrasound, 5 patients had normal monophasic baseline flow of cavernousal arteries (Figures 3 & 4).



Figure 3 : Doppler USG of penis showing good cavernousal arterial flow (At 1 month follow up).



Figure 4: Post operative photo (at 1 month follow up) .

At 6 months, the patients were called again for evaluation of their potency status. Only 5 patients had morning erection. After assessment with international index of erectile function (IIEF5) questionnaire, we found that patients with morning erection had an average score of 22.8 whereas patients without morning erection had an average score of 5.87. After 6 months, 5 out of 12 patients had recovered their potency (41.66%)(Figure 5). 7 patients lost their potency (58.11%). Patients who presented earlier had better potency outcome than those who presented late. All 3 patients who presented within 48 hours recovered their potency (100%). None of the patients who presented after 72 hours recovered their potency . Only 2 patients out of 6 (33.33%) who presented between 48 -72 hours recovered their potency.

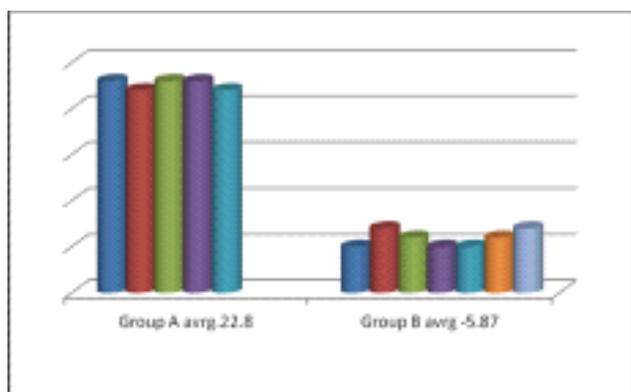


Figure 5 : IIEF 5 questionnaire point table of the patient at 6 month follow -up. (Group A -Had morning erection. Group B -Patient without morning erection.)

DISCUSSION :

Based on etiology and pathophysiology, priapism is of 3 types: ischemic, non-ischemic, and stuttering. Ischemic priapism, also termed veno-occlusive or low flow priapism, is a persistent erection usually associated with pain in the penis and marked by rigidity of the corpora cavernosa and little or no cavernous arterial inflow [5]. It is due to trapping of mixed venous blood in the penis creating venous congestion and ultimately ischemic environment in the corpora. It is the most common type and accounts for almost 95% of all cases [6]. The tissue ischemia and increased pressure generated within the corporal bodies lead to rigidity and pain in the penis, classically seen in ischemic priapism. Various studies have shown that ischemic priapism lasting longer than 24 hours results in erectile dysfunction with incidence as high as 90% [7]. Ischemic priapism represents a Urological emergency. Understanding the risk factors, commonly associated disease states will help the clinician to make an accurate diagnosis. The treatment may involve aspiration/irrigation with sympathomimetic drugs, surgical shunts, and penile prosthesis implantation. Non-ischemic priapism, also termed high-flow or arterial priapism, is a persistent erection caused by unregulated cavernous arterial inflow [5]. It occurs generally as a result of trauma resulting in an arteriolar-sinusoidal fistula. Non-ischemic priapism can also result from congenital arterial malformations, iatrogenic insults and following shunt procedures for ischemic priapism(8). In non-ischemic priapism, cavernous environment does not become ischemic secondary to the continuous influx of arterial blood [5]. As a result, the corpora are tumescent but not rigid, and patients typically do not complain of pain with erection [9]. For this reason, non-ischemic priapism is not an emergency and does not require immediate intervention. Stuttering priapism, also termed intermittent or recurrent priapism, is characterized by recurrent episodes of ischemic priapism. These episodes typically last less than 4 hours prior to remission [9,10]. Erections commonly arise during nocturnal sleep, or preceding or following sexual stimulation [11]. These episodes may increase in frequency and duration compromising the patient's quality of life and potentially developing into major episodes of ischemic priapism. Therefore, all episodes of recurrent priapism that progress to prolonged, painful erections should be treated promptly, according to the guidelines set for ischemic priapism [12]. Priapism represents a great challenge in therapeutic management as it carries with it significant

risk of structural damage to the penis and permanent erectile dysfunction [11]. Management should begin with a detailed history and physical examination. Diagnosis should focus on identifying any contributory/predisposing conditions [5,11]. Physical examination includes inspection and palpation of the penis, to assess the tumescence or rigidity, degree of corporal body involvement, and presence of tenderness [13]. In ischemic priapism, a cavernous blood gas analysis should be done for differentiating it from other variants of priapism [14]. The aspirated blood is dark and hypoxic in these cases. Typical blood gas values show a partial pressure of oxygen (pO₂) of less than 30 mmHg, partial pressure of carbon dioxide (pCO₂) of greater than 60 mmHg, and a pH of less than 7.25. Conversely, in non-ischemic priapism, the blood is bright red and oxygenated with cavernous blood gas values of a pO₂ greater than 90 mmHg, pCO₂ less than 40 mmHg, and pH 7.40 [5]. Other laboratory testing such as complete blood count, white blood cell differential, and platelet count to detect evidence of acute infections or hematologic abnormalities should be included in the diagnostic workup. Reticulocyte counts and hemoglobin electrophoresis may signify the presence of Sickle Cell Disease/trait or other hemoglobinopathies [5]. Penile imaging is useful in the diagnosis of otherwise equivocal cases and also helpful in the follow up for assessing treatment success. Colour duplex ultrasonography (CDU) of penis and perineum can evaluate intracorporeal arterial blood flow in real time which is helpful to differentiate highflow priapism from lowflow one [5,12,15]. Penile magnetic resonance imaging (MRI) has been shown to provide an assessment of smooth muscle viability in patients presenting with priapism. Potential benefits of this procedure include its ability to predict nonviable corporal smooth muscle after episodes of priapism as well as detecting other conditions such as malignant infiltration and segmental cavernosal thrombosis [16]. The most common complication of priapism is erectile dysfunction, which can occur in as many as 59% of cases [5,17]. Timely treatment is crucial for recovery of erectile function which may be seen in up to 44% of patients who experience priapism for 24-36 hours [18]. First-line therapy for patients with episodes of acute ischemic priapism is aspiration and irrigation of cavernous blood with or without intracavernous α -agonist injection. This can be performed by a dorsal nerve block or local penile shaft block [11]. A greater resolution of ischemic priapism and low post therapy erectile dysfunction is achieved following injection of a sympathomimetic agent with or without irrigation

(43-81%) than after aspiration alone with or without irrigation (24-36%). During phenylephrin injection, patients should be monitored for hypertension, headache, reflex bradycardia, tachycardia, palpitations, and arrhythmia [5]. Studies suggest that repeated injections and aspiration should continue for at least up to 1 hour prior to proceeding with second-line interventions in patients presenting with a priapism of less than 24 hours [5,8]. Clinical indicators that suggest failure of first-line intervention include recurrence of corporal rigidity, acidosis of cavernous blood gas analysis, absence of cavernosal artery inflow on penile color duplex ultrasonography (CDU) or elevated intracorporal pressures [8]. Ischemic priapism lasting more than 48 hours is unlikely to resolve with intracavernous injection/irrigation therapy alone. These patients should be counselled for immediate surgical shunting after a trial of intracavernous injection/irrigation [5,11,19]. There are four types of shunts: percutaneous distal shunts, open distal shunts, open proximal shunts, and vein anastomoses/shunts [8]. Guidelines advocate for an aggressive approach in treating refractory priapism cases in a serial fashion from distal to proximal shunts to vein shunting in order to achieve penile flaccidity [8,9]. Currently, data are limited to recommend one shunt over another. The summary data generated by the American Urological Association panel for treatment of priapism shows resolution rates of 74% for Al-Ghorab, 73% for Ebbehøj, 66% for Winter, 77% for Quackels, and 76% for Grayhack procedures. Erectile dysfunction rates are higher for the proximal or vein shunts (Quackels and Grayhack, roughly 50%) than for the distal shunts (25% or less) [5]. In addition, the duration of priapism is critical to the development of erectile dysfunction. So patients presenting with prolonged priapism are at risk for subsequent loss of potency irrespective of the treatment modality used. As priapism and its subsequent effects can be a litigious issue, it is very essential to give a clear counselling to the patient, informing him that the long standing priapism alone is a risk factor for erectile dysfunction, and that any shunt procedure might not modify the risk or procedure itself sometimes can result in loss of potency. There are no defined indications for implanting a penile prosthesis in patients with priapism. Acute priapism episodes lasting for more than 72 hours may be definitively treated with penile prosthesis implantation [20,21]. It may also be done in the non-acute setting where priapism has resulted in significant penile deformity and loss of potency [22,23].

Conclusion :

Low flow priapism is a Urological emergency. Prompt intervention will help to preserve the erectile function. Degree of erectile dysfunction (ED) depends on duration of symptoms and intervention.

Occasionally multiple procedures may be required to achieve detumescence. Delayed presentation is associated with corporal fibrosis and ultimately erectile dysfunction.

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Case Report

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Residual gall stones with subhepatic abscess

ABSTRACT

Stone spillage occurring during cholecystectomy may sometimes lead to a rare complication such as abscess formation if the stones are not completely retrieved or cleared off. The abscess may be located in and around the gall bladder fossa or in a distant, remote site and it usually occurs within 4 to 9 months after the initial operation but it happened after 8 years in our patient.

Key Words : *Laparoscopic cholecystectomy; Residual gall stones; Stone spillage; Abdominal abscess; Risk factors*

Introduction :

Laparoscopic cholecystectomy has become the standard procedure of choice for the surgical treatment of gall bladder diseases since its introduction in 1987. The surgical community is on the pursuit to determine the full spectrum of complication associated with this technique ranging from bile duct injury, superficial wound infection to those of late or remote clinical manifestation. It is worth mentioning that one such distant complication which can occur during laparoscopic technique is perforation of gall bladder and stone spillage particularly while dissecting an acutely inflamed gall bladder (15%) resulting either late intraperitoneal or retroperitoneal abscess. The incidence of perforation is believed to exceed that of traditional open method although not officially reported in the literature. Most cases of post cholecystectomy abdominal abscess formation being reported to occur between 4 months to 12 months however the incidence seems low but there may be a patient population who are at risk of developing such a long term complication arising out of retained stones.

Case Report:

A 55 yrs old lady was referred to our general surgery OPD from neurology as MRI of upper abdomen showed a right sided sub hepatic abscess of 80 mm x 40 mm size. Her chief complaint was right upper abdomen pain radiating to the back occurring since later part of February 2016. She was having moderate to severe, persistent pain which aggravated on movement but not accompanied by fever, vomiting, urinary or bowel symptoms. She had undergone laparoscopic cholecystectomy in 2008 at another hospital in the city but no previous documents were presented. Clinically her general physical state was normal but deep tenderness was elicited in the right hypochondrium. Her blood parameters showed mild leucocytosis but significantly high random blood sugar level. She was advised admission and surgery on the same day of consultation but she requested for conservative treatment hence injection ertepenem 1gm IV once daily was started and diabetologist opinion was obtained. 2 days later she

reported again with severe right hypochondrium pain and exhaustion, hence CT upper abdomen contrast study was done which revealed the same status of the abscess in the hepatorenal pouch. After necessary preoperative preparation, she was convinced for diagnostic laparoscopy under general anaesthesia with option for conversion to open and accordingly the subhepatic abscess was drained through a right subcostal incision as laparoscopy failed to locate the abscess due to dense adhesion in the perihepatic and stomach regions. The thick creamy white pus was evacuated through a blunt dissection starting from the right paracolic gutter and 8 pieces of medium sized calculi were detected in the abscess cavity. Pus culture showed presence of E.coli and was sensitive to tigecycline and gentamicin in descending order.

Discussion:

Residual gall stones can give rise to abscess either in the subhepatic + subphrenic spaces (intraperitoneal) or in the retroperitoneal areas including abdominal wall and it may occur weeks, months or even years after cholecystectomy [1-3]. It is less frequently seen after 12 months postoperatively and only one case of abscess formation being reported which occurred after 15 years of laparoscopic cholecystectomy [4]. Longest reported

abscess formation was an open cholecystectomy after 20 yrs. Gall bladder perforation (10-40%) during laparoscopic cholecystectomy or extraction by squeezing through the port can lead to stone spillage (7.3%) and its likely disappearance [5]. Papasavas found only 2 cases of stone spillage in open cholecystectomy, but they found same complication in 127 cases of laparoscopic cholecystectomy [6]. Risk factors of abscess formation are older age, male sex, acute cholecystitis, pigment stones, number of stones (>15), size of the stone (>1.5 cms) and perihepatic localization of lost stones [7, 8].

Conclusion:

Abdominal wall or cavity abscess secondary to retained stones occurring after many years of cholecystectomy is a rare entity. Delayed abscess development can be anticipated if above mentioned risk factors are considered and its incidence is directly proportional to lost stones [8] occurring due to stone spillage in the event of cholecystectomy. Regular use of endobag or any other cost effective alternatives can minimize stone spillage. Moreover, documentation of gall bladder rupture with stone spillage during surgery is crucial in terms of informing the patient for future follow up and identifying this late complication.

Fig 1 : MRI upper abdomen



Fig 2 : Residual stones



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Case Report

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Pancreatico-duodenectomy for pancreatico-pleural fistula: a rare indication

ABSTRACT

Pancreatico-pleural fistula is a rare complication of acute and chronic pancreatitis. Diagnosis requires a high degree of suspicion and is often delayed. The present case is a 10 year old boy who presented with dyspnoea and right sided pleural collection. Following thoracocentesis, there was repeated collection of pleural fluid at quick intervals, hence an intercostals chest drainage tube was inserted. The pleural fluid had markedly raised lipase and amylase levels. MRCP revealed a fistulous communication between the pancreas and the right pleural cavity. The patient did not improve with conservative treatment, therefore, Whipple's pancreaticoduodenectomy was done. The patient recovered well and has been asymptomatic till last follow up.

Key Words : *Pancreatitis; pancreatico-pleural fistula; pleural effusion; whipple's operation; pancreatico-duodenectomy.*

INTRODUCTION :

Pancreatico-pleural fistula (PPF) is a rarely encountered complication of acute and chronic pancreatitis. It is even rarer in children. Diagnosis requires a high degree of suspicion and is often delayed. Symptoms that suggest this condition include dyspnoea, abdominal pain, cough and chest pain. Analysis of the pleural fluid for raised amylase will confirm the diagnosis and investigations like CT scan, MRCP and ERCP will establish the fistulous communication between pleural cavity and the pancreas. Treatment starts with chest drainage, parenteral nutritional support and administration of intravenous octreotide. When these conservative measures fail surgical intervention in the form of endoscopic sphincterotomy and stent placement or pancreatic resection with drainage procedure is required.

Case report :

A 10 year old male child presented with dyspnoea, fever and pain abdomen. His Chest X-ray showed right sided massive pleural effusion. Thoracocentesis was done to relieve symptom. However there was rapid recollection of fluid leading to difficulty in breathing. HRCT thorax was done which showed massive right sided pleural effusion and collapse and consolidation of underlying lung parenchyma. Thus, a right sided intercostal water seal drainage was placed. The pleural fluid showed markedly raised amylase (3123.7 U/l) and lipase (1596 U/l) levels.



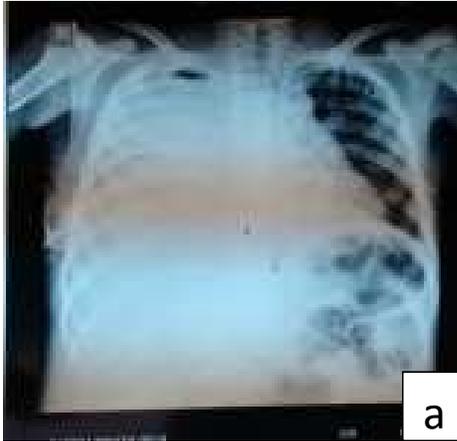


Fig 1 a & b: chest X-ray and HRCT thorax showing rt. sided massive pleural effusion and collapse of underlying lung parenchyma

CECT abdomen showed features suggestive of acute pancreatitis with multiloculated collection in right hepatorenal pouch and ascites. An MRCP revealed irregular dilatation of the main pancreatic duct (7mm) and its side branches with markedly atrophied pancreatic parenchyma. It also showed long irregularly outlined fistulous tract from major pancreatic duct (MPD) in the region of the head of the pancreas, extending along right subhepatic space and

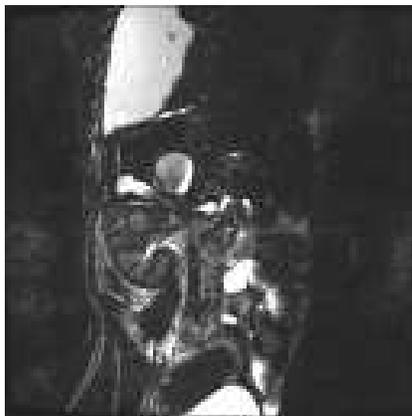


Fig 2: MRCP showing collection in hepatorenal pouch with fistulous tract communicating the pleural cavity with the dilated MPD

hepatorenal pouch into the right pleural cavity via a well-defined transdiaphragmatic tract. The transdiaphragmatic tract was noted in the posterolateral aspect of hemi diaphragm.

The patient was initially put on total parenteral nutrition and subcutaneous octreotide. After two weeks, oral feeding was started. The pleural drainage decreased to some extent but persisted. The patient developed right sided pyothorax for which right sided thoracotomy and decortication was done. The lung expanded following this but pleural drainage persisted. Hence pylorus preserving pancreaticoduodenectomy was done two weeks later. The patient had uneventful recovery and was discharged on 10th post-operative day. He has been on follow up for 3 months without any symptoms.



Fig 3: Resected specimen of head of the pancreas with part of duodenum. The arrow mark shows dilated MPD with proximal narrowed segment.



Fig 4: After completed pancreatico-jejunostomy, choledocho-jejunostomy and gastro-jejunostomy

DISCUSSION:

PPF is considered a rare pathology [1-3]. Its incidence is estimated at 0.4% in patients with pancreatitis and 4.5% in those presenting with a pancreatic pseudocyst [4]. Although pleural effusion is a common entity in children, right sided haemorrhagic pleural effusion due to PPF is rarely encountered. Hence, diagnosis of this condition requires a high degree of clinical suspicion.

Pleural effusion from pancreatic pathology is usually reactive and transudative in nature and so, self-limiting. The fluid amylase level remains below 100 IU/litre. However, effusion due to PPF is recurrent, massive and refractory to drainage procedure. Fluid protein, amylase and lipase levels are markedly raised [2,5,6].

Patients diagnosed with PPF are middle-aged (40-50 years), predominately male (83%), with chronic pancreatitis related mainly to alcohol abuse (67%) [5]. Clinical presentation of PPF varies between patients due to different levels of coexisting pancreatic injury and the amount of fluid collected in the pleural cavity. Ali et al reported that the most common symptoms are related to the pleural effusion [5]. These symptoms include dyspnea, cough, and chest pain. Abdominal pain was reported in only a third of the cases.

CT scan can identify a fistula in only 33-47% of cases [1,5]. The reason may be limited contrast resolution of CT in the case of narrow and tortuous canal with poorly enhancing walls, which is frequently hidden within pancreatic pseudocysts [1,5,7,8].

Magnetic resonance cholangiopancreatography (MRCP) is considered the choice of investigation as It allows for mapping of the ductal anatomy along with pathologic changes in adjacent structures that may provide with important information to understand the local anatomy and to plant an optimal treatment [1,5,7,9-11]. Endoscopic retrograde cholangiopancreatography (ERCP) is the second effective modality for diagnosis and treatment of PPF, with sensitivity of 46-78% [1,5].

Therapeutic options for PPF include conservative treatment, endoscopic management, and surgery. Medical treatment is usually attempted for 2-3 weeks using analogs of somatostatin to reduce pancreatic secretion and pressure in pancreatic ducts [1,10]. The success rate of this approach is between 30-60% [5,12]. ERCP allows for simultaneous assessment of the fistula and for decompression of ducts. Endoscopic procedures include sphincterotomy, stone removal, balloon stricture dilatation, and stenting [1,5]. Surgery is necessary in cases of noninvasive treatment failure, which is frequent in patients with severe pancreatic duct strictures and multiple polymorphic pseudocysts [1,5,7]. Depending on the local anatomy, distal pancreatectomy, pancreatojejunostomy or pancreaticoduodenectomy may be performed [1,5]. The present case initially showed some response to the conservative management, as the fistula output decreased to some extent. However, persistence of symptoms and fistula drainage as well as demonstration of a strictured segment in proximal main pancreatic duct led us to decide in favour of surgical intervention.

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Case Report

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Tuberculosis of Liver: A Case Report

ABSTRACT

Tuberculosis of liver is a rare disease. Pain in right hypochondrium and hepatomegaly are the main symptom and sign. A case of tuberculosis of liver is reported, that presented with hepatomegaly and pain in the right upper abdomen. Diagnosis of hepatic tuberculosis was made after laparotomy and liver histology. The patient recovered completely after a course of antitubercular therapy.

Key Words : Tuberculosis; hepatomegaly; liver biopsy; antitubercular therapy.

INTRODUCTION :

Tuberculosis of liver is an uncommon disease. Liver involvement may occur in untreated or inadequately treated case of pulmonary tuberculosis. Clinical pictures may be variable. Common symptom and signs include pain in right upper abdomen and hepatomegaly. Laboratory investigations and imaging methods are often normal. Diagnosis of tuberculosis of liver rests on histology of the lesion. A rare form of tuberculosis of liver is presented here.

Case Report :

The patient was a 41 years old male with complaints of distension and pain of the right upper abdomen for last 8 months. The pain was dull aching in nature and there was no aggravating or relieving factors. On examination the patient was pale and a lump of 3"x2" size was palpable in the right hypochondrium arising from the right lobe of the liver, firm in consistency, with irregular surface and ill defined margins. Examination of chest and C.V.S were found to be normal.

Prior to hospitalisation the patient received a course of antimicrobial drugs for two weeks without any improvement. Patients investigation showed, Hb -50%, T.L.C.-7500/mm³, ESR-92 mm at the 1st hour. Liver Functions Tests were within the normal limits. Hepatitis Profile A,B,C-Negative. X Ray Chest and Abdomen were normal. U.S.G. showed thickened gall bladder and hepatomegaly with coarse echo texture (Fig. 1). A contrast CT scan was abandoned due to sensitivity of the patient to contrast medium.

[N.B.: This article was submitted by Dr. Banamali Nath long back and was pending for major revision. The editor has revised the article to be published in the current issue as a tribute to late Dr. Nath].





Fig.1

An exploratory Laparotomy was planned. Abdomen was opened by upper right paramedian incision. Visceras were examined and found to be normal. The right lobe of liver was macroscopically pale with whitish appearance. Lower part of the right lobe was enlarged and granular. The gall bladder was normal. A wedge biopsy was taken from the enlarged part of the right lobe of the liver. Bleeding from the liver was controlled by gel foam packing. The abdomen was closed in layers.

Histology confirmed the lesion as hepatic granuloma with central caseating necrosis, suggestive of hepatic tuberculosis (Fig. 2). The patient was treated with 3 drug antitubercular regime comprising of isonex, rifampicin & ethambutol daily for 2 months followed by isonex & rifampicin daily for the next 7 months. After one month of treatment, the patient showed marked improvement and the patient reported full recovery after completion of the course.



Fig. 2: Granulomatous lesion with central necrosis in liver parenchyma.

Discussion

Tuberculosis of the liver is a rare entity and occur in 50-80% of patients who are dying of pulmonary TB

[1]. Most of the cases remain clinically silent and a conclusive diagnosis is possible only by histology. With the increasing resurgence of TB, the incidence of hepatic TB has also been increasing [2].

Liver has rich blood supply and with large number of reticuloendothelial cells, it is a common site for granuloma formation. A majority of granulomas are usually located near the portal tract and there is only mild perturbation of hepatic function, so most patients are minimally symptomatic or asymptomatic. Primary hepatic TB is rare because low oxygen tension in the liver is unfavorable for growth of mycobacteria [3]. Hepatic TB secondary to pulmonary or intestinal tuberculosis is more common. Reed classified hepatic tuberculosis into three forms: tuberculosis of the liver associated with generalized miliary tuberculosis, primary miliary tuberculosis of the liver, and primary tuberculoma or abscess of the liver [4]. The present case had primary tuberculoma (Type 3).

Hepatic tuberculoma lack specific signs or symptoms. Hepatomegaly is usually found with an increase in alkaline phosphatase and normal transaminase levels [2,4-8]. Jaundice, if present is often confused to be caused by hilar tumors, and some liver tuberculoma patients may be associated with portal hypertension or biliary tract bleeding [9,10]. Imaging techniques are useful in making the diagnosis of tuberculoma or tubercular abscess. On CT, the liver tuberculoma appears as an un-enhancing, central, low density lesion owing to caseation necrosis with a slightly enhancing peripheral rim corresponding to surrounding granulation tissue [11-13]. CT may show multiple lesions of varying density, indicating that there are lesions in different pathologic stages coexisting in hepatic TB, including tuberculous granuloma, liquefaction necrosis, fibrosis or calcification [14]. MRI of hepatic tuberculosis shows a hypointense nodule with a hypointense rim on T1-weighted imaging and hypointense, isointense or hyperintense with a less intense rim on T2-weighted imaging, and peripheral enhancement or internal septal enhancement on post-contrast MRI [12,15]. Cao et al, reported that contrast-enhanced ultrasonography may be helpful in differentiating hepatic TB from other hepatic focal lesions [17]. In recent years, the F-18 FDG PET/CT is widely used in the diagnosis and staging of malignant tumors, but prone to misdiagnosis in the diagnosis of hepatic tuberculosis, because the hepatic tuberculosis also showed FDG-avid [18].

Zheng et al reported five cases of hepatic tuberculosis, in 3 of them a diagnosis was possible on fine needle aspiration biopsy[16]. Clinical presentations were pain abdomen, fever, lethargy, jaundice, raised

alkaline phosphatase level and ESR. Quadruple therapy including isonex, rifampicin, pyrazinamide and ethambutol was employed and therapy continued for 1 year. Three patients required surgical procedures: for acute acalculus cholecystitis and enucleation of mass, for excision of a left hepatic lobe mass thinking it to be a cholangiocarcinoma and in the 3rd case for biopsy of a hilar mass causing jaundice. All the patients recovered well except one, who had received a renal transplantation two months before diagnosis of hepatic pathology. Immunological investigations are also at times unreliable and non specific. Final diagnosis rests on exploratory laparotomy and biopsy.

Very often, a diagnosis of amoebic tuberculosis have been made in hepatic tuberculosis cases, particularly in developing countries where amoebic hepatitis is common. Liver function also remains within normal limits in early stage of the disease, making clinical diagnosis very challenging. Image examination associated with image-guided fine needle aspiration biopsy is the best diagnostic method. Anti-TB treatment is effective in most of cases. However, if there are indications for surgery or difficult to diagnose, surgical procedures along with anti-tubercular drug therapy could be adopted.

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Case Report

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Pyloric atresia Type II and epidermolysis bullosa : A Case Report

ABSTRACT

Pyloric atresia is a very rare cause of gastrointestinal obstruction in neonates and usually occurs as an isolated anomaly. The coexistence of pyloric atresia (PA) and genetically mediated epidermolysis bullosa (EB) is rare but a well documented neonatal surgical emergency. The course of PA-EB is usually severe; extensive lesions generally have a fatal outcome in the neonatal period due to sepsis and fluid loss. Herein, we report a successful management of a case of congenital PA and EB.

Key Words : Pyloric atresia; Epidermolysis bullosa; Gastroduodenostomy.

INTRODUCTION :

Pyloric atresia is a very rare congenital anomaly. It comprises 1% of all intestinal atresia and its incidence is approximately 1 in 100,000 newborn [1]. The association of Pyloric atresia(PA) and epidermolysis bullosa(EB) is rare but is a known distinct clinical entity with autosomal recessive inheritance. In 20% of the cases epidermolysis bullosa is associated [2]. High mortality is reported in PA-EB due to complications related to EB [3]. Early diagnosis and prompt treatment of PA is associated with very low morbidity and mortality, of which main causes are septicaemia and electrolyte disturbance.

Presentation

A Case Report:

A 1.570 kg female baby was born by spontaneous delivery to a 24 year lady gravida 2 at 34 weeks of gestation. No antenatal details were available. A non-consanguineous marriage parent from a low socio-economic condition, of manual tea garden workers with no family history of any bullous disorder. On second day of life, the baby presented with non-bilious vomiting and on fourth day of life, she was referred for surgical opinion. On examination, the neonate was hemodynamically stable. There was no abdominal distension but well demarcated single areas of peeled skin were documented over right lower abdominal wall [Fig. 1a]. It was again observed that on fourth post-operative day onwards new blisters continued to develop spontaneously and without any apparent trauma [Fig. 1b]. Histopathological finding of a skin biopsy were compatible with EB (intraepidermal split). Systemic examination revealed no other abnormality. X-ray abdomen with air contrast study revealed a single gastric gas bubble with paucity of air distal to the stomach [Fig. 2]. Routine haematological investigations were within normal limits. At laparotomy, pyloric atresia was confirmed type II (obstructed pyloric canal by a solid cord tissue) and gastroduodenostomy



was performed [Fig. 3]. Post-operative recovery was uneventful. Oral feeding was started on fifth day post-op and was well tolerated. The baby passed stool from sixth post-operative day onwards. Within the first two weeks after surgery, the epidermal lesions started to heal [Fig. 4]. The baby was discharged on 22nd post-operative day on full oral feeding and remains well at 12 months of life.

Discussion :

Pyloric atresia (PA) is a rare neonatal surgical emergency with obscure aetiology and its incidence has been reported as 1 per 100,000 live births [1]. Three anatomical subgroups of PA consist of (I) pyloric membrane alone, (II) obstructed pyloric canal by solid cord tissue, (III) atrophic pylorus with a gap between stomach and duodenum [4]. Sencan, A et al. mentions that PA is also associated with EB and/ or aplasia cutis congenitalis, as seen in our patient [5].

The association of PA and EB was first described by Swinburne and Kohler in 1968 [6]. Both PA and EB are very rare and an autosomal recessive entities. Current evidence-base molecular evaluation suggest that EB-PA association is a hemidesmosomal variant [7]. Genetic associations have been linked to mutation in three genes: ITGB4(beta 4 integrin) in 80%, ITGA6(alpha 6 integrin) in 5% and PLECI(plectin) in 15% [8].

EB represents a heterogeneous group of rare inherited connective tissue disorders, characterised fragility of the skin and mucous membrane, which manifest by spontaneous or minimal traumatic blister formation. Three major varieties of EB have been identified depending on the ultrastructural level of basement membrane cleavage responsible for blister formation: EB simplex (intraepidermal separation), junctional epidermolysis bullosa (JEB: skin separation in the lamina lucida or central basement membrane zone [BMZ]) and dystrophic epidermolysis bullosa [9]. All three variants of EB, simplex, junctional and dystrophic, have been associated with PA but most common association is junctional epidermolysis bullosa. EB simplex was associated in our case, which is also a rare entity.

Neonates with PA usually present with non-bilious vomiting soon after birth. Plain radiographs of the abdomen confirm diagnosis of PA, which shows dilated stomach with the typical single gas bubble appearance and a paucity of gas distally as seen in our case [4].

The treatment of PA is surgical and the approach depends upon the anatomical subgroups. The treatment choice for pyloric membrane (type I) is excision of

membrane and Heineke-Mikulicz pyloroplasty or Finney [1,10], while type II or type III, end-to-end or the diamond shaped (Kimura method) gastroduodenostomy is preferred. Our case was a PA type II variety and gastroduodenostomy was performed.

We emphasize on early diagnosis and prompt treatment of pyloric atresia, together with current neonatal supportive care and nutritional support have significantly improved the survival rate in these patients.

Figure. I : a. Black arrow shows preoperative single peeled skin lesion over the right lower abdomen and b. White arrows shows post-operatively continued new blister formation with peeled skin lesions development over both extremities.



Figure. 2 : Shows pre-operative air contrast X-ray abdomen with single gastric air bubble and a paucity of air distally to stomach.



Figure. 3 : White arrow shows Pyloric Atresia type II (obstructed pyloric canal by long solid cord tissue), black arrow shows gastric antrum and brick arrow shows first part of duodenum.



Figure. 4 : Shows post-operative healing of epidermal skin lesions.



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Case Report

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Prune Belly syndrome: A Case Report

ABSTRACT

Prune belly syndrome is a rare condition characterized by the triad of 3 major features: deficiency of abdominal muscles, hydroureteronephrosis and cryptorchidism. While the unusual prune like appearance of the abdomen is the hallmark that usually identifies these patients, their underlying renal function is the most important factor in determining their overall survival. We are reporting a 2 year old male child presenting with abdominal wall laxity, bilateral cryptorchidism, and increased frequency of micturition. USG abdomen revealed bilateral hydroureteronephrosis and empty scrotal sacs, while MCUG showed pine cone shaped bladder and dilated, tortuous ureters. Staged surgical reconstruction was carried out successfully. The patient has been doing well and is continent.

Key Words : Prune belly syndrome; cryptorchidism; hydroureteronephrosis; urosepsis.

INTRODUCTION :

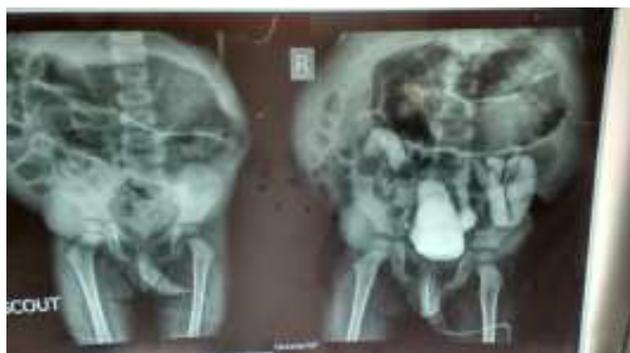
Prune Belly syndrome (PBS) is a rare malformation disorder (1 in 30,000 - 1 in 50,000 live births), affecting almost exclusively males (>95%), and characterized by a triad of clinical features including urinary tract anomalies, abdominal wall deficiency and bilateral cryptorchidism. The protruding hypoplastic abdominal wall looks like a dried prune, hence the name Prune Belly[1]. While the unusual "prunelike" appearance of the abdomen is the hallmark that usually identifies these patients, their underlying renal function is the most important factor in determining their overall survival.

CASE REPORT:

A 2 year old male child presented with laxity of the abdomen since birth and increased frequency of micturition especially during the day since 6 months. These complaints were accompanied by on and off low-grade fever since 3 months which was relieved by medications. Appetite was good and bowel habit was normal. Abdominal examination revealed a distended abdomen with thin wrinkled skin. Scrotal sacs were empty. Other systemic examination revealed no abnormality. Ultrasonography of abdomen revealed bilateral hydroureteronephrosis and both testes were located intraabdominally. Micturating cystourethrography (MCUG) showed pine cone shaped bladder, with bilateral grossly dilated and tortuous ureters with Grade V vesicoureteric reflux (VUR). (Fig.1)



Fig. 1 : MCUG showing pine cone shaped bladder with bilateral megaureters



In view of urosepsis, bilateral low loop cutaneous ureterostomies with right sided stage 1 Fowler Stephen's orchiopexy was done (Fig. 2). The child improved following this and he was discharged a week later. 6 months later, a cystoscopy was done to rule out any bladder outlet obstruction, followed by bilateral Cohen's cross trigonal ureteric reimplantation. Bilateral orchiopexy (right side 2nd stage Fowler Stephens) was done in the same sitting (Fig. 3). The patient had uneventful recovery and was discharged 15 days later. The patient was put on chemoprophylaxis and has been doing well for last 6 months.

Fig. 2 : The child with bilateral low loop cutaneous ureterostomies were performed.



Fig. 3 : Both ureters were dissected out, trimmed and cross trigonal re-implantation was performed.



Discussion:

Although PBS (also known as Eagle Barrett syndrome) is characterized by the classical triad of urinary tract anomalies, deficient abdominal musculature, and bilateral cryptorchidism, association with other anomalies including musculoskeletal, cardiovascular, pulmonary and genital malformations have been reported in the literature [2-5]. The aetiology of PBS is unclear and possible familial genetic inheritance was reported in some of studies [6-8].

Although the primary molecular defect underlying PBS remains unclear, clinical studies have given rise to two main pathogenic hypotheses? mesodermal defect hypothesis and the urethral obstruction malformation complex hypothesis. According to mesodermal defect hypothesis aberrant development of the derivatives of the first lumbar myotome in early gestation leads to a patchy muscular deficiency or hypoplasia of the abdominal wall as well as to urinary tract abnormalities [9]. The second hypothesis proposes that pressure atrophy of the abdominal wall muscles occurs when urethral obstruction leads to massive distension of the bladder and ureters [10]. Bladder distension would also interfere with descent of the testes and thus be responsible for the bilateral cryptorchidism. PBS is rare in females, with fewer than 30 cases reported in the literature [11]. More complex morphogenesis of the male urethra may be the possible cause of obstructive anomalies at several levels indicating the higher incidence of this syndrome in males. According to the classification of Woodard, the present case falls on category II (Table 1) [12].