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CONTENTS

Original Article

Prevalence of abdominal aorta aneurysm and associated risk factors in Abha city, Saudi Arabia

Abdullah Alhaizaey, Ahmed Azazy, Mohammed Asiry, Mohammed Alsayed, Mustafa Abbass, Abdulrhman Hassan, Ahmed Gamil, Musaed Alghamdi

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Abstract

Objective: The objective was to assess the prevalence and risk factors of abdominal aorta aneurysms (AAAs) in the general population.

Materials and Methods: We carried out a prospective, interventional study with patients aged over 60 years screened in the Asir Central Hospital Vascular Department from March 2017 to March 2018. Ultrasound was used to AAA screening. The maximal anteroposterior (AP) and transverse (LL) diameters of the suprarenal and infrarenal aorta were measured in each patient. AAA was defined as aortic dilatation >29 mm in the AP or LL plane. All cases with an aortic diameter >25 mm were included in the study.

Results: Our study included 701 patients (531 male, 170 female; age 60–102 years). Most were Saudi nationals (87.6%). There were some smokers (1.3%), 277 (39.5%) had diabetes mellitus, and 233 (31.8%) had hypertension. Fifty-one percent of patient had ischemic heart disease (7.3%), and 13.4% had hypercholesterolemia. Patients were classified into three groups: normal aortic size of 657 patients (93.7%); aortic ectasia 26–29 mm of 24 patients (3.4%); and AAA ≥30 mm of 20 patients (2.9%). The overall prevalence of AAA was ≥30 mm (2.9%) and there is significant relation with hypertension ($P < 0.05$).

Conclusion: Asymptomatic AAA is prevalent in our area. We may need to implement a regular screening program for men aged >60 years, especially high-risk patients to reduce AAA rupture, emergency AAA repair, and mortality.

Keywords: Abdominal aortic aneurysm, prevalence, risk factor, screening, ultrasound

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INTRODUCTION

Abdominal aorta aneurysm (AAA) is a dilated aorta 1.5 times more than normal aorta at the level of the renal arteries, or an AAA is diagnosed when the aortic diameter exceeds 3.0 cm.[1]

The risk of AAAs increases dramatically in the presence of the following factors: age ≥60 years, hypertension, and smoking. The likelihood that an aneurysm will rupture depends on aneurysm size, continued smoking, expansion rate, and persistent hypertension. Most of AAAs discovered incidentally while the patient underwent radiological investigations due to other purposes.[2]

The prevalence of AAA has been increasing for the past two decades, which possibly correlates to the increased average life span and development of diagnostic tools and screening programs.[3] The prevalence of AAA is 1%–7%

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in Western population[4] and 5% in men over 65 years of age.[5] A ruptured AAA can be fatal; therefore, a screening program is recommended for populations at increased risk.

Epidemiological studies of AAA have shown that the annual incidence of new AAA diagnoses is approximately 0.4%–0.67% in Western population, despite the evolution of our understanding and treatment of AAA in the past few decades, it continues to be a major threat to health because it has an overall mortality of 80% in the event of rupture.[6] Early identification of patients with AAA and offer of timely elective repair remains to be the most reliable strategy for prevention of death from ruptured AAA.[7]

Ultrasonography is accepted as the standard screening method for AAA because it has a high sensitivity (94%–100%) and specificity (98%–100%) and has no radiation exposure.

Surgical repair and endovascular aneurysm repair have similar outcomes, but endovascular repair is lower perioperative mortality and less invasive than surgical repair.[8]

This study was designed to assess the prevalence of AAA in Abha, Saudi Arabia. In addition, the study aimed to define risk factors associated with high prevalence of the disease, to provide information concerning which subset of individuals from the population could benefit from screening.

Statistical analysis

Statistical analysis was done using IBM SPSS software version 20 Description of quantitative data was done by mean and standard deviation (SD), and for qualitative data, percentage was used. Chi-square test was used to compare qualitative data between groups, and *t*-test was used to compare quantitative data between groups. Odds ratio (OR) with 95% confidence interval (CI) was used to describe degree of association between variables.

MATERIALS AND METHODS

We carried out a prospective, interventional study in which participants were screened in the Asir Central Hospital Vascular Department using ultrasound to screen for AAA from March 2017 to March 2018.

Males and females aged ≥60 years were included in the study. The study was explained to the participants; those who agreed to participate were first interviewed according to a survey that covered medical history, current therapy, smoking, and basic clinical data (blood pressure, heart rate, body weight, and height); any required additional tests were ordered accordingly.

Abdominal ultrasound scan was performed on all patients by a specialized radiologist from our hospital using B-mode ultrasonography machine. The maximal anteroposterior (AP) and transverse (LL) diameters of the suprarenal and infrarenal aorta were measured in each patient. AAA was defined as any aortic dilatation >29 mm in the AP or the LL plane. All cases with an aortic diameter >25 mm were registered. Ultrasound surveillance for patients with AAAs can be done regularly every 12 months for aneurysm 3–4 cm, every 6 months for aneurysm 4–4.5 cm, and every 3 months for aneurysm >4–5 cm. The aneurysms reaching a diameter of 5.5 cm in male and 5 cm in female, aneurysms with an expansion rate >5 mm in 6 months, and symptomatic patients were evaluated for management with computed tomography with arterial phase contrast.

RESULTS

A total of 701 patients (531 males and 170 females) were included in the study. Their ages ranged from 60 to 102 years with a mean ± SD (68.1 ± 10 years). The majority of them were Saudi nationals (87.6%). Few were smokers (1.3%), 277 (39.5%) had a history of diabetes mellitus (DM), and 233 (31.8%) had a history of hypertension. Ischemic heart disease (IHD) was present in 51 patients (7.3%), and 13.4% of the patients had hypercholesterolemia [Table 1].

Based on the abdominal ultrasound results, patients were classified into three groups: normal aortic size of 657 patients (93.7%), aortic ectasia 26–29 mm of 24 patients (3.4%), and AAA ≥ 30 mm of 20 patients (2.9%). The AAA prevalence was 3.5% among females and 2.8% among males [Table 1].

Among the risk factors for AAA, hypertension was significantly related to the presence of AAA ($P < 0.05$; OR: 3.3; 95% CI: 1.3–8.3). Among patients with AAA, 60% were hypertensive compared to 30% of the normal participants. Other risk factors such as age, gender, nationality, smoking, DM, hypercholesterolemia, IHD, history of vascular disease, medications, and family history of AAA were not significantly related to the presence of AAA [Table 2].

Table 1: Patient characteristics (*n*=701)

	Count (%)
Gender	
Female	170 (24.3)
Male	531 (75.7)
Nationality	
Saudi	584 (87.6)
Egyptian	28 (4.2)
Yemini	28 (4.2)
Syrian	15 (2.2)
India	12 (1.8)
Nationality	
Saudi	584 (87.6)
Non-Saudi	87 (12.4)
Aortic diameter (mm)	
AAA (≥ 30)	20 (2.9)
Ectasia (26-29)	24 (3.4)
Normal (<26)	657 (93.7)
Smoker	9 (1.3)
Diabetic	277 (39.5)
Hypertensive	223 (31.8)
Hypercholesterolemia	94 (13.4)
IHD	51 (7.3)
Medications	3 (0.4)
Vascular disease	10 (1.4)
Family history	4 (0.6)

AAA: Abdominal aorta aneurysm, IHD: Ischemic heart disease

Table 2: Risk factors for abdominal aorta aneurysm

	AAA, count (%)		*P**	OR (95% CI)
	No AAA (*n*=681)	AAA (*n*=20)		
Gender			>0.05	-
Female	164 (24.1)	6 (30)		
Male	517 (75.9)	14 (70)		
Nationality				
Saudi	565 (87.3)	19 (95.0)	>0.05	-
Egyptian	27 (4.2)	1 (5.0)		
Yemini	28 (4.3)	0 (0.0)		
Syrian	15 (2.3)	0 (0.0)		
India	12 (1.9)	0 (0.0)		
Nationality				-
Saudi	565 (87.3)	19 (95.0)	>0.05	
Non-Saudi	82 (12.7)	1 (5.0)		
Smoker	9 (1.3)	0 (0.0)	>0.05	-
Diabetic	269 (39.5)	8 (40.0)	>0.05	-
Hypertensive	211 (31.0)	12 (60.0)	<0.01	3.3 (1.3-8.3)
Hypercholesterolemia	93 (13.7)	1 (5.0)	>0.05	-
IHD	51 (7.5)	0 (0.0)	>0.05	-
Medications	3 (0.4)	0 (0.0)	>0.05	-
Vascular disease	10 (1.5)	0 (0.0)	>0.05	-
Family history	4 (0.6)	0 (0.0)	>0.05	-
Age (years)**, mean±SD	68.1±10.0	68.4±10.3	>0.05	-

*Test of significance is Chi-square test, **Test of significance is *t*-test.
SD: Standard deviation, AAA: Abdominal aorta aneurysm,
IHD: Ischemic heart disease, OR: Odds ratio, CI: Confidence interval

DISCUSSION

AAA screening decrease mortality due to AAA by 4/1000.[9-12] Moreover, in comparison of AAA screening with breast cancer and colorectal cancer screening program, it showed decrease mortality by 0.7/1000, whereas decrease mortality was 1.5/1000.[13]

Despite this, as the national AAA screening program continues, the issue of a lower rate of AAA incidence persists, as does the question of targeting resources to improve yield and therefore success.[12]

This study indicates a shortfall in uptake of screening in Abha, Saudi Arabia, among different races of both males and females aged ≥60 years. Results have shown a significant difference in the incidence of AAA between different age groups, patients with no risk factors and those with at least one cardiovascular, family, or lifestyle risk factor.

Our prevalence data are in keeping with those of that of literature. Our overall prevalence for AAA ≥ 30 mm (2.9%) is remarkably similar to that seen in the study by Mani *et al*.[14]

There has been considerable interest in the long-term treatment of individuals with nonaneurysmal aortic diameters at screening. In multicenter aneurysm screening study, the rate of ruptured AAAs appeared to start increasing at approximately 8 years after baseline screening.[11]

A multicenter study found that 26% of men and women with subaneurysmal aortic diameters of 2.5–2.9 cm developed an AAA larger than 5.4 cm in diameter within 10 years.[15] Together, these studies suggest that men with aortic diameters in the 2.5 -to 2.9 cm range are at risk of incident AAAs although the benefit of surveillance of these men remains to be established. Interestingly, aortic diameters larger than 2.5 cm in men are also associated with future (nonaneurysmal) cardiovascular events.[16]

Age has a dramatic effect on the incidence of AAA. In Rochester, USA, AAA incidence was essentially zero in individuals below the age of 49 years, increasing from 2.1 in the 40–49-year age group to 2.83 in those over 80 years of age.[17] This was clear in our study, where the mean age was 68.1 years.

Men have 4–5 times risk chance to get AAAs in comparison with women according population-based studies.[18] Surprisingly in our study, AAA prevalence was 3.5% among females and 2.8% among males [Table 1]. This pattern should be reviewed because our results were similar to those of a screening study performed in the Jeddah area by al-Zahrani *et al*.[19]

The absence of a relationship between hypertension and AAA is consistent with data from several previous

studies[20,21] whereas others found hypertension to be associated with AAA.[22,23] In the large Aneurysm Detection and Management (ADAM) Study,[24] hypertension was only marginally associated with AAA; in our study, we found a strong correlation between hypertension and AAA.

Unruptured aneurysms that are not repaired often gradually enlarge. The majority of aneurysms discovered in screening are small and do not require surgical repair, but need a regular surveillance. The risk of rupture generally increases as the diameter of the aneurysm increases. According to results of two recently published large AAA screening trials,[25] the U.K. Small Aneurysm Trial and the ADAM study, the rupture risk of aneurysms that were 4.0–5.5 cm in diameter was 1.0% and 0.5%/year, respectively. In comparison, other older studies reported an annual rupture risk of 3.4% for aneurysms that were 5.0–5.9 cm in diameter.[26] This necessitates regular surveillance, which was done in our cases.

CONCLUSION

The results of our screening study show a prevalence of asymptomatic AAA in a general population in our area, which is comparable to that of similar series carried out in other countries. We also conclude that implementation of a regular screening program for males aged above 60 years, especially high-risk patients, is required.

AAA screening program will help in the avoidance of AAA rupture, emergency AAA procedures, and death within 30 days of an AAA procedure and resulted in a significant decrease of morbidity and mortality for such diseases.

Recommendation

Decreasing of AAA-specific mortality rate after applying of the screening program may also be affected by other factors, including increased use of endovascular repair, falling prevalence of the disease, better perioperative outcome, and increased life expectancy.

However, despite a falling prevalence, contemporary AAA screening in men remains cost-effective because of counterbalancing the lower prevalence with improved surgical outcome and increased longevity.

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Conflicts of interest

There are no conflicts of interest.

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Original Article

Experiences of choledochal cyst in pediatric and adult population: A case series

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Abstract

Introduction: Choledochal cysts are a relatively uncommon disease of the bile duct which is the cystic dilatation of the intra- or extrahepatic bile ducts. Literature is scarce on its clinicopathological behavior among children and adults and its comparison.

Methods: The retrospective study was aimed to study the clinical characteristics, management, and complications between pediatric and adult patients with choledochal cysts.

Results: There was higher female preponderance with male to female ratio of 3:17 in pediatric age group while 1:4 in adult age. Abdominal pain was more common among adults, while palpable mass was more common among pediatrics population ($P < 0.05$). Jaundice was more evident in the pediatric age group, yet the classic triad of choledochal cyst (abdominal pain, jaundice, and a palpable mass) was not observed in any age group. About 76% of the cysts were type 1 cysts, which was more common among pediatric age group (40% vs. 90% $P < 0.05$) while with adults presented more with a type IVA cyst (60% vs. 10% $P > 0.05$). No patients with type II, type III, type IVB cysts, or type V were found. Sixteen patients underwent Lilly technique, with resection of the choledochal mucosa and Roux-en-Y hepaticojejunostomy, while nine patients underwent resection of the choledochal cyst and Roux-en-Y hepaticojejunostomy. Patients who received total excision had fewer surgical complications in both groups.

Conclusion: Although there is a significant difference in the clinical characteristics of choledochal cysts between children and adults, yet early detection and necessary surgery is essential for patients with choledochal cysts.

Keywords: Adult population, choledochal cyst, comparison, outcomes, pediatric

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INTRODUCTION

Choledochal cysts are congenital conditions involving cystic dilatation of bile ducts. It is a fairly uncommon anomaly of the biliary system.[1]

Its incidence is 1:100,000–1:150,000 live births in the West while it is 1:1000 in Asia with two-third of cases reported from Japan[2] with female preponderance.[3]

This surgical illness usually manifests with classic triad of intermittent abdominal pain, jaundice, and a right upper

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quadrant abdominal mass. It usually leads to obstruction of the bile ducts and retention of bile. Although it is usually found in infants and children yet can go undiagnosed and may manifest in adulthood, about 60% of cases diagnosed in the first decade of life.

The importance of choledochal cysts lies in the lethal complications such as biliary stasis, cholangitis, cholelithiasis, pancreatitis,[3-6] and malignant transformation.[7-11] Even with adequate surgical treatment, long-term complications develop and include anastomotic stricture, cholangitis, biliary cirrhosis, and biliary tract malignancy.[4,12,13]

To date, complete excision of the cyst and bilioenteric anastomosis is the treatment of choice.[14] with various types of drainage procedures such as Roux-en-Y hepaticojejunostomy, hepaticoduodenostomy, and jejunal interposition hepaticoduodenostomy.[15] However, laparoscopic excision of choledochal cyst and hepaticoduodenostomy is advised.

The lower incidence of this disease among the adults in western literature and variability in the clinical presentation and outcomes between pediatrics and adult population,[4,5] the direct comparison between the two is lacking in literature.[16] Thus, the study was aimed to identify the differences in presentation, types, management, and outcomes in both the groups.

METHODS

We conducted a retrospective analysis of 25 patients with choledochal cyst operated at Era's Lucknow Medical College and Hospital (ELMCH) from January 2011 to December 2016. The study was ethically approved by institutional review board in accordance with the ethical standards of the responsible committee on human experimentation (institutional or regional) and with the Helsinki Declaration of 1975, as revised in 2000.

For inclusion in the study, we considered patients of both genders, aged between 6 months and 60 years, whose surgery was performed at the Department of Surgery of ELMCH and attending at least three visits postoperatively.

We analyzed the demographic data, presenting complaints, age at which the diagnosis was made, applied surgical procedures, complications, anatomical classification of cysts, and postoperative outcomes.

Of the 25 cases identified, all patients had complete treatment and follow-up records for analysis. All patients received surgical treatment, Lilly technique, with resection of the choledochal mucosa and Roux-en-Y hepaticojejunostomy in 16 while in another 9 patients, resection of the choledochal cyst and Roux-en-Y hepaticojejunostomy was carried.

Statistical analysis of data was performed using Statistical package for social sciences, version-23 (SPSS-23, IBM, Chicago, USA) and MedCalc statistical software were used for data analysis. Statistical significance was set at $P < 0.05$.

RESULTS

Among the 25 patients, male to female ratio was 3:17 in the pediatric age group, whereas 1:4 in adult age group with higher female predominance in both the groups. Majority of the cases were found among females <18 years of age with 75% of patients being <10 years.

Clinical symptoms, operative findings, and the Todani classification are shwon in Tables 1 and 2.

Clinical symptoms

In the pediatric age group, the most common clinical manifestation was abdominal pain in 95% of cases (19 patients), followed by jaundice, present in 30% of patients (6 cases). Five patients had a palpable abdominal mass (25%).

In adults, pain abdomen was common presentation among all the patients (100%). Only one patient had palpable abdominal lump, while none had jaundice. Thus, abdominal pain was more common among adults while palpable mass was more common among pediatrics population ($P < 0.05$).

Acute cholangitis was present in 1 child (5%).

The classic triad of choledochal cyst (abdominal pain, jaundice, and a palpable mass) was not observed in any age group.

Table 1: Patient characteristics

Total	Pediatrics (*n*=20)	Adults (*n*=5)	*P*
Mean Age	4.2 years	38.5 years	<0.05
Sex	3:17	1:4	<0.05
Male:Female			
Presentation			
Abdominal pain	19	5	<0.05
Abdominal mass	5	1	NS
Jaundice	6	0	<0.05
Associated complications			
Cholangitis	1	0	<0.05
Pancreatitis	0	0	NS
Biliary malignancy	0	0	NS
Biliary stricture	0	0	NS

Table 2: Cysts types

Total Todani's 's classification	Pediatrics (*n*=20)	Adult (*n*=5)	*P*
Type 1a	10	1	<0.05
Type 1b	0	0	NS
Type 1c	8	0	<0.05
Type IVa	2	3	NS
Type V	0	0	NS

Ultrasonography (USG) of the abdomen was done in all patients, followed by contrast-enhanced computed tomography (CT).

The mean interval between diagnosis and surgery was 3 weeks.

Cyst type

About 76% of the cysts were type 1 cysts with adults presented with type l less commonly (40% vs. 90% $P < 0.05$). No patients with Type II, Type III, Type IVB cysts, or Type V were found. Adults were more likely to present with a Type IVA cyst (60% vs. 10% $P > 0.05$) [Table 2].

Surgical treatment

In 16 patients, we applied the Lilly technique, with resection of the choledochal mucosa and Roux-en-Y hepaticojejunostomy. In nine patients, we performed resection of the choledochal cyst and Roux-en-Y hepaticojejunostomy. All adult patients underwent choledochal cyst resection and Roux-en-Y hepaticojejunostomy without exception.

Complications

Two patients (10%) had immediate postoperative complications, one in pediatric age group and one in adult age group, both having a biliary leak from the bilioenteric anastomosis, which were managed successfully by conservative means.

Two patients (10%) both in adult age group developed acute cholangitis out of which one was managed successfully by IV antibiotics and supportive care while other patients expired.

One patient died of postoperative complication after developing acute severe cholangitis.

All patients had a grossly normal-appearing liver at laparotomy.

None of the excised cysts had malignant degeneration or metaplasia at histopathology.

All patients received antibiotic cover for 1 month and ursodeoxycholic acid for 3 months.

The mean duration of postoperative follow-up was 30 months.

All patients showed clinical improvement, with no evidence of jaundice, biliary stricture, or any malignant transformation.

DISCUSSION

Choledochal cysts are congenital malformations of bile ducts that represent a major diagnostic and therapeutic challenge for the surgeon, demanding a high degree of suspicion and efficient etiological investigation for the correct diagnosis and institution of appropriate treatment.[17]

The treatment should consist of resection of the dilated portion of the extrahepatic biliary tree with reconstruction with a Roux-en-Y hepaticojejunostomy. When intrahepatic dilatation is very extensive, liver transplantation is an alternative.

We report our experience about the clinicopathological differences between pediatric and adult patients with choledochal cysts in our institute. The classic triad of jaundice, abdominal pain, and abdominal mass was often seen in pediatric patients than in adults in the previous study[14,15] was not evident in our study.

There is no consensus in the literature about the most common clinical presentation. Some authors believe abdominal pain to be the main signal, similar to the result obtained in this sample. Other studies, however, report that jaundice is the most prevalent symptom.[18,19]

We observed more of abdominal pain as the chief complaints in children and adults while abdominal mass of classical presentation more among children. Jaundice was more evident among children, as was the observation from others in literature.[16] Lilly argued that jaundice should be more commonly seen in infants and abdominal pain in older patients, probably due to the better capacity to verbalize.[19]

The observation of the predominance of female patients in this sample, consisting of 85%, is consistent with other studies on choledochal cysts.

The age at which the diagnosis was made, with the majority of patients in the first decade of life, agrees with other reports, where 80% of cases are diagnosed before 10 years of life, whereas only 20% of common bile duct cysts are seen in adulthood.

Although abdominal pain, jaundice, and abdominal mass are described as the classic triad of choledochal cyst, their association was not observed in this study, in disagreement with data reported by other authors.[10]

Although the concept that this presentation would be more frequent in children than in adults, it is believed that early diagnosis in the current era would prevent the choledochal cyst from greatly increasing in volume. None of our patients had any associated complications like stricture or stones except 1 having cholangitis. This largely depends on the type of cyst and to the duration and severity of biliary stasis.

Currently, the preoperative diagnosis of choledochal cyst is mostly done by the USG, which has high sensitivity for the diagnosis of biliary tract disease. It was the first complementary method used in all our patients and in other studies as well.[20] In all seven patients who underwent magnetic resonance imaging (MRI) the diagnosis was correctly achieved, demonstrating that this method is a more sensitive diagnostic tool for the evaluation of diseases of the biliary tract than CT [Figure 1].

Three-dimensional reconstruction of MRI cholangiopancreatography allows better anatomical evaluation of biliary tract and pancreas. Another innovation is the virtual cholangioscopy, which allows preoperative radiological exploration of the bile ducts.[12]

According to medical literature, choledochal cysts type I correspond to up to 90% of all cases, confirming the demonstrated in this series, where the majority of cysts (85%) represented a fusiform dilatation of the common bile duct. Choledochal cysts in children were predominantly type I cystic lesions (18 out of 20), whereas type IV cysts were more common in adult patients (3 out of 5).

The current treatment is total resection of the cyst since 1980s; however, associated complications and accompanying malignant disease alter the further management in terms of extension of resection, reconstruction method, and need for hepatic resection. Thus, the type of choledochal cyst becomes the most critical variable. Fortunately, our patient population were of Type 1 and Type 4 and underwent bilioenteric anastomosis associated with resection of the mucosal lining of the cyst, the Lilly technique, in 16 patients and Roux-en-Y hepaticojejunostomy was performed in 4 patients with low incidence of postoperative complication [Figures 2-4].

In this series, the low incidence of postoperative complications and good clinical outcome suggests that it is technically possible to safely perform bilioenteric anastomosis, as previously described.

CONCLUSION

Given all these data, we conclude that abdominal pain in childhood must remain a warning sign for congenital malformations of the biliary tract. Choledochal cysts in children were predominantly type I cystic lesions, whereas type IV cysts were more common in adult patients. The

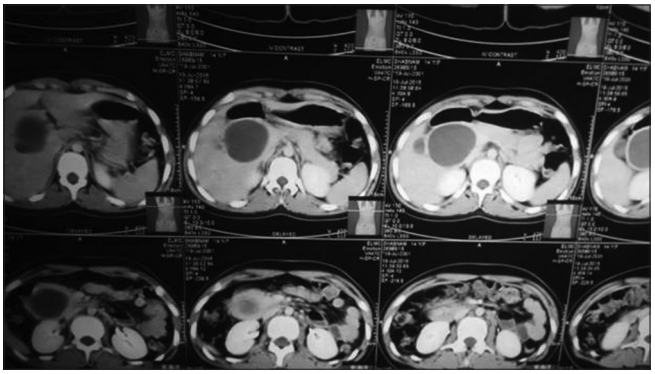

Figure 1: Contrast-computed tomography films

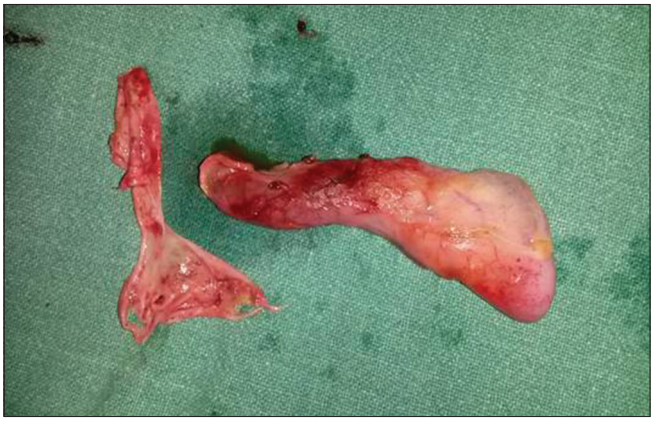

Figure 2: Postoperative specimen sample of gallbladder and cyst

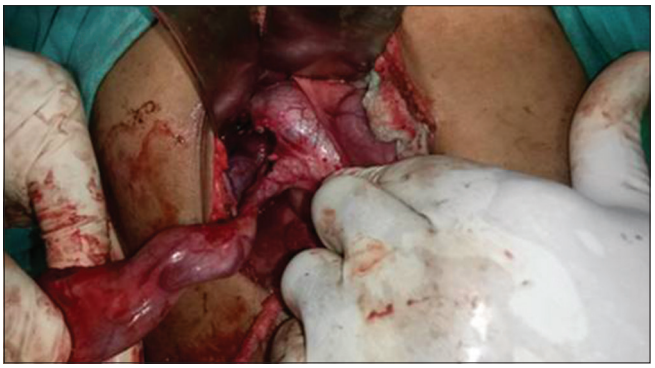

Figure 3: Intraoperative picture of gallbladder and cyst wall

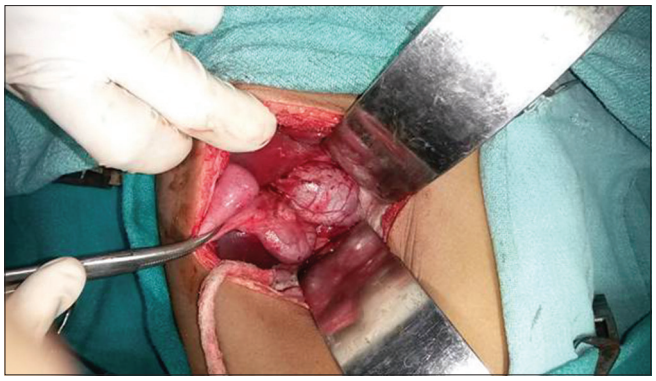

Figure 4: Intraoperative picture of cyst

surgical treatment of choledochal cysts, with resection and hepaticojejunostomy, is the treatment of choice and is safe even in young children even when symptoms are minimal. Patients who received total excision had fewer surgical complications in both groups. Diagnosis and treatment should be early to avoid a greater involvement of the hepatic parenchyma, whose severity depends on the degree of obstruction and time course.

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Conflicts of interest
There are no conflicts of interest.

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Original Article

Diagnostic accuracy of triple test in breast pathologies of women above 20 years of age

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Abstract

Background: This study is to establish the diagnostic accuracy of triple test in assessing breast pathologies in women above 20 years of age taking their histopathological report as standard.

Patients and Methods: In our analytical study, we included women presenting with a complaint of breast lump or change in breast texture in an age group of above 20 years. Systematic clinical examination was done followed by mammography and finally fine-needle aspiration cytology (FNAC) for tissue sampling. Lesions were considered triple test positive, if lesions were FNAC positive and any one of the remaining two modalities also gave a positive (malignant) interpretation, supporting FNAC, but each of three components must be negative for labeling triple test as negative. Postoperatively, cumulative results were compared with histopathology reports and statistical parameters such as specificity, sensitivity, positive predictive value, negative predictive value, and accuracy of triple test were calculated.

Results: We have obtained 100% sensitivity using triple test in all age groups when each element was interpreted as malignant and 100% specificity ($P < 0.001$) when each element was interpreted as benign with diagnostic accuracy of almost 100% in concordant cases. It was recommended that in cases, where all three modalities are not in agreement for benign pathology and in FNAC positive cases where other two parameters are not in agreement, and lesion is interpreted as suspicious on triple test, the nature of the lesion must be ascertained by excision biopsy.

Conclusion: Triple test of breast pathologies is a reliable method and allows detection of breast pathologies in an effective manner, and undue delay in treatment can be minimized by using this modality in limited resource country.

Keywords: Breast cancer, clinical examination mammography, fine-needle aspiration cytology, triple test

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INTRODUCTION

Breast (mammary gland) is a distinguishing feature of class Mammalia; from puberty to death, the breast is subjected to constant physical and physiological alteration that relates to menses, pregnancy, gestation, and menopause.

Nearly, half of the population is of females, and they are likely to suffer from diseases of breast any time after puberty. Breast problems make up to 20% of the workload of a surgical outpatient department in the UK.[1] Patients commonly present, complaining of lump in the breast, pain, and nipple discharge. Although the most common

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cause of symptoms is benign breast diseases. But as the life expectancy is increasing incidence of carcinoma breast is also increasing.

Many times, it becomes difficult to clinically differentiate between benign and malignant lesions, especially in early stages. Furthermore, cancer awareness has created phobia in minds of most of women, and they want to be sure that they are not suffering from cancer breast, as name itself is taken, by general masses to be a forebearer of death. In many countries, increasing numbers of women now undergo screening for malignant breast disease and ask for further management of their asymptomatic breast disease. Previously for clinically suspicious lesions biopsy was only reliable tool but with newer imaging and pathological evaluation techniques, the field of diagnosis has been revolutionized. However, main problem is none of the tools, for diagnosis is 100% accurate. Hence, to minimize delay in treatment and to avoid unnecessary outpatient follow-up and open biopsy, many breast clinics have evolved a policy of "triple test" with immediate reporting to provide a "One Stop diagnostic Service"[2] where patients are evaluated by history and physical examination, imaging (mammography, breast USG, and MRI) and FNAC or core needle biopsy to establish a diagnosis and management plan for each patient on the day of the clinic visit.

PATIENTS AND METHODS

This analytical study was conducted in the Department of Surgery, Ruxmaniben Deepchand Gardi Medical College and Allied Hospitals, Ujjain, in collaboration with Pathology and Radiology departments. Women presenting with breast lump or change in the nature of breast with the age above 20 years were selected. Diagnosed cases of breast abscess, antibioma, and malignancy were not included in study. We have included 420 patients in our study.

Relevant history was taken and clinical examination done. Mammography included two standard views, i.e., mediolateral oblique and a craniocaudal view. After imaging patients were evaluated by FNAC for cytology. Interpretations of clinical examination, mammography, and FNAC were tabulated as benign, malignant, and suspicious, respectively. The triple test assessment was done and interpretation drawn. Lesions were considered triple test positive if lesion was FNAC positive with any one of remaining two modalities was also positive (malignant), but each of three components must be negative for labeling triple test as negative. Finally, results were compared with histopathological reports.

Statistical calculation comprised of sensitivity, specificity, positive predictive value, negative predictive value and accuracy of triple test, for evaluating breast pathology, considering histopathology as standard.

RESULTS

The age group of patients (n = 420) observed in this series ranged from 20 to 72 years. Number of malignant cases were 138(32.8%) whereas benign cases reported were 282 (67.2%), [Table 1 and Figure 1]. With mean age at presentation of benign diseases was 29.86 years and for malignancy was 51.84 years' youngest patient who was detected malignancy was 28 years, and the oldest patient was 72 years old. The most common age group of incidence for malignancy was 36-45 years (34.69%) and the most common age group in benign pathology was 20–26 years (47.06%) [Table 2 and Figure 2].

Present study shows that clinical assessment alone was having sensitivity of 69.39% and specificity of 83.12% with overall diagnostic accuracy of 72%, results of mammography alone showed sensitivity of 62.79%, specificity of 85.13% with overall diagnostic accuracy of 75.27%, FNAC alone have given sensitivity of 94.25% and specificity of 85.92% with overall diagnostic accuracy of 78.47%. When two elements were, combined for assessment results obtained were as; combination of clinical assessment and FNAC yielded sensitivity of 96% and specificity of 86.95% with overall accuracy of 80.60%, whereas, combined assessment with clinical assessment and mammography yielded sensitivity of 65.81% and specificity of 82.86% with overall accuracy of 78.57% [Table 3 and Figure 3]. The accuracy of triple assessment in concordant cases was 100%, with overall test accuracy of 82.15%. Triple test showed a positive and negative predictive value of 100% with sensitivity of 97.17% and specificity of 86.96% which was greater than individual test or two modalities used in combination [Tables 3, 4 and Figure 3].

DISCUSSION

The accurate diagnosis of breast lump through cost-effective and less time-consuming manner should be of top priority in a country like India where health care facilities are not at par with developed world. Being the most common cause of malignancy in female's timely diagnosis and treatment is of paramount importance. The diagnosis of breast pathology can be done by various modalities namely clinical examination, radiological examination (USG, mammography, and MRI), and pathological examination (FNAC, trucut biopsy, core needle biopsy, and incisional biopsy). The accuracy of each test varies when used individually whereas the combination of all

the three modalities enhances accuracy significantly. The triple assessment or triple test which includes clinical examination, mammography, and FNAC yields superior results [Tables 5-8].

We have identified studies from the literature that addressed the same research question as this study [Tables 5-8].

Our results support the findings of other studies that combined clinical, imaging, and cytological (FNAC) assessment is diagnostically more accurate than individual or combination of two diagnostic modalities [Table 3].

In our study, the clinical assessment was found to have sensitivity of 69.39% and specificity of 83.12% with overall diagnostic accuracy of 72%. Sensitivity and specificity of clinical examination in our study is less that the most of the available literature [Table 8].

This difference in sensitivity may be due to the fact that few of the patients in our study were found to have malignant lesion in very younger age group with the youngest female being 28 years of age; and there is always an unavoidable bias toward benign pathology in younger age group.

In our study, results of imaging of breast gave sensitivity of 62.79% and specificity of 85.13% with overall diagnostic accuracy of 75.27%, which are on the lower side of the available literature [Table 7].

The reason may be breast imaging is an operator-dependent process[17] and technical team expertise can significantly change quality of film and reporting process other reason may be technically related to resolution of the machine.

FNAC results in our study have given sensitivity of 94.25% and specificity of 85.92% with overall diagnostic accuracy of 78.47%, which is in accordance with most of the available literature.

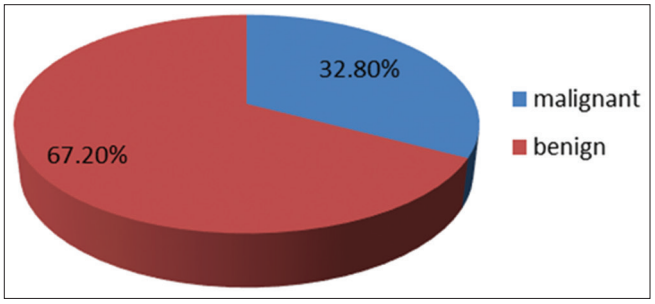

Figure 1: Pie chart showing histopathological distribution of sample

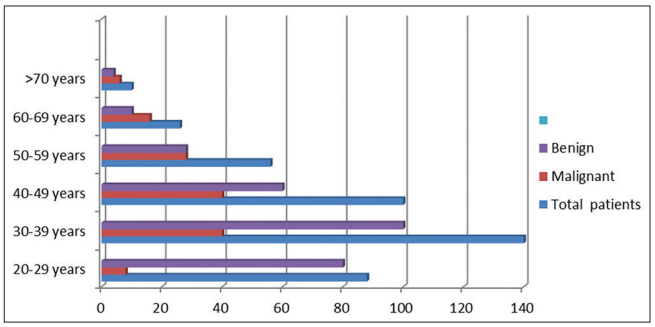

Figure 2: Bar chart for age group distribution of histopathological lesions

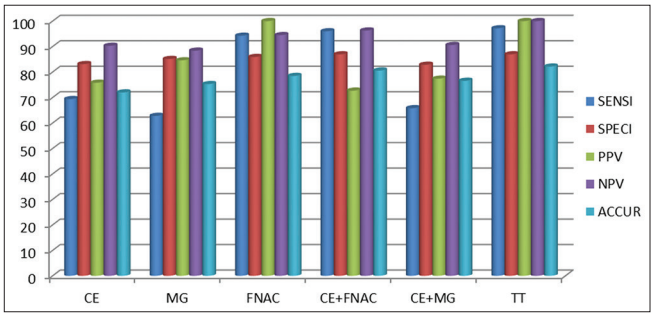

Figure 3: Graphical depiction of statistical parameters of diagnostic modalities alone and in combination. SENSI: Sensitivity, CE: Clinical examination, SPECI: Specificity, MG: Mammography, PPV: Positive predictive value, FNAC: Fine-needle aspiration cytology, NPV: Negative predictive value, TT: Triple test, ACCUR: Accuracy

Table 1: Histopathological distribution of breast masses

Malignant cases, *n* (%)	Benign cases, *n* (%)
138 (32.8)	282 (67.2)

Table 2: Histopathological distribution of breast lumps in various age groups

Age group (years)	Number of patients	Malignant cases, *n* (%)	Benign cases, *n* (%)
20-29	88	8 (9)	80 (91)
30-39	140	40 (28.5)	100 (71.5)
40-49	100	40 (40)	60 (60)
50-59	56	28 (50)	28 (50)
60-69	26	16 (61.5)	10 (38.5)
>70	10	6 (60)	4 (40)

Table 3: Statistical comparison of diagnostic modalities

Measures	CE (%)	MG (%)	FNAC (%)	CE + FNAC (%)	CE + MG (%)	TT (overall) (%)
Sensitivity	69.39	62.79	94.25	96	65.81	97.17
Specificity	83.12	85.13	85.92	86.95	82.86	86.96
PPV	75.8	84.6	100	72.7	77.4	100
NPV	90.3	88.4	94.5	96.3	90.6	100
Accuracy	72	75.27	78.47	80.60	76.57	82.15

FNAC: Fine-needle aspiration cytology, CE: Clinical examination, MG: Mammography, PPV: Positive predictive value, NPV: Negative predictive value, TT: Triple test

Table 4: Triple test result compared with histopathology in concordant and nonconcordant cases

Triple-test results	Interpretation	Number of lesions	Histopathology	
			Benign	Malignant
Concordant	Benign	270	270	
	Malignant	96		96
Nonconcordant	Suspicious	54	12	42
Total		420	282	138

Table 5: Comparative study chart for triple assessment

Authors	Sensitivity (%)	Specificity (%)	PPV (%)	NPV (%)	DA (%)
Butler *et al*.[3]	-	100	-	-	-
Morris A *et al*. (1998)[4]	100	100	100	100	100
Steinberg *et al*.[5]	95.5	100	100	-	-
Kulkarni *et al*.[6]	100	100	-	-	100
Morris KT *et al*. (2002)[7]	100	100	-	-	100
Ghafouri *et al*. (2006)[8]	63.6	94.4	100	100	100
Mokri and Guity *et al*. (2012)[9]	98	100	100	97	99
Ghimire B *et al*. (2008)[10]	100	95.2	96.7	-	98
Jan M, Mattoo JA *et al*. (2010)[11]	100	99.3	93.3	100	99.3
Bhatti and Gilani *et al*. (2010)[12]	100	100	-	-	100

NPV: Negative predictive value, PPV: Positive predictive value, DA: Diagnostic accuracy

Table 6: Comparative study chart for FNAC (cytological) assessment

Authors	Sensitivity (%)	Specificity (%)
Butler *et al*.[3]	96	66
Morris A *et al*. (1998)[4]	92	66.7
Kulkarni *et al*.[6]	86.66	94.64
Morris KT *et al*. (2002)[7]	87	80
Ghafouri *et al*. (2006)[8]	18.2	99.9
Mokri and Guity *et al*. (2012)[9]	87	86
Eltahir *et al*. (1999)[13]	88.7	99.1

Table 7: Comparative study chart for imaging assessment

Authors	Sensitivity (%)	Specificity (%)
Butler *et al*.[3]	94	73
Eltahir *et al*. (1999)[13]	93.2	96.7
Moss HA (1999)[14]	78.9	96.7
Kerlikowske K (2011)[15]	84	81.9
Rahman MZ (2011)[16]	82.76	90.36

Table 8: Comparative study chart for clinical assessment results

Authors	Sensitivity (%)	Specificity (%)
Butler *et al*.[3]	96	66
Morris A *et al*. (1998)[4]	96	100
Kulkarni *et al*.[6]	90	91.07
Morris KT *et al*. (2002)[7]	92	96
Mokri and Guity *et al*. (2012)[9]	89	90
Eltahir *et al*.[14]	93.5	98.1

In our study, combined assessment with clinical examination and FNAC yielded sensitivity of 96% and specificity of 86.95% with overall accuracy of 86.60% which is in agreement with Kulkarni *et al*.[6] sensitivity 93.33% specificity 87.50% in the view of higher diagnostic accuracy than single diagnostic modality. Morris *et al*. used a scoring system for triple test;[7] in their study, they have included 113 patients. Benign lesions were given a score of 1, suspicious lesions 2, and malignant lesions 3 on each modality. The final score was obtained by adding individual scores. Results were then compared with histopathological reports. The triple test was found to have an accuracy of 100%. They suggested that breast pathologies with score less than four are benign, those with score of five should undergo biopsy and lesions with six and higher score can undergo definitive treatment. Thus unnecessary biopsies can be avoided. Morris *et al*.[4] also supported this fact in their study on 261 female patients with breast lesions with diagnostic accuracy of 100% results were obtained by Mansoor and Zahrani.[18] Kachewar and Dongre in their study of 200 female patients obtained sensitivity of 97.44% and specificity of 100% for triple test[19] Similar results were obtained by Ghafouri *et al*.[8] and Ghimire B *et al*.[10] Kaufman *et al*. in their study of 234 patients also concluded that triple test was more sensitive (100%) and specific (57%)[17] as compared to individual test used alone, thus unnecessary biopsies could be avoided saving time and money of patient.

CONCLUSION

The triple test for assessment of breast lesions is a reliable method and allows the detection of breast pathologies in an effective manner, and undue delay in treatment can be minimized using this modality in limited resource country.

Three potential sources of error are suggested

- Interobserver variation in clinical assessment, breast imaging, and cytological analysis[20]
- Unavoidable bias toward benign pathology in younger age groups
- Histopathology was considered gold standard in this study and all other studies. However, in accuracies and possibility of human errors are always there
- In the present study, all patients presented with breast lump so our study have a limitation that no comments could be given on triple assessment of impalpable malignant/benign breast lesions.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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Original Article

Lichtenstein repair using lightweight mesh versus laparoscopic total extraperitoneal repair using polypropylene mesh in patients with inguinal hernia: A randomized study

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Abstract

Background: With the introduction of mesh for repair of inguinal hernia, the focus of surgeons has shifted to postoperative pain and quality of life (QOL). As compared to open procedures, laparoscopic procedures have been found to be associated with less pain and faster recovery. The present study was designed to assess whether this holds true when open Lichtenstein repair is done using lightweight mesh (LWM) because, in previous studies where laparoscopic inguinal hernia repair is compared to open Lichtenstein repair, heavyweight mesh (HWM) was used for both techniques. HWM was used for total extraperitoneal (TEP) in the current study because of higher recurrence associated with LWM.

Materials and Methods: This prospective randomized study was done on 60 patients divided into two groups: the Lichtenstein group and the TEP group. Patients were followed at 1 week, 1 month, and 6 months for any postoperative complication. QOL was assessed using hernia-specific Carolinas Comfort Scale.

Results: No statistically significant difference was observed between the two groups with regard to various postoperative complications. Only one recurrence was detected at 1 month in TEP group. The mean groin pain was significantly less in TEP group at 24 h, 1 week, and 1 month ($P < 0.05$). The sensation of mesh was significantly less in Lichtenstein group at 24 h and 1 week ($P \leq 0.001$) but comparable to TEP at 1 month and 6 months. The difference in movement limitation was not significant at any time between the two groups.

Conclusion: Except for less pain in the early postoperative period TEP does not offer any advantage and Lichtenstein repair using LWM can still be considered as the best option for inguinal hernia repair, especially in countries where resources are limited.

Keywords: Hernia, inguinal, Lichtenstein, quality of life, total extraperitoneal

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INTRODUCTION

Inguinal hernia surgery is one of the most commonly performed procedure by general surgeons, but the debate on ideal technique is yet to resolve. Earlier inguinal hernia repairs were assessed on the basis of recurrence. The recurrence rate associated with nonmesh repair was up to 15%. With the introduction of mesh, this recurrence rate

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dropped significantly and irrespective of the technique, open or laparoscopic, reported recurrence rate with mesh is 2%–3%.[1] Although the use of mesh has decreased the recurrence rate, it is found to be associated with increased incidence of chronic groin pain, foreign body sensation, and limitation in physical activity. The incidence of chronic pain after mesh repair varies from 10%–30% to 2%–20% of these patients experience limitations in their daily activities.[2] Therefore, more recently attention has shifted to quality of life (QOL) and postoperative pain after hernia surgery repair. Different techniques using different types of meshes were thus compared to address this issue.

In patients treated with open Lichtenstein's repair, it is found that lightweight mesh (LWM) is associated with less postoperative pain with comparable recurrence rate to heavyweight mesh (HWM).[3] LWM is thin and has large pore size as compared to conventional polyester or polypropylene HWM. LWM initiates less foreign body reactions and is found to be better biocompatible as compared to HWM.

Among the laparoscopic repairs, total extraperitoneal (TEP) repair is preferred over transabdominal preperitoneal repair (TAPP) as the peritoneal cavity is not entered; and thus, it reduces the chances of visceral injury, adhesion formation, and development of port site hernia. In terms of mesh, recent studies have shown that LWM in TEP does offer any additional benefit over HWM as far as postoperative pain and QOL is concerned, and the recurrence rate is higher with LWM. Therefore, HWM is the preferred choice for TEP repair.[4]

In comparison to laparoscopic TEP repair, Lichtenstein is found to be inferior in terms of postoperative pain and return to normal activity. However, in most of the identified randomized controlled trials (RCTs), where Lichtenstein is compared to TEP, HWM was used for both the techniques [Table 1].[5-14] The present study was, therefore, designed to find whether this also holds true when Lichtenstein using LWM is compared to laparoscopic TEP repair using HWM.

MATERIALS AND METHODS

The study was conducted in the Department of General Surgery, Government Medical College, and Hospital Chandigarh from 2016 to 2018. Patients over 18 years of age undergoing elective unilateral inguinal hernia repair were included in the study. Patients with bilateral hernia, complete hernia, recurrent hernia, strangulated hernias, prior lower abdominal surgery, and prior radiation exposure to lower abdomen were excluded from the study.

A total of 60 patients were randomized into one of the two groups: the Lichtenstein group and the TEP group. Randomization was achieved through sealed envelope technique. If a patient did not agree to the procedure after randomization, he was excluded from the study. No blinding was possible owing to different nature of the two procedures. For Lichtenstein repair, poliglecaprone 25/polypropylene LWM (Ultrapro - Ethicon) was used, and for TEP, polypropylene three-dimensional mesh (Bard) was used. A detailed information sheet was entered for each patient which included the informed written consent, demographic details, relevant history pertaining to inguinal hernia, general and local physical examination, routine hematological investigations along with electrocardiograph and chest X-ray. Details regarding the type of procedure performed, type of anesthesia used, type of mesh placed, and duration of postoperative stay were also noted down.

After discharge, patients were followed at 1 week, 1 month, and 6 months. QOL in the postoperative period was assessed by the Carolinas Comfort Scale (CCS) questionnaire, which included the sensation of mesh, pain, and movement limitation with various activities.[15] This questionnaire was filled by the patients at each follow-up. A thorough local examination was performed to look for any seroma, hematoma formation. Status of the wound was examined, and staplers were removed on the 1st follow-up. During the follow-up, patients were also examined for any testicular atrophy, mesh infection, and hernia recurrence.

CCS is a hernia-specific QOL survey. It evaluates the incidence and severity of pain, activity limitation, and mesh sensation in seven different activities, as well as pain and mesh sensation at rest. CCS scores are reported on a 6-point Likert scale – 0: No symptom, 1: Mild but not bothersome

Table 1: Lichtenstein versus total extraperitoneal trials

Trial	Lichtenstein		TEP	
	Cases (*n*)	Mesh	Cases (*n*)	Mesh
Heikkinen *et al* ., 1998[5]	23	PPM	22	PPM
Lal *et al* ., 2003[6]	25	PPM	25	PPM
Gokalp *et al* ., 2003[7]	62	PPM	61	PPM
Andersson *et al* ., 2003[8]	87	PPM	81	PPM
Lau *et al* ., 2006[9]	100	PPM	100	PPM
Eklund *et al* ., 2006[10]	706	PPM	665	PPM
Vidović *et al* ., 2007[11]	233	PPM	112	PPM
Langeveld *et al* ., 2010[12]	324	PPM	336	PPM
Wang *et al* ., 2013[13]	84	Vypro	84	Vypro
Dhankhar *et al* ., 2014[14]	30	PPM	29	PPM

TEP: Total extraperitoneal, PPM: Polypropylene (HWM), Vypro: Polypropylene with polyglactin (LWM), HWM: Heavyweight mesh, LWM: Lightweight mesh

symptoms, 2: Mild and bothersome symptoms, 3: Moderate and/daily symptoms, 4: Severe symptoms, and 5: Disabling Symptoms. Maximum scores of 0 (none) or 1 (minimal and not bothersome) were classified as asymptomatic, whereas scores of 2 (minimal but bothersome) or higher were considered symptomatic. To compare each QOL domain, the mean score from all activities corresponding to that domain is calculated. Then the overall mean for a domain is calculated separately for both the groups at various follow-up visits and compared.

Descriptive statistics were used. Data were expressed in terms of mean ± standard deviation. Differences between two groups were determined by Mann–Whitney test and Wilcoxon-signed ranks test. All $P < 0.05$ was considered statistically significant. The statistical analysis was carried out using IBM Corp. Released 2013. IBM SPSS Statistics for Windows, Version 22.0. (IBM Corp., Armonk, NY).

RESULTS

There was no significant difference in patient and disease characteristics at the time of operation [Table 2]. The mean hospital stay after TEP was 1.56 (range 1–2 days). Nineteen patients were discharged on postoperative day 1 and 11 were discharged after 2nd postoperative day. The mean hospital stay after open repair was 1.46 days (range 1–2 days). Twenty-two patients were discharged on postoperative day 1 and 8 were discharged on the 2nd postoperative day. There was no significant difference in hospital stay between the two groups [Table 3].

The peritoneal breach was the only intraoperative complication that occurred in 3 (10%) patients in TEP group. Of the three patients, two were asymptomatic and were discharged at 48 h postoperatively. One patient who was discharged at 48 h came back with the symptoms of abdominal distension and inability to pass stools and flatus on the 5-postoperative day. Abdominal X rays showed multiple air-fluid levels suggesting acute intestinal obstruction [Figure 1]. He was kept nil per oral and Ryle's tube drainage, but symptoms did not relieve. On exploratory laparotomy, loop of bowel was found herniating through the peritoneal defect. No intraoperative complication was seen in the Lichtenstein group.

No statistically significant difference was observed between the two groups with regard to various postoperative complications such as seroma, hematoma, wound infection, and testicular atrophy at follow-up visits. However, one recurrence was detected at 1 month in TEP group.

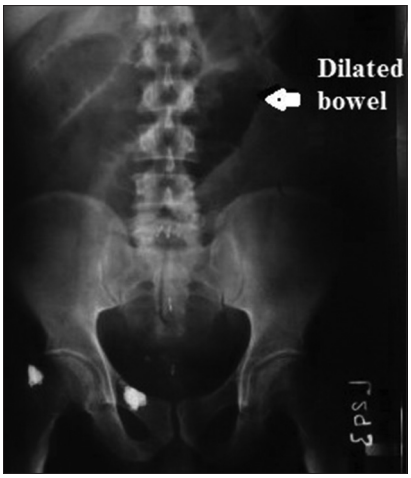

Figure 1: Plain X-ray of total extraperitoneal patient showing multiple air-fluid levels

Table 2: Baseline characteristics

	TEP (30)	Lichtenstein (30)
Age (mean±SD)	51.47±11.27	45.43±19.37
Duration of symptoms >1 year, *n* (%)	13 (43.3)	17 (56.7)
Hernia characteristics, *n* (%)		
Right sided	18 (60.0)	20 (66.7)
Direct	14 (46.7)	9 (30)
Direct + indirect	0	2 (6.6)

SD: Standard deviation, TEP: Total extra peritoneal

Quality of life

QOL was assessed using CCS. Of 30 patients included in TEP using HWM, 2 were excluded from the study as 1 patient developed intestinal obstruction on 5th postoperative day and underwent emergency laparotomy and the other had hernia recurrence at 1 month. Further, one patient in Lichtenstein using LWM did not respond after 1 month and one patient in TEP using HWM did not responded at 6-month period. Only 23 patients who underwent TEP and 22 patients who underwent Lichtenstein repair could respond to "sensation of mesh" questionnaire as others were unable to understand what is being asked.

The mean groin pain at any point of time was less in TEP group than Liechtenstein group. The difference was significant at 24 h and 1 week and 1 month. The sensation of mesh was less in Lichtenstein using LWM as compared to TEP using HWM. This was statistically significant at 24 h and 1 week postoperatively. Movement limitation in TEP using HWM was less than Lichtenstein using LWM. This was not statistically significant at any time. The overall QOL was better in TEP using HWM than Lichtenstein using LWM. This was statistically significant only at 1-week postoperative period [Table 4 and Figure 2].

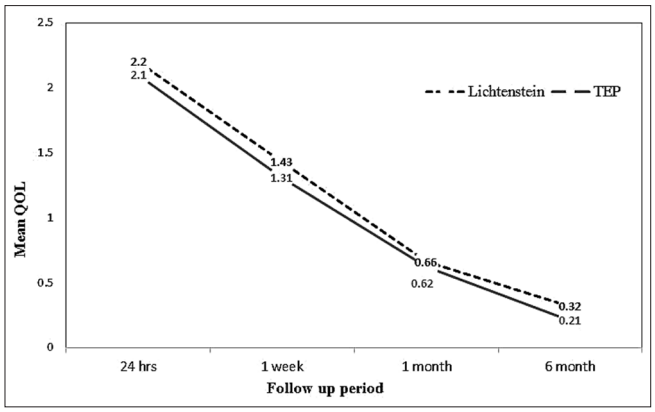

Figure 2: Overall quality of life on Carolinas Comfort Scale

Table 3: Postoperative hospital stay

Time at discharge (h)	Lichtenstein group, *n* (%)	TEP group, *n* (%)	*P*
24	22 (73.3)	19 (63.3)	0.40
48	8 (26.7)	11 (36.7)	0.40

TEP: Total extraperitoneal

Table 4: Carolinas Comfort Scale outcome

Time	Lichtenstein group	TEP group	*P*
Mean±SD for groin pain			
24 h	2.58±0.398	2.04±0.413	0.000
1 week	1.76±0.303	1.10±0.379	0.000
1 month	0.878±1.01	0.441±0.482	0.043
6 months	0.443±0.928	0.140±0.253	0.100
Mean±SD for the sensation of mesh			
24 h	1.97±0.321	2.248±0.296	0.001
1 week	1.427±0.272	1.76±0.315	0.000
1 month	0.819±1.58	1.19±0.374	0.228
6 months	0.618±1.60	0.622±1.47	0.991
Mean±SD for movement limitation			
24 h	2.06±0.275	2.01±0.332	0.556
1 week	1.10±0.369	1.08±0.197	0.789
1 month	0.286±0.240	0.281±0.180	0.940
6 months	0.167±0.249	0.128±0.180	0.499
Mean±SD for QOL			
24 h	2.205±0.212	2.10±0.25	0.093
1 week	1.43±0.17	1.31±0.22	0.029
1 month	0.66±0.84	0.62±0.23	0.833
6 months	0.32±0.37	0.21±0.14	0.156

TEP: Total extraperitoneal, SD: Standard deviation, QOL: Quality of life

DISCUSSION

Tension-free mesh repair is now the standard technique for adult inguinal hernia. The recurrence rate after mesh repair is as low as 0.3%–2.2%. As a result, nowadays, the area of interest in hernia surgery has shifted to postoperative pain and QOL. With the introduction of TEP and TAPP for repair of inguinal hernias, various studies have revealed that laparoscopic repairs are associated with less postoperative pain and complications as compared to open procedures. However, at the same time, there are reports in the literature which have questioned the superiority of minimally invasive procedures over open procedures. A recent meta-analysis of thirteen RCTs, including 3279 patients, comparing Lichtenstein and TEP in the treatment of inguinal hernias concluded that there is insufficient evidence to determine the greater effectiveness between TEP and Lichtenstein mesh techniques.[16]

The present study was designed to provide further insight into this area of research and to contribute a little to existing literature. The aim of this was to compare the best available open hernia repair technique with the best available laparoscopic technique. LWM was used for Lichtenstein repair, as LWM is associated with less chronic pain, groin stiffness, and foreign body sensation as compared to HWM. For TEP, HWM was used because various studies have found that LWM increases recurrence rate. Recently concluded RCT (TULP trial), in which lightweight (Ultrapro) was compared to heavyweight (Prolene), also found that use of LWM in laparoscopic inguinal repair is associated with statistically higher recurrence rate and does not offer any additional benefit in terms of postoperative pain.[4]

In our study, both the groups were comparable in terms of the patient demographics and no significant difference in the intraoperative complications was observed between two groups. The peritoneal breach was the only intraoperative complication that was encountered in TEP group. Subsequently, one of these patients presented with intestinal obstruction due to herniation of loop of bowel through peritoneal rent. A similar experience was reported by Andersson *et al*. in their study, where one patient who underwent TEP hernia repair presented with small bowel obstruction 3 days after surgery due to herniation.[8] Complex anatomy and presence of vital structures in the preperitoneal space predispose TEP to serious complications such as vascular injury, urinary bladder, or intestinal perforation. However, with gain in experience the incidence of these complications can be brought down significantly and comparable to open hernia repair. Gokalp *et al*. in their study found that there was no significant difference in terms of incidence of intraoperative complications in the two groups.[7] Lau *et al*. found similar results with only a few minor complications occurring in either surgical group. No major complication such as visceral injury or intestinal obstruction occurred.[9] Various postoperative complications such as urinary retention, constipation, seroma formation, hematoma, and wound infection were similar among both the groups. This is comparable to the results of various other RCTs.

In terms of hospital stay, one can presume that laparoscopic procedures shorten the hospital stay. However, nowadays, even open hernia repairs are done on daycare basis in most of the centers. We also observed that the patient who underwent Lichtenstein repair using LWM repair had a comparable length of hospital stay (1.46 days) to TEP using HWM (1.56 days), with no statistical difference.

To assess QOL after mesh hernioplasty, hernia-specific instrument, i.e., CCS is found to be a useful tool to effectively understand how surgical repair with mesh will affect patient QOL. It was developed by physician and researchers from Carolina laparoscopic and advanced surgery program, to monitor QOL in patients undergoing hernia repair. It measures the severity of pain, mesh sensation, and movement limitations during various day-to-day activities. In our study, almost all patients could make the components of CCS, except that 13 patients (25%) were unable to make out the difference between pain and sensation of mesh. Various other studies have also shown that CCS is feasible, easy to use with high acceptance rate in patients undergoing hernia repair with mesh.[17]

The overall reported incidence of chronic pain after herniorrhaphy is 12%; 18% in patients who undergo open surgery and 6% in patients who are treated laparoscopically.[18] The International Association for the Study of Pain has defined this chronic pain as pain lasting for longer than 3-month postoperatively.[19] The EU Hernia Trialists Collaboration review of 2003 patients treated by laparoscopic or open mesh repair showed that a significantly smaller number of laparoscopically treated patients developed a chronic pain state.[20] The Cochrane review on laparoscopic versus open inguinal hernia repairs reviewed 41 published reports of eligible trials that involved 7,161 participants and showed that there was less persistent pain (290/2,101 vs. 459/2,399) in the laparoscopic groups.[21] In our study, the mean groin pain was less in TEP using HWM as compared to Lichtenstein using LWM, at 24 h, 1 week, and 1 month postoperatively, and it was statistically significant. However, chronic pain was comparable in both the groups at 6 months' period. This could be attributed to the fact that LW mesh used in Lichtenstein was associated with less scar tissue formation and less chronic inflammatory reaction. Consequently, the advantages of laparoscopic surgery on chronic pain were nullified by the usage of LWM in Lichtenstein group.

The sensation of mesh was significantly low in Lichtenstein group using LWM as compared to TEP using HWM in the immediate postoperative period (24 h and at 1 week). The sensation of mesh at 1-month and 6-month period was less in Liechtenstein group using LWM as compared to TEP using HWM but not statistically significant. A plausible explanation to this fact could be that due to decreased groin pain in TEP group, patients focused more on the sensation of mesh whereas in Lichtenstein group due to more pain, patients paid less attention to foreign body sensation in early postoperative period. At 1 month and 6 months, when pain had reduced in both the groups, the sensation of mesh was comparable in both the groups. The difference in movement limitation with TEP group using HWM and Lichtenstein using LWM was not statistically significant at any time. This is contrast to other studies which reported more sensibility disorder and movement restriction following Lichtenstein repair with polypropylene mesh.[22]

It was thus observed that apart for less pain in the early postoperative period, TEP does not offer any additional benefit over Lichtenstein repair using LWM. Moreover, TEP is associated with certain disadvantages. The operative time and cost involved with TEP is significantly higher as compared to Lichtenstein repair.[7] Furthermore, there is a long learning curve for TEP as compared to Lichtenstein repair. The reported learning curve for TEP is between 30 and 250 surgeries. Neumayer *et al*. found higher recurrence rate (>10%) for surgeons who have performed less than 250 procedures.[23] Therefore, authors are of the opinion that Lichtenstein repair using LWM is good option for repair of uncomplicated inguinal hernia, particularly in developing nations where resources are limited.

CONCLUSION

The results of Lichtenstein technique for repair of inguinal hernia using LWM are comparable to laparoscopic TEP repair. Apart from less pain in the early postoperative period TEP repair do not offer any other benefit. However, large multicentered randomized trials are needed to further substantiate this fact, as a number of cases in the present study were small.

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Conflicts of interest
There are no conflicts of interest.

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Original Article

Right congenital diaphragmatic hernia: Four cases and literature review

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Abstract

Background: Left-sided Bochdalek congenital diaphragmatic hernia (CDH) is the most common type of CDH. The right-sided Bochdalek CDH is rare and usually contains only the liver as its content. We describe four children with right CDH managed at our institution over 3 years.

Materials and Methods: We present a retrospective analysis of four patients with right CDH managed at our institution from 2012 to 2015 with respect to age at presentation, sex, presenting symptoms, investigations, associated anomalies, management, and outcome.

Results: The average age at presentation was 1.5 years, with range being 5 days to 4 years. The three older children presented with a history of frequent respiratory tract infections. The neonate was a preterm low-birth weight baby and had respiratory distress since birth. A chest radiograph suggested right pneumothorax for which intercostal chest drain insertion was done. A repeat chest X-ray showed suspicion of right-sided CDH which was confirmed on computerized tomography. All four patients underwent right thoracotomy with repair of the CDH. The neonate expired on the 4th postoperative day. The other three children had an uneventful postoperative recovery.

Conclusion: Right-sided CDH is rare and has varied presentations and poorer outcome.

Keywords: Bochdalek, respiratory tract infection, right congenital diaphragmatic hernia

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INTRODUCTION

The history of congenital diaphragmatic hernia (CDH) in medical literature dates back to 1679 when CDH was first incidentally noted by Lazarus Riverius during an autopsy of a 24-year-old person.[1] In 1761, Giovanni Battista Morgagni described the classic anterior diaphragmatic hernia which has been named after him.[1] In 1848, Victor Alexander Bochdalek described both left and right posterolateral CDH, which today is referred as Bochdalek hernia.[1,2]

Thus, there are three types of CDH – posterolateral Bochdalek hernia, anterior Morgagni hernia, and hiatal hernia.[1] The left-sided Bochdalek hernia is the most common type occurring in approximately 85% of cases.[1] In this type of CDH, the large and the small bowel with or without intra-abdominal solid organ may be herniated into the thorax. The incidence of right-sided Bochdalek CDH is rare and occurs in approximately 13%–20%;[1,3] it contains only the liver along with some portion of the small bowel.[1,4] CDH can be isolated (nonsyndromic) or familial (in about 2% of cases) – which can be autosomal

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recessive, or X-linked.[1] The various syndromes which have CDH as a component are Fryns, Donnai–Barrow, Beckwith–Wiedemann, Simpson–Golabi–Behmel, Coffin–Siris, and Denys–Drash syndromes. Karyotype anomalies are seen in approximately 4% of cases – trisomy 13 and 18, tetrasomy 12p mosaicism, and deletions of 1q, 8p, and 15q.[1,2] CDH may be complicated and associated with other anomalies such as gastric volvulus, rotational abnormalities and midgut volvulus (incidence rate 30%–62%), gastric or intestinal perforations, and left ventricular hypoplasia of heart.[1,5]

Right CDH has a varied presentation and poorer prognosis. We present four children with right CDH managed at our institution over 3 years.

MATERIALS AND METHODS

Case records of four patients with right CDH managed at our institution from 2012 to 2015 were retrospectively analyzed with respect to age at presentation, sex, presenting symptoms, investigations, associated anomalies, management, and outcome.

All patients were admitted. Routine blood investigations were sent for all the patients. X-ray of the chest and abdomen; Ultrasonography (USG); two-dimensional (2D) echo; and computed tomography of the chest were done. After stabilization, the patients were taken for surgery.

RESULTS

Demographic profile

A total of four children with right CDH were treated over a 4-year period. The average age at presentation was 1.5 years, with the range being 5 days to 4 years. There were three males and one female.

Clinical presentation

The three older children presented with a history of frequent respiratory tract infections. They were investigated by the pediatricians and were diagnosed to have right CDH on X-rays and computed tomography of the thorax [Figure 1]. A 2D echo and abdominal ultrasound ruled out other associated anomalies in these patients. The neonate was a preterm low-birth weight baby delivered by the cesarean section and had respiratory distress since birth. He was given bag and mask ventilation after birth. A chest radiograph suggested right pneumothorax, for which intercostal chest drain insertion was done. However, there was no improvement in respiratory distress. A repeat chest X-ray showed suspicion of right-sided CDH which was confirmed on computerized tomography.

Management and outcome

All four patients underwent right thoracotomy with repair of the CDH [Figures 2 and 3]. All four patients had the right lobe of the liver as the content. The neonate required postoperative ventilator support but succumbed on the 4th postoperative day. The other three children had an uneventful postoperative recovery. They are asymptomatic on follow-up.

DISCUSSION

Right CDH is rare and accounts for about 13%–20% of all CDHs.[3] The embryogenesis of CDH has been postulated to be the failure in the closure of pericardioperitoneal canal by pleuroperitoneal membranes, which occurs during the 8th gestational week.[6] It is hypothesized that an early return of the foregut into the coelomic cavity results in wide

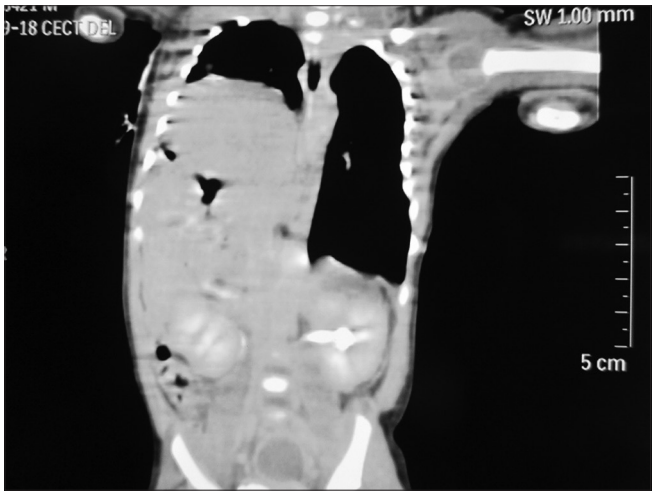

Figure 1: Coronal section of computed tomography of the thorax showing right congenital diaphragmatic hernia

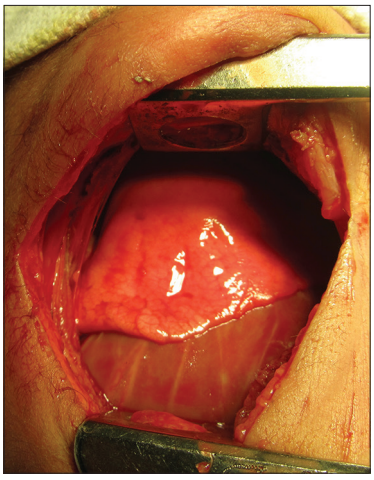

Figure 2: Intraoperative image showing right congenital diaphragmatic hernia with the liver (arrow) herniating into the right hemithorax

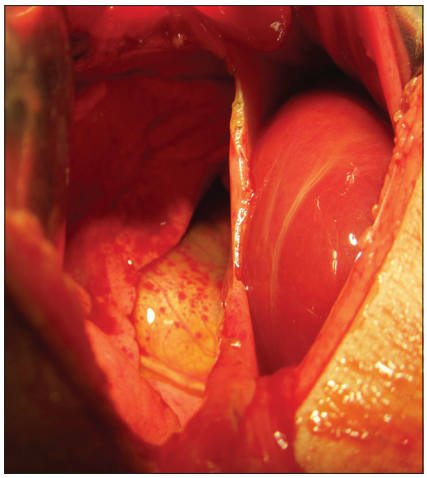

Figure 3: Intraoperative image showing the right-sided diaphragmatic defect with edges of the defined

posterolateral spaces and dysfunctional pleuroperitoneal membranes.[6] The abdominal viscera consequently herniate into the thorax. As a result, this causes pulmonary hypoplasia by compression of the growing lung.[6] The left CDH is more common than right CDH because of the early closure of right pleuroperitoneal opening; however, there are no postulated theories to explain this.[6]

Most of the patients present in neonatal age group with respiratory distress.[7] However, a delayed clinical presentation has been reported in approximately 5%–30% of the right CDH patients.[3] Late-onset right CDH can present itself in various ways in the form of respiratory distress, intestinal obstruction, jaundice, or failure to thrive.[3] It has been postulated that partial liver displacement, which occurs in most right-sided CDH patients, may block the further herniation of hollow viscera.[7] That is the reason why in the right CDH group most children have respiratory symptoms only.[7]

When the respiratory distress occurs in a baby with right CDH and group B streptococcal (GBS) sepsis, the radiological features represent GBS pulmonary inflammatory conditions and may be incorrectly interpreted as right-sided pneumonia, pleural effusion, or even pneumothorax.[3]

More than half of patients present with right-sided pleural effusion. Its cause has been speculated to be due to hepatic venous outflow obstruction which results in vascular congestion and a transudate exiting through the liver surface.[3] When liver is the only herniated organ, there would be no intestinal gas shadow in the right chest to give a clue to the underlying pathology. Erroneous chest tube drainage may be done for an apparent pleural effusion which may actually be a right CDH, with the liver being the reason for the clinical percussion dullness or radiographic opacification.[3] Deviation of the esophageal portion of the nasogastric tube to the left side along with the vertical orientation of the intra-abdominal portion of the nasogastric tube may provide the only clue to the diagnosis of right CDH in such cases.[3]

The most common radiological finding of right CDH is the opaqueness of the right hemithorax usually associated with mediastinal shift to the contralateral side on plain chest radiograph.[7] Computed tomography has been considered the ideal noninvasive technique for diagnosis – the presence of diaphragmatic defect, size, exact location, and contents of the various types of diaphragmatic hernia can be very well evaluated.[7]

Antenatal diagnosis of right CDH is difficult. The liver is the most common herniated into the thoracic cavity, and by analogy in the ultrasonographic echogenicity between the liver and lung, it becomes difficult to diagnose the right CDH.[8] Portal blood flow, ascites, or positioning of the gallbladder in the thoracic cavity may provide important diagnostic clues to the antenatal diagnosis of right CDH.[8] An intrathoracic kidney could also be a diagnostic clue for the diagnosis of right CDH.[8]

Right CDH, being rare, there are difficulties in evaluating its prognosis.[8] The current literature comparing the outcome of right CDH when compared to left CDH is inconsistent.[9] There are several studies in the literature which have described poorer postnatal outcomes in right CDH when compared with left CDH;[8,10-12] however, others have found no difference between the two types of CDH.[8,13,14] However, right CDH has been stated in the literature to carry disproportionately high mortality and morbidity.[15]

Herniation of the liver which is almost always present in right CDH has been perceived as an indicator of poor prognosis; however, this significance has only been shown for left CDH.[8] Liver herniation causes caval compression, leading to reduced preload and impaired cardiac output.[10] Moreover, the right lung normally accounts for 57% of the total functional lung volume; hence, any developmental abnormality of the right lung may have a bigger clinical impact.[8] Third, the size of defect necessary to permit herniation of the liver on the right side is likely to be substantially larger than that of a left-sided CDH. This explains the higher rates of patch repair required to manage right CDH.[8]

The significance of liver herniation and lung-to-head ratio have been documented as prognostic factors only

for left CDH.[8] As most fetuses with right CDH have a liver herniation, this finding cannot be a significant antenatal prognostic factor in right CDH.[8] Similarly, there is controversy regarding the utility of lung-to-head ratio measurements for predicting survival in the right CDH.[8]

CONCLUSION

Right CDH is rare and carries high rates of morbidity and requirement of extracorporeal membrane oxygenation and patch repair. The outcomes reported in the literature are mixed. The clinical presentation can be delayed and confused with pleural effusion and pneumonia. The outcome for patients with delayed presentation has been reported to be good.

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Conflicts of interest
There are no conflicts of interest.

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Original Article

A comparison of developing breast cancer-related lymphedema between mastectomy with reconstruction and mastectomy alone among breast cancer patients in Saudi Arabia

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Abstract

Context: The study was undertaken to compare developing breast cancer-related lymphedema between those who underwent mastectomy with reconstruction and mastectomy alone.

Aims: One of the most feared consequences after a mastectomy is breast cancer-related lymphedema (BCRL). However, few papers have questioned whether breast reconstruction impacts the development of lymphedema. This study aims to determine if breast reconstruction has an effect on the incidence of BCRL. Furthermore, the effect of the time (immediate vs. delayed) and type (implant based vs. autologous) of breast reconstruction on the development of BCRL will be evaluated.

Settings and Design: We conducted a retrospective cohort study on 320 patients who underwent mastectomy with reconstruction and mastectomy alone between January 1, 2007, and December 31, 2017, at King Abdulaziz Medical City – Jeddah.

Subjects and Methods: We conducted a retrospective cohort study on 320 patients who underwent mastectomy with or without breast reconstruction between January 2007 and December 2017. We reviewed patient medical records progressively to extract patients' characteristics, operative details, and lymphedema information. We divided our sample into two main groups: patients who underwent mastectomy with reconstruction and mastectomy alone. Mastectomy with reconstruction group was subdivided into immediate or delayed reconstruction and autologous or implant-based reconstruction.

Statistical Analysis Used: Statistical analysis was performed using the Statistical Package for the Social Sciences version 20.

Results: Of the total sample size 320, only 78 (24.4% (underwent mastectomy with breast reconstruction (Group 1), while the rest 242 (75.6%) underwent mastectomy alone (Group 2). From both the groups, 24 (7.5%) patients developed lymphedema; there was no significant difference between the breast reconstruction and development of lymphedema ($P = 0.67$). We subdivided Group 1 (patients who underwent mastectomy with breast reconstruction) into immediate breast reconstruction (40 patients, [51%]) and delayed breast reconstruction (38 patients, [49%]). In comparison between immediate versus delayed breast reconstruction, there was no significant difference

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between immediate and delayed breast reconstruction ($P = 0.67$). In terms of the type of reconstruction, we further subdivided Group 1 (patients who underwent mastectomy with breast reconstruction) into implant-based breast reconstruction (42 patients, [54%]), and autologous breast reconstruction (36 patients, [46%]). In comparison between implant-based versus autologous breast reconstruction, there was no significant difference between implant-based and autologous breast reconstruction ($P = 0.66$).

Conclusions: Although our result is insignificant, it suggests that patients who underwent mastectomy with reconstruction have a lower incidence of BCRL in comparison with those who underwent mastectomy alone. Moreover, our result suggests that immediate breast reconstruction and implant-based breast reconstruction have a lower incidence than delayed and autologous breast reconstruction. Further studies are needed to determine if the breast reconstruction has an effect on the development of lymphedema.

Keywords: Breast cancer, lymphedema, mastectomy, mastectomy with reconstruction

INTRODUCTION

The most common cancer affecting women worldwide is breast cancer.[1] Due to screening programs, early intervention, and improved treatment, the number of cancer survivors has increased.[1] Despite the advancement in procedural treatments, mastectomies (removal of breast tissue) are still common and are accounting for approximately 20%–30% of treated women with breast cancer.[2] One of the most feared consequences after mastectomy is breast cancer-related lymphedema (BCRL).[3] It is characterized by chronic swelling, localized pain, atrophic skin findings, and recurrent infections. All of these complications have severe psychological and physiological impacts.[3] Moreover, lymphedema has a potential effect on the quality of life due to body image changes, alterations in arm function, and increased complications such as infection and cellulitis.[4]

Clinicians have been recently focused on how to reduce the complications of breast cancer treatment so that breast cancer survivors can have a better quality of life.[5] BCRL- is one such complication that affects approximately 6%–30% of breast cancer survivors.[6] It has been associated with the body mass index (BMI), mastectomy, axillary dissection, axillary irradiation, and lymph node status.[5]

Breast reconstruction after mastectomy is increasingly performed and has become the standard of care in breast cancer management.[7] The goal of breast reconstruction after mastectomy is to restore a breast mound and to maintain health-related quality of life in breast cancer survivors.[8] Few papers have questioned whether breast reconstruction impacts the development of lymphedema.[5,7,8-10] The influence of breast reconstruction on postoperative lymphedema is yet to be clarified. Recent literature has suggested a beneficial reduction in lymphedema after both delayed and immediate breast reconstruction.[11,12] However, the results of these studies remain highly controversial.[3] According to a recent study published in 2015, reconstruction does not appear to alter lymphedema risk, whereas postoperative radiation therapy, obesity, and extensive axillary dissection greatly increase the risk.[3] Another study was conducted in 2012 suggested that patients who undergo breast reconstruction have a lower incidence and a delay in the onset of BCRL compared with patients who undergo mastectomy alone.[5]

Method of breast reconstruction and the development of lymphedema is another aspect to be discussed. Few studies have focused specifically on whether the reconstruction method affects the development of lymphedema.[13]

In this study, the incidence and time of developing BCRL- in mastectomy with breast reconstruction patients will be compared with those who underwent mastectomy alone. The other risk factors that were mentioned in the literature such as the BMI, axillary dissection, axillary irradiation, and lymph node status will be considered as well. Furthermore, the method of the reconstruction whether it was autologous or implant-based breast reconstruction will be compared in terms of BCRL- development. This study will contribute to the literature and may help the clinicians and the patients to take a decision regarding the breast reconstruction and the method of the reconstruction.

SUBJECTS AND METHODS

Study design

We conducted a retrospective cohort study on 320 patients who underwent mastectomy with reconstruction and mastectomy alone between January 1, 2007, and December 31, 2017, at King Abdulaziz Medical City – Jeddah. The study was approved by the Institutional Review Board at King Abdullah International Medical Research Centre, Ministry of National Guard Health Affairs. Patients who had lymphedema before the surgery, had lymphedema

with 2 weeks of the surgery and resolved spontaneously within 30 days, with missing data, or loss to follow-up were excluded from the study.

Data collection

We used patient files to review medical records progressively from the date of the procedure until the last follow-up. We extracted patients' demographic variables and medical histories from medical records including age, BMI, smoking, comorbidities (diabetes mellitus [DM], hypertension [HTN], and coronary artery disease [CAD]), preoperative chemotherapy or radiotherapy, and postoperative chemotherapy or radiotherapy. Patients' surgical histories were also extracted including mastectomy date and if it was skin-sparing or nipple-sparing, axillary intervention, development of BCRL-, and breast reconstruction. Axillary interventions were classified as without intervention, sentinel lymph node dissection (SLND), and axillary lymph node dissection (ALND). Lymphedema was diagnosed on the basis of clinical diagnosis and arm circumference measurements that documented by the physiotherapy department. Breast reconstruction was classified as immediate or delayed and further classified into autologous or implant based. To compare the development of BCRL between patients who underwent mastectomy with reconstruction and mastectomy alone, we divided our sample into two main groups: patients who underwent mastectomy with reconstruction and mastectomy alone. Mastectomy with reconstruction group was subdivided into immediate or delayed reconstruction and autologous or implant-based reconstruction.

Outcomes

The outcomes of this study were the rates of lymphedema among patients who underwent mastectomy with reconstruction and compare it with the patients who underwent mastectomy alone. We also compared the rates of lymphedema between the different types of reconstruction (immediate vs. delayed and autologous vs. implant based).

Statistical analysis

Statistical analysis was performed using the Statistical Package for the Social Sciences version 20 (IBM Corp. Released 2011. IBM SPSS Statistics for Windows, Version 20.0. IBM Corp., Armonk, NY). For expressing the qualitative variable, frequency and percentage were used and for stating the quantitative variable, mean and standard variable were used for normally distributed data, and in case of skewed data, median and interquartile range were used. When comparing the qualitative variables, Chi-square test/ Fisher's exact test was used and found the association of the risk factors, with dependent variable regression analysis being done. $P < 0.05$ was considered statistically significant.

RESULTS

Over the study period, 320 patients were identified. The median age was 48 years, while for the BMI, the median was 28. For the other variables, 24% were diabetic, 28% hypertensive, 4% smoker, 2% had CAD, 45% underwent preoperative chemotherapy, 8% underwent preoperative radiotherapy, 61% underwent postoperative chemotherapy, 46% underwent postoperative radiotherapy, and only 7.5% developed lymphedema [Table 1]. For the axillary intervention, 211 (66%) patients underwent ALND, 37 (12%) patients underwent SLND, and 72 (23%) patients had no axillary intervention. For those who developed lymphedema, the average time from the date of surgery until the date of lymphedema 8.9 months and further details are shown in Table 2. The incidence of BCRL was 7.5% in our sample. While applying the Chi-square test to assess the association between lymphedema and the risk factors, it was found that no significant association between lymphedema and age, BMI, DM, HTN, smoking, CAD, or postoperative radiotherapy. However, there was a significant association between lymphedema and preoperative chemotherapy, preoperative radiotherapy, and postoperative chemotherapy. In stepwise

Table 1: Comorbidities and risk factors for developing lymphedema

	Yes, *n* (%)	No, *n* (%)
Diabetes	77 (24)	243 (76)
Hypertension	91 (28)	229 (72)
Smoking	14 (4)	306 (96)
Coronary artery disease	7 (2)	313 (98)
Preoperative chemotherapy	143 (45)	177 (55)
Preoperative radiotherapy	25 (8)	295 (92)
Postoperative chemotherapy	194 (61)	126 (39)
Postoperative radiotherapy	148 (46)	172 (54)
Lymphedema	24 (7.5)	296 (92.5)

Table 2: Time from the date of surgery until the date of lymphedema

Time to develop lymphedema (months)	*n* (%)
2	1 (4)
3	1 (4)
4	4 (16)
5	3 (12)
6	3 (12)
7	2 (8)
8	1 (4)
9	2 (8)
10	2 (8)
11	3 (12)
12	1 (4)
22	1 (4)
24	1 (4)
Total	25 (100)

logistic regression analysis, "risk factors of preoperative chemotherapy and postoperative chemotherapy were significantly associated with lymphedema." The results of binary logistic regression model illustrate that those who said "yes" to preoperative chemotherapy are 2.5 times more likely to get lymphedema as compared to those who said "no." In addition, those who said "yes" to postoperative chemotherapy are 3.3 times more likely to get lymphedema as compared to those who said "no." Of the total sample size 320, only 24% (78) underwent mastectomy with breast reconstruction (Group 1), while the rest (76%, [242]) underwent mastectomy alone (Group 2). From both the groups, 7.5% (24) patients developed lymphedema; there was no significant difference between the breast reconstruction and development of lymphedema ($P = 0.67$) [Table 3]. We subdivided Group 1 (patients who underwent mastectomy with breast reconstruction) into immediate breast reconstruction (40 patients, [51%]) and delayed breast reconstruction (38 patients, [49%]). In comparison between immediate versus delayed breast reconstruction, there was no significant difference between immediate and delayed breast reconstruction ($P = 0.67$) [Table 4]. In terms of the type of reconstruction, we further subdivided Group 1 (patients who underwent mastectomy with breast reconstruction) into implant-based breast reconstruction (42 patients, [54%]) and autologous breast reconstruction (36 patients, [46%]). In comparison between implant-based versus autologous breast reconstruction, there was no significant difference between implant-based and autologous breast reconstruction ($P = 0.66$) [Table 5].

Table 3: Lymphedema versus no lymphedema

Breast reconstruction	Lymphedema		Total, *n* (%)	*P*
	Yes, *n* (%)	No, *n* (%)		
No	19 (79)	223 (75)	242 (76)	0.67*
Yes	5 (21)	73 (25)	78 (24)	
Total	24	296	442	

*Chi-square test (there is no significant differences in the percentages among lymphedema for breast reconstruction)

Table 4: Immediate versus delayed breast reconstruction

Breast reconstruction	Lympedema		Total, *n* (%)	*P*
	Yes, *n* (%)	No, *n* (%)		
Yes "Immediate"	2 (40)	38 (52)	40 (51)	0.67*
Yes "Delayed"	3 (60)	35 (48)	38 (49)	
Total	5	73	78	

*Fisher's exact test (there is no significant differences in the percentages among lymphedema for breast reconstruction)

Table 5: Implant based versus autologous breast reconstruction

Type of reconstruction	Lympedema		Total, *n* (%)	*P*
	Yes, *n* (%)	No, *n* (%)		
Implant based	2 (40)	40 (55)	42 (54)	0.66*
Autologous	3 (60)	33 (45)	36 (46)	
Total	5	73	78	

*Fisher's exact test (there is no significant differences in the percentages among lymphedema for the type of reconstruction)

DISCUSSION

As a result of advances in breast cancer treatment, physicians and patients give more attention for the quality of life after the treatment.[14] One of the most feared complications of breast cancer treatment is lymphedema which results from disruption to the lymphatic system.[14] According to a recent meta-analysis, one in every five patients developed BCRL following breast cancer treatment.[15] Few papers have questioned whether breast reconstruction impacts the development of lymphedema.[5,7-10] The influence of breast reconstruction on postoperative lymphedema is yet to be clarified. Recent literature has suggested a beneficial reduction in lymphedema after both delayed and immediate breast reconstruction.[8,9]

The average time from the date of surgery until the date of lymphedema is 8.9 months. It may be possible that this average time has been impacted by the axillary intervention (66% of the sample had ALND); according to this result from the recent study, early-onset lymphedema (<12 months postoperatively) was associated with ALND.[16] For other factors, our analysis showed a significant association between lymphedema and preoperative chemotherapy, preoperative radiotherapy, and postoperative chemotherapy.

In comparison with a recent study that published in 2017, they found the incidence of lymphedema 9.1%, while in our sample, it was 7.5%.[17] The relatively lower incidence of lymphedema in our sample may be due to differences in patients' characters, treatment course, and rehabilitation program. For the 7.5% who developed BCRL, 79% of them had mastectomy alone, while only 21% had a mastectomy with breast reconstruction, which suggests that breast reconstruction might decrease the possibility of developing BCRL. The result of the study conducted in 2012 supports our findings; it showed that patients who undergo breast reconstruction have a lower incidence and later onset of BCRL compared with patients who undergo mastectomy alone.[5] Another study published in 2015 suggested that reconstruction does not appear to alter lymphedema risk, which indicates that further study should conduct to determine whether breast reconstruction really decreases the incidence of BCRL or not.[3]

Miller *et al*. in the study suggested that immediate implant reconstruction does not increase the risk of lymphedema compared to mastectomy alone.[7] Our result showed

that those who developed lymphedema from breast reconstruction group, 40% of them underwent immediate breast reconstruction, while the higher percent underwent delayed breast reconstruction (60%).

Lee *et al*. noted that autologous reconstruction was associated with a significantly reduced risk of lymphedema compared with that for tissue expander/implant reconstruction.[13] However, our result suggested against these findings, implant-based breast reconstruction group had a lower incidence of lymphedema when compared with autologous breast reconstruction. Further studies are needed to compare immediate versus delayed breast reconstruction and implant-based versus autologous breast reconstruction since our result conducted on a small population.

CONCLUSIONS

Although our result is insignificant, it suggests that patients who underwent mastectomy with reconstruction have a lower incidence of BCRL in comparison with those who underwent mastectomy alone. Moreover, our result suggests that immediate breast reconstruction and implant-based breast reconstruction have a lower incidence than delayed and autologous breast reconstruction. Further studies are needed to determine if the breast reconstruction has an effect on the development of lymphedema.

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Conflicts of interest

There are no conflicts of interest.

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Case Report

Multiple wandering jejunal intussusceptions in an adult patient with celiac disease: Atypical presentation

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Abstract

Intussusception is a rare presentation of celiac disease. In this report, we describe the condition of a 28-year-old Saudi male known case of brucellosis diagnosed and treated 10 months ago. The patient presented to the hospital complaining of diffuse intermittent dull abdominal pain with mild abdominal distention for 1-month duration, which starts gradually with progressive course. Patient also was given history of generalized fatigability for the same duration. These symptoms were associated with a weight loss about 10 kg in the last 6 months. Abdominal examination revealed soft and lax abdomen with mild tenderness at epigastria area. Computed tomography (CT) abdomen showed jejunal loop intussusceptions with diffuse jejunal wall thickening. Gastroscopy was done and showed fissuring of duodenal folds. Biopsy report came later as duodenal villa atrophy with increased intraepithelial lymphocytosis. The patient diagnosed as celiac disease. After establishing the diagnosis, dietary advice was given and he was discharged for outpatient department follow-up. Currently, the patient remains well and repeat CT scan showed persistence of multiple intussusceptions, though at different sites of the jejunum.

Keywords: Abdominal pain, coeliac disease, intussusception, repeated, small bowel disease

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INTRODUCTION

Coeliac disease or celiac disease is a long-term autoimmune disorder that primarily affects the small intestine.[1] Coeliac disease is caused by a reaction to gluten, a group of various proteins found in wheat and in other grains such as barley and rye.[2-4] Classic symptoms include gastrointestinal (GI) problems such as chronic diarrhea, abdominal distention, malabsorption and among children failure to grow normally.[5] Nonclassic symptoms are more common, especially in people older than 2 years. There may be mild or absent GI symptoms, a wide number of symptoms involving any part of the body or no obvious symptoms.[6-8] Intussusception is a medical condition in which a part of the intestine folds into the section immediately beside it. It typically involves the small bowel and less commonly the large bowel.[9-10] Intussusception is rare in adults. Reported cases of intussusception in celiac disease suggest that it may be asymptomatic, transient, and limited to the small intestine and rarely requires surgical intervention;[11]

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however, enteropathy-associated T-cell lymphoma should be considered in the differential diagnosis.[12]

CASE REPORT

A 28-year-old Saudi male with past history of brucellosis diagnosed and treated 10 months ago. The patient presented to the hospital complaining of diffuse intermittent dull abdominal pain with mild abdominal distention 1-month duration which starts gradually with progressive course. Patient also was given history of generalized fatigability for the same duration. Nausea with occasional vomiting was also recorded by the patient. These symptoms were associated with weight loss of about 10 kg in the last 6 months. No history of fever or night sweating was observed. No history of GI bleeding was noted. No history of skin rash or joint pain or bone pain was also observed. The patients had neither jaundice nor history of chest pain, shortness of breath, or cough. No history of dysuria or change in color or frequency of urine was noted. No history of similar attack before. No significant family history. In addition, surgery or drug history was negative.

The patient was diagnosed as brucellosis case 10 months ago as he complained of fever and arthralgia, which started on antibiotics, but they noticed that time he had picture of unexplained iron deficiency anemia, and then patient missed the follow-up.

Clinical examination

The patient was conscious, oriented, pale, but neither jaundice nor lymphadenopathy. Body mass index was 19.5. The patient was vitally stable and afebrile. Chest and cardiovascular examinations were free.

Abdominal examination revealed soft and lax abdomen with mild tenderness at epigastria area. No organ enlargement was detected, but bowel sound was recorded. Lower limb examination was normal.

Investigation

Laboratory investigations revealed white blood cell = 9.7, Hb = 10.5 g/dl, MCV = 66.1, MCH = 19.1, PLT = 306, and INR = 1.1.

Liver function test and renal profile were normal.

Ferritin was low, normal thyroid function test, tissue transglutaminase IgA was positive with high titer.

Imaging

Computed tomography (CT) abdomen showed jejunal loop intussusceptions with diffuse jejunal wall thickening Figure 1. Gastroscopy was done and showed mild gastric erythematous mucosa at gastric antrum with fissuring and scalloping of duodenal folds of second part of duodenum (5 biopsies were taken) Figure 2.

Duodenal biopsy report showed duodenal villa atrophy with increase intraepithelial lymphocytosis which going with celiac disease. The patient diagnosed as celiac disease.

Treatment

After final diagnosis, the patient started on gluten-free diet and dietician referral was done, while micronutrient deficiencies were also corrected. The patient followed by

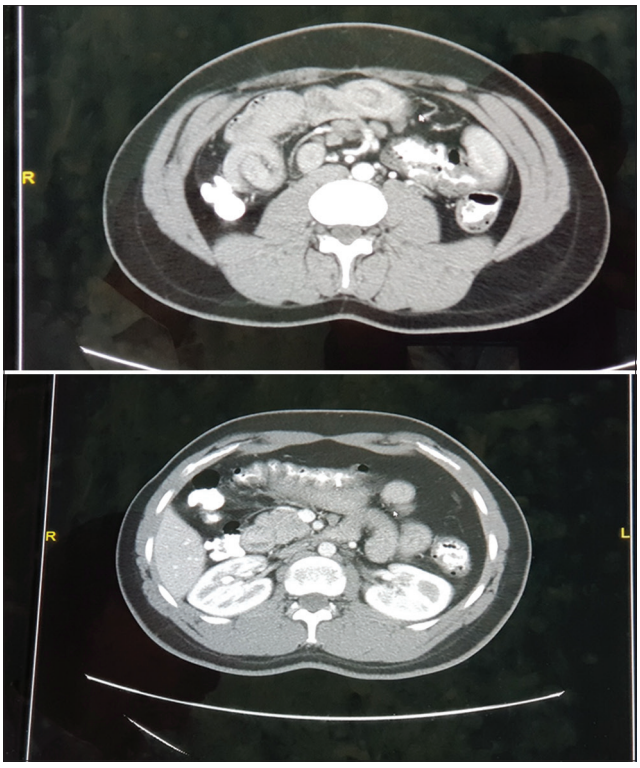

Figure 1: Computed tomography abdomen showing jejunal loop intussusceptions with diffuse jejunal wall thickening

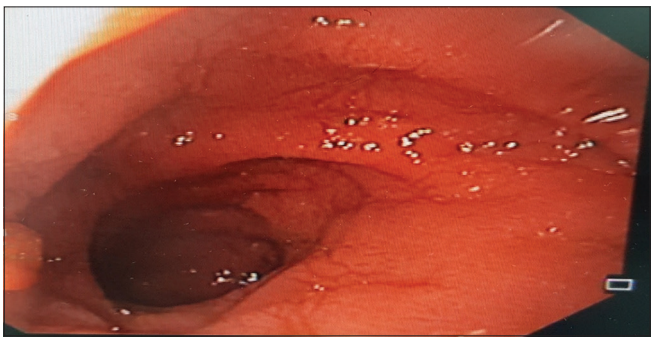

Figure 2: Upper gastrointestinal endoscopy showing mild fissuring and scalloping of duodenal folds in the second part of duodenum

general surgeon regarding the intussusception, which does not need any surgical intervention.

Outcome and follow-up

The patient was seen after 3 months, clinical symptoms were improved, with neither more abdominal pain nor distention. The patient had good appetite and no more fatigability. His body mass index was improved to 24, and his abdomen was soft lax with no tenderness. Patient laboratory investigations also were improved as Hg was increased to 12 g/dl, and the celiac serology titer was improved while patient was on strict gluten free diet. Follow-up CT abdomen revealed persistence of multiple intussusceptions, though at different sites of the jejunum. The patient instructed to be on strict gluten-free diet with follow-up in clinic after 2 months.

DISCUSSION

Coeliac disease is an autoimmune GI disorder caused by permanent intolerance to ingested gluten in genetically susceptible individuals.[13-15] Its magnitude was reported among Caucasians in Europe, North and South America, Australia, and the Middle East to be as high as 1 in 100.[16,17] Conventionally, coeliac disease clinically presented with symptoms of intestinal malabsorption disorder, resulting in weight loss, diarrhea, steatorrhea, or abdominal distension. Regarding these typical clinical presentations, coeliac disease can be presented by other atypical complaints such as isolated subclinical iron deficiency anemia, osteoporosis, neurologic disease, nonspecific abdominal symptoms, dermatitis herpetiformis, or even intussusception.[18,19] In recent years, there has been increasing recognition that the mode of presentation of coeliac disease may be changing.[20] It often presents with symptoms not previously considered to be characteristic of the disease.[21,22]

Intussusception is not a commonly recorded complication of celiac disease. In 1968, Ruoff *et al*.[23] stated the occurrence of intussusception in adult celiac disease for the first time. An occurrence of intussusception among adults with celiac disease had been reported. Although more common among children, intussusception not confirmed among childhood celiac disease.[24] Germann *et al*.[11] in 1997 recorded celiac disease as an uncommon cause of recurrent intussusception in children for first time. Then, Mushtaq *et al*.[25] reported three children with intestinal intussusception in conjunction with celiac disease and recommended that the finding of transient small bowel intussusception should be considered clinically for celiac disease. Another many cases of intussusception among cases of coeliac disease were recorded.[26-30]

All these reported associations between intussusception and coeliac disease were in concordance with these cases reported in this study as the patient reported here also complained of abdominal pain with bowel motion disturbances, especially constipation, and after clinical examination and biopsy, he was confirmed as a case of intussusception for coeliac disease patient.

Intussusception can be a clinical complication of celiac disease so that celiac disease should be considered in patients with intussusception and growth failure, especially in unusual age range.

CONCLUSION

Small bowel intussusception in adult with suspected celiac disease initially should be considered and managed expectantly rather than by early surgical intervention. The finding of transient small bowel intussusception should prompt investigation for celiac disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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Case Report

Bronchoesophageal fistula, a rare complication post laparoscopic sleeve gastrectomy: A case report and literature review

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Abstract

Acquired bronchoesophageal fistula (BEF) and tracheoesophageal fistula are rare disorders that result from medical disease or secondary to a complication of a procedure, most commonly due to the prolonged high-pressure endotracheal or tracheostomy cuffs in the presence of nasogastric tube in the esophagus. Rarely, esophageal injuries can result in BEF; presentation is usually after 1 week of the procedure, and the treatment is esophageal stenting or clips in the early phase, and failure will need definitive surgical management. Here, we present our case of a complex BEF post laparoscopic sleeve gastrectomy that required endoscopic and surgical management. This is a case report and literature review.

Keywords: Bronchoesophageal fistula, esophageal stents, sleeve gastrectomy complications, tracheoesophageal fistula

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INTRODUCTION

Obesity is raising both in developed and developing countries due to the changes in lifestyle and food habits. Surgical treatment of obesity has revolutionized in recent years with laparoscopic sleeve gastrectomy (LSG), being performed more commonly. Hemorrhage and leak are common complications of LSG. Late complication like bronchoesophageal fistula (BEF) is rare to find in literature. We present our case of LSG, which was complicated by the development of BEF and its management.

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CASE REPORT

A 35-year-old female presented to obesity clinic with body mass index of 39 and with unremarkable past history except primary infertility. She failed to reduce her weight by diet and regular physical exercises referred from her obstetrician for consideration of bariatric surgery. After full assessment and preparations including esophagogastroduodenoscopy (EGD), the patient underwent uneventful LSG on May 28, 2015, with normal water-soluble oral contrast swallow and meal study day 1

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postoperative; the patient started on clear fluid and discharged home with postbariatric diet, medicinal, and exercise instructions. Seen after 2 weeks in bariatric surgery clinic and her condition was stable, her wounds were checked and advanced to soft diet and multivitamins and proton-pump inhibitor medications were prescribed. Five weeks later, the patient presented with intractable vomiting and severe epigastric pain that was not responding to medical therapy, eventually resulted in electrolyte imbalance and hence she was admitted in a surgical ward for investigation and fluid replacement. Esophagogastroduodenoscopy (EGD) with minimal insufflation was done and showed stenosis at the site of the cardia (mid-sleeved stomach). Through-the-scope balloon dilatations were done up to 12 mm at the stricture site with no improvement in symptoms. Two days later, another EGD was done, and another narrowing was found 5 cm proximal to pylorus in addition to the previous one. A 15-cm long self-expandable totally covered metallic stent (SEMS) applied from the distal esophagus to the first part of the duodenum. Two weeks later, the patient still had recurrent nausea and vomiting, so another EGD showed migration of the SEMS distally. Readjustment of the stent was done and her vomiting improved, but 2 days later, the patient started to have tachycardia, hematemesis, melena, and hemoglobin dropped from 11.7 to 9.7 in 1 day. She was treated conservatively with close observation, pantoprazole infusion, and kept Nill Per Mouth diet. Total parental nutrition was started. Four days later, the patient had developed tachycardia, hypotension, shortness of breath, and pallor. Her hemoglobin level dropped to 4 this time, so urgent EGD was done after blood and fluid resuscitation which revealed ulceration at the upper limit of the stent (distal esophagus), argon beam applied over ulcer and the patient was kept under observation; there was no more hemoglobin drop, and the patient was tolerating oral feeding and was discharged home in stable condition 4 days later. Two weeks after her last admission, she presented to the emergency room with repeated vomiting and epigastric pain; ultrasound abdomen was done which showed gallbladder stones without evidence of cholecystitis. A water-soluble contrast swallow and meal study ruled out leak or obstruction with stent in place. Almost 6 weeks later, she was readmitted with recurrent vomiting and signs of dehydration and electrolyte imbalance as well as productive cough. A supportive management was initiated, and EGD was performed and removal of stent was done. There was suspicion of a fistula opening at the distal esophagus, so a computed tomography (CT) of the chest with oral and intravenous (IV) contrast showed dilated and thickened terminal esophagus with a complex fistula between the lower esophagus and the airways of the medial basal segment of the right lower lobe [Figures 1 and 2].

Bronchoscopy confirmed the presence of fistula in the distal right basal medial bronchus of the lower lobe. The patient was offered surgical management, but she refused initially, because of the patient's and her family's wish to avoid surgical management and as her hemodynamic status was stable with parenteral nutrition and broad spectrum antibiotics and antifungal therapy; she wanted to try more conservative management approach if she can avoid going through major surgery to repair the fistula. We entertained the trial of less invasive endoscopic and conservative management, which included NPO, high dose proton-pump inhibitor (PPI) therapy, IV broad-spectrum antibiotics, and antifungal, total parenteral nutrition and chest physiotherapy. Other attempts to place endoscopic SEMS stent with follow-up barium meal showed persistent leak of fistula and distal migration of the stent [Figure 3]. EGD and retrieval of the stent and placement of Ovesco clip (over-the-scope clip [OTSC]) was done. One week later, CT chest showed persistent of the fistula. The decision of operative surgical intervention was discussed again with the patient and her family who agreed to proceed for high-risk surgery to close the fistula, repair of esophageal fistula site, segmental lung resection and laparoscopic cholecystectomy.

Intraoperative bronchoscopy showed the site of fistula at right lower basal medial segment, and EGD confirmed location of fistula, at distance 35 cm with multiple holes seen at the esophageal side. After preparations, double-lumen lung isolation, central line, Foley catheter, and pneumatic compression, the patient was placed in left lateral decubitus position, and right 7th intercostal space posterior thoracotomy was performed. We were able to identify the fistula site. Excision of fistulous tract was done, leaving 8 cm defect in lateral wall of the lower esophagus with thick mucosa that has been debrided to bleeding edges.

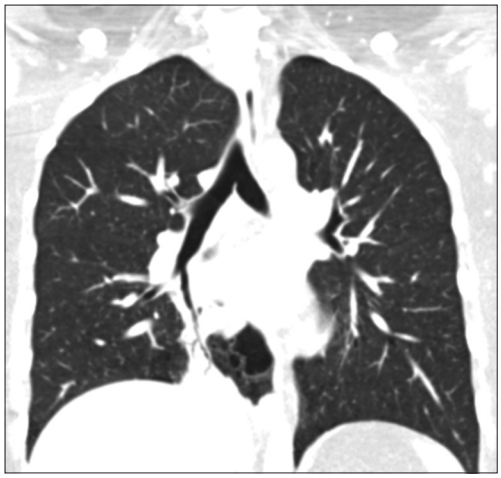

Figure 1: Computed tomography chest showed the fistula tract between the lower esophagus and the right lower lobe bronchus

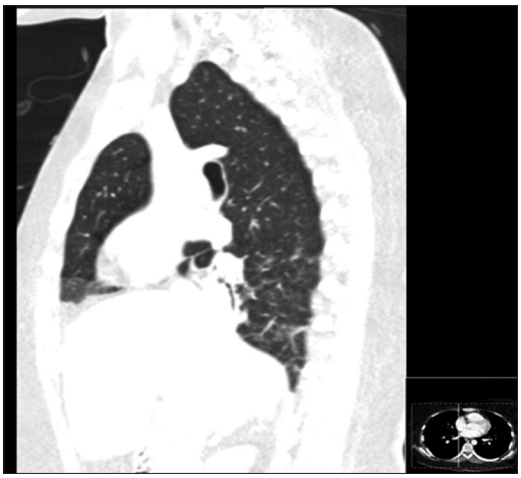

Figure 2: Sagittal view of computed tomography chest that showed the fistula site clearly delineates between the lower esophagus and the segmental bronchus of the right lower lobe

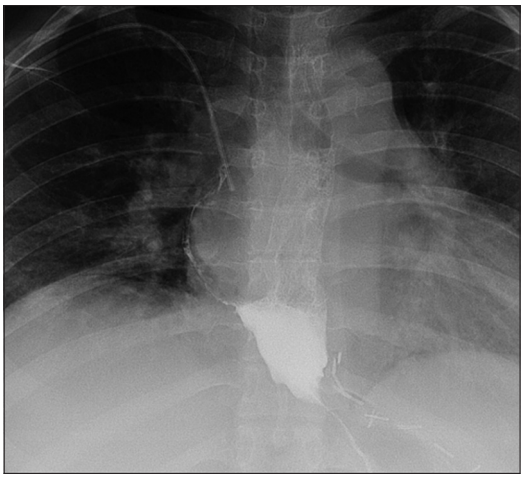

Figure 3: A water-soluble contrast swallow study clearly showed the fistula tract between the dilated distal esophagus and the right lower lobe bronchus

Nonanatomical segmental resection was done with blue stapler 60 of the medial basal segment of right lower lobe. Air/water leak test was done which showed no air leak from the lung resection stapler line. A traction diverticulum was noted in the esophagus due to chronic inflammation, and stapling of the esophageal defect was done using stapler 60 TL green over in placed EGD to close the lateral defect without narrowing the esophageal lumen. The second layer of interrupted 3/0 PDS buttress sutures was carried out, followed by intercostal muscle flap and BioGlue to cover the fistula site. Air/water leak test was negative for leak in the esophagus, then insertion of two chest tubes, followed by abdominal laparoscopy for the removal of gallbladder and insertion of feeding jejunostomy tube in the same operative setting. Postoperative day 2, a water-soluble swallow and meal contrast study showed no leak and free passage of contrast to the duodenum. The patient started on oral fluid diet day 4 postoperative and advance to full fluid diet. The patient was discharged 10 days later in good condition with planned clinic follow-up in 2 weeks; pain medications, multivitamins, and PPI were prescribed to the patient.

On follow-up clinical visits, the patient was tolerating oral diet and her symptoms of reflux and obstruction improved. EGD at 6 months and CT chest showed no evidence of fistula recurrence and resolution of esophagitis with normal-looking sleeve stomach. At 1 year postoperative, the patient got pregnant and delivered a healthy baby girl with cesarean section approach.

DISCUSSION

LSG has emerged as the procedure of choice for morbid obesity.[1] It is a restrictive procedure, which works by restricting food intake and producing early satiety by removing ghrelin-producing portion of stomach.[2]

BEF is defined as an abnormal connection between bronchial tree and esophagus. Common causes are malignancies, but other causes such as infections, esophageal injury, surgery, foreign body, prolonged endotracheal intubation, ingestion of chemical products, and swallowed dental prosthesis have been reported.[3-6] Diagnosis is usually delayed due to nonspecific symptoms with the most commonly pathognomonic symptom of cough or chocking during ingestion of food or fluid. Other presentations include recurrent Pneumonias and lung abscesses or gastrointestinal (GI) bleeding. Diagnosis requires a high index of suspicion. Imaging studies include plain chest radiography, contrast study, CT of the chest, EGD, and bronchoscopy. Bronchoscopy may reveal inflammatory changes; a discrete focus of heaped-up granulations, visualization of the fistula orifice, and appearance in the bronchus of dyed instilled in the esophagus would be diagnostic. Braimbridge and Keith classified tracheoesophageal fistula and BEF after reviewing 23 cases, Type Ia wide-necked congenital diverticulum of the esophagus. Stasis may occur in the dependent tip, which becomes inflamed and perforates into the lung. Type 2a short track runs directly from the esophagus to the lobar or segmental bronchus. Type III consists of a fistulous track connecting the esophagus to a cyst in the lobe, which in turn communicates with the bronchus. In Type IV, the fistula runs into a sequestrated segment, which is recognized by the presence of a systemic arterial supply from the aorta.[7]

Following sleeve gastrectomy, bleeding occurs in 1%–6% of cases.[8] Leaks occurs up to 5% of cases.[1] Gastric

leak occurs mainly in the upper part of the stomach due various factors such as high intragastric pressure, impaired peristaltic activity, and ischemia, thereby decreasing oxygen supply and resulting delayed healing. Staple line dehiscence and thermal damage are other possible causes of leak. Early leaks are detected 1–4 days postoperatively, intermediated 5–9 days, and late after 10 days. Another classification is Type 1 when leak is local without dissemination or spillage and Type 2 when dissemination to peritoneal or thoracic cavity. If extraluminal leak is diagnosed late, then it may result in peritonitis, sepsis, gastrocutaneous fistula, or organ failure and even death.[9] Early diagnosis of leak is crucial for proper management; tachycardia and fever are two constant indicators in many studies.[10,11]

The principles for successful fistula management are control of sepsis, establishment of a good nutritional status, pulmonary support rehabilitation, and endoscopic and/or surgical repair. In our case, the fistula appeared after the iatrogenic injury occurred during EGD, and the trial to remove the covered SEMS from the stomach and necrosis in the lower esophageal wall with argon beam coagulation (ABC) was the most likely culprit. Initial management includes the treatment of infection associated with the fistula formation (mediastinitis), endoscopic management OTSC,[12] SEMS,[13] and Amplatzer vascular plug.[14] SEMS has been widely used for malignant palliation, leaks, perforation, and fistula, with a high successful rate. Swinnen *et al*. reported the complications of SEMS, spontaneous migration occurred in 11.1% of stents, and there were minor complications (dysphagia, hyperplasia, and rupture of coating) in 20.9% and major complications (bleeding, perforation, and tracheal compression) in 5.9%.[13] OTSC is a new endoscopic modality, used for closure of full-thickness GI tract fistula tracts. Zolotarevsky *et al*. reported one case of BEF in a woman with esophageal diverticulum, with successful management and closure of fistula. We believe that the failure of conservative approach in our patient was due to many reasons such as chronic fistula, poor nutrition status, presence of reflux, and obstructive symptoms such as recurrent forcible vomiting which may lead to stent migrations and possible erosions, another cause in this patient fistula development that the fact of using ABC in inflamed bleeding wall of esophagus, which leads to necrosis of the wall and the fistula tract formation. The fact of finding multiple holes and defect with the development of esophageal diverticulum indicates the chronicity of the fistula which considers another reason for failure of conservative therapy with less invasive approaches for fistula closure with stent or clips applications. In chronic fistula, surgical approach is considered the gold standard. The standard operative procedure consists of right posterolateral thoracotomy or minimally invasive approach through video-assisted thoracoscopy, exposure and division of the fistula, primary repair of bronchus and esophagus, and pedicle tissue interposition in addition to J feeding tube through abdominal approach for postoperative nutrition. We have chosen the thoracotomy approach due to dense adhesions encountered in the chest cavity with the chronic and large defects in the esophagus. Complications after surgical repair of BEF are often due to poor general condition of the patient and pulmonary failure. Mangi *et al*. study showed that 6 out of 13 patients discharged home on postoperative day 10 and the other patients had complications including persistent pinhole fistula, thoracic duct leak which required re-operation, prolonged intubation and respiratory toilet, transient subglottic edema requiring re-intubation, and thoracentesis for persistent pleural effusion.

CONCLUSION

BEF is a rare complication of sleeve gastrectomy, which is difficult to diagnose and require intensive management, nutritional support, control of sepsis, leak management, and eventually definitive surgical management, which is the gold standard. Endoscopic therapy includes SEMS and OTSC still good modalities with acceptable successful rates that can be used in selective cases with proper patient selection in acute presentation of BEF.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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