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Benign biliary stricture and its rare association—Mirizzi syndrome: A case series and literature review

CS Wong, AK Al-Ajami, JM Crotty, SA Naqvi

ABSTRACT

Introduction: Mirizzi syndrome is a rare variant of obstructive jaundice due to extrinsic compression of common bile duct (CBD) or common hepatic duct (CHD) by an impacted gallstone in the infundibulum (Hartmann's pouch) or neck of the gallbladder or in the cystic duct. The diagnosis of this rare condition is important because its presence is associated with an increased risk of bile duct injury when a standard cholecystectomy is carried out. Case Series: A series of three similar cases of benign bile duct stricture secondary to cholecystolithiasis (Mirizzi syndrome) are reported. Their clinical symptoms, diagnosis and management and outcomes were not identical. Conclusion: To reach the final correct diagnosis of a rare variant of gallstone-related disease is always challenging. Recognition of several different clinical presentations of a same disease is important in order to avoid any complications during an operation. There should be a careful diagnostic approach towards Mirizzi syndrome before laparoscopic cholecystectomy is planned.

Keywords: Jaundice, Benign bile duct stricture, Mirizzi syndrome, MRCP, ERCP

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INTRODUCTION

Gallstone disease is increasingly common in the Western world. Cholesterol stones account for the vast majority of gallstones in western countries. The spectrum of symptomatic gallstone disease may be varied depending on the location of gallstones in the biliary tract. This could lead to many different clinical manifestations which include acute or chronic cholecystitis, cholangitis, gallstone pancreatitis, and small bowel obstruction secondary to gallstone ileus.

Mirizzi syndrome is a rare complication of gallstones. It was first described by Pablo Luis Mirizzi in 1948 [1]. It is a rare form of obstructive jaundice which accounts only for 0.1% of all patients with gallstone disease [2]. It occurs in 0.3–3% of patients undergoing cholecystectomy [3]. Complications of Mirizzi syndrome include biliary enteric fistula and biliary stricture. The development of fistulae is uncommon, and when they occur, are either cholecystoduodenal or cholecystocolic [4, 5]. The most common cause of benign bile duct strictures is injury to the bile duct during cholecystectomy. Other causes may include chronic pancreatitis, common bile duct stones, acute cholangitis, biliary obstruction due to cholecystolithiasis (Mirizzi syndrome), postransplantation stricture, primary sclerosing cholangitis (PSC), and biliary enteric anastomosis. While, pathophysiology of biliary obstruction in Mirizzi syndrome is usually due to
extrinsic compression, those biliary strictures of the common hepatic or common bile duct related to the same syndrome are not well documented. It is important to rule out pancreaticobiliary malignancy causing stricture. In one study, it was found that biliary brush cytology had 56.2% sensitivity and 100% specificity in diagnosing biliary and pancreatic malignancy [6]. We report a case series of Mirizzi syndrome including three patients who presented with different demographic profile, clinical presentation, management and outcome. Preoperative recognition of this is important for a surgeon because laparoscopic treatment of Mirizzi syndrome has a relatively high rate of complication [7].

**CASE SERIES**

**Case 1:** A 51-year-old woman with background medical history of hypertension was presented to the surgical service with sudden onset of epigastric pain for four days. The pain was described as *burning* in nature which radiated to the back and usually occurred in the evening after eating meal. No relieving features. There was no previous history of jaundice, pale stool or dark colored urine. She denied any history of alcohol consumption. On clinical examination, her temperature was 36.0°C, pulse rate 81 beats per minute, blood pressure 139/70 mmHg, respiratory rate 20. Her abdomen was not distended but diffuse tenderness in the epigastric region on deep palpation. Murphy’s sign was negative.

Full blood count revealed an elevated WBC 13.60×10⁹/L, Hb 13.0 g/dL, Platelet 324×10⁹/L. Serum sodium Na 138 mmol/L, serum potassium 3.1 mmol/L, urea 1.5 mmol/L, creatinine 60 mmol/L, bilirubin (total) 104 µmol/L, alkaline phosphatase (ALP) 202 IU/L, gamma-glutamyl transferase (α-GT) 320 IU/L, alanine transaminase (ALT) 274 IU/L, amylase 30 IU/L, C-Reactive Protein (CRP) 189 mg/L and erythrocyte sedimentation rate (ESR) 88 mm/h. Urine dipstick was negative for glucose, bilirubin, ketone, blood and nitrates, but with traces of protein and leukocytes.

An ultrasound examination of the abdomen (Figure 1) scan showed a borderline dilated common bile duct (CBD) measuring between 6 and 8 mm. The intrahepatic ducts were dilated. There were multiple stones inside the gallbladder. The gallbladder wall was thickened and inflamed in appearance. The MRCP examination (Figure 2) confirmed multiple large gallstones and a diffusely gallbladder wall. There appeared to be a Mirizzi’s-type compression the bile duct confluence with dilatation of the intrahepatic ducts. However, the distal CBD appeared normal with no visible dilation or intraductal calculi.

She was initially managed conservatively with intravenous antibiotic and adequate analgesia. Endoscopic ultrasound (EUS) and endoscopic retrograde cholangiopancreatography (ERCP) were performed. Gallstones were identified within the gallbladder. The CBD measured 4.5 mm. Biliary tree stricture was found at the level between common hepatic duct and upper common bile duct. Biliary brush cytology (BBC) was performed. Amsterdam stent (10 French, 9 cm straight) was inserted into the biliary systems. Stricture dilatation was also performed. Subsequently, she had delayed (interval) cholecystectomy.

Five months later, she was readmitted with a clinical diagnosis of mild pancreatitis (Glasgow Score of two for...

**Figure 1:** Ultrasound examination of the abdomen shows a borderline dilated common bile duct measuring 7.6 mm (Case 1).

**Figure 2:** MRCP examination of patient with Mirizzi syndrome shows narrowing of the proximal CBD (white arrow). The intrahepatic ducts are dilated (white-dashed arrow) (Case 1).
raised white cell count and mildly low serum calcium level). Laboratory investigation showed an elevated WBC 16.57×10⁹/L with predominantly neutrophilic 14.55×10⁹/L, Hb 14.8 g/dL, Platelet 318×10⁹/L. Serum sodium 136 mmol/L, serum potassium 3.1 mmol/L, urea 3.9 mmol/L, creatinine 73 mmol/L, bilirubin (total) 87 µmol/L, ALP 239 IU/L, α-GT 692 IU/L, ALT 764 IU/L, and a very high serum amylase level 1408 IU/L. Calcium 1.94 mmol/L, lactate dehydrogenase (LDH) 527 IU/L, glucose 5.9 mmol/L and albumin 40 g/L. She was hypoxic but no derangement of metabolic component on the arterial blood gas (pH 7.37, pO₂ 9.5 kPa, pCO₂ 5.8 kPa, bicarbonate 24.5 mmol/L, and base excess −1.0 mmol/L).

A repeat ultrasound abdomen (Figure 3) showed mildly dilated CBD, measuring 7 mm in diameter, with mild prominence of left intrahepatic ducts. A repeat MRCP (Figure 4) showed filling defects in the lower extrahepatic CBD with moderate distension of the upper tract. The appearances suggested the possibility of intraductal calculi. The ERCP was repeated and balloon dilatation performed. Biliary stricture was observed in the lower CBD. However, despite the MRCP appearance, no stones were retrievable during the procedure. Biliary brush cytology did not reveal any evidence of malignant cells. Cellular specimen composed of sheets of benign and reactive epithelial cells. Her symptoms had settled with supportive measures and discharge well.

Figure 3: Repeat ultrasound examination shows a mildly dilated CBD measuring 7 mm (Case 1).

Figure 4: Repeat MRCP examination shows dilated CBD with a filling defect suggesting intraductal calculi (black arrow). The intrahepatic ducts are also dilated (white-dashed arrow) (Case 1).

Case 2: An obese 90-year-old woman with severe chronic obstructive pulmonary disease, ischemic heart disease and hypothyroidism was admitted with jaundice, nausea and feeling unwell for one week. She also complained of having mild right upper quadrant pain associated with passing dark-colored urine and pale stool. She denied any recent of loss of appetite or loss of weight. She denied any history of alcohol consumption. On examination, she was afebrile, the temperature was 36.6°C, pulse rate 74 beats per minute, blood pressure 130/65 mmHg, respiratory rate 18. Right upper quadrant and epigastrum tenderness were elicited. Her abdomen was soft and Murphy’s sign was negative.

Full blood count revealed an elevated WBC 8.22×10⁹/L, Hb 13.7 g/dL, Platelet 258×10⁹/L. Serum sodium 139 mmol/L, serum potassium 4.1 mmol/L, urea 4.7 mmol/L, creatinine 62 mmol/L, bilirubin (total) 171 µmol/L, ALP 508 IU/L, α-GT 604 IU/L, ALT 105 IU/L, amylase 53 IU/L. Urine dipstick was negative for urinary tract infection.

Ultrasound examination of the abdomen (Figure 5) revealed a contracted gallbladder with shadowing of gallstones. The CBD measured 9.0 mm in diameter with moderate intrahepatic duct dilatation. MRCP (Figure 6) revealed a large calculus in the neck of the gallbladder which lay in close proximity to the common hepatic duct. There was an elongated stricture of the common hepatic duct with marked dilation of the intrahepatic biliary tree. The lower extra hepatic common bile duct was also distented but without visible filling defect distally. The pancreas and pancreatic duct appeared normal. The appearances were those of a stricture of the common hepatic duct causing marked intrahepatic ductal distension. It was not clear whether this was due to the Mirizzi’s-type effect upon the bile duct from the adjacent gallbladder calculus or there was an intrinsic stricture of the duct.

Cholangiography revealed a tight common hepatic duct stricture with proximal intrahepatic ductal dilation. Malignancy of the biliary tree was suspected. Biliary
brush cytology was performed which reported a rare group of atypical ductal cells present in an inflammatory background, favoring a regenerative process. A Cotton Leung (10 French, 10 cm straight) biliary stent was inserted and biliary stricture dilatation was also performed.

A further work-up showed she had elevated tumor markers [CA-199 97.3 U/mL (Reference range: 0–37 U/mL) and CA-125 117.0 U/mL (Reference range: 0–35 U/mL)]. Carcinoembryonic antigen (CEA) was of normal limits (3.3 ng/mL), reference range: 0–3.4 ng/mL. The CT examination of the abdomen showed large stones in the gallbladder. No discrete pancreatic head mass, lymphadenopathy, liver mass or other finding of note was seen.

In view of her age and coexisting medical conditions, she was considered not fit to proceed with surgical treatment. After sometime, she suddenly deteriorated and died from acute heart failure.

**Case 3:** A 45-year-old male was presented with a history of right upper quadrant pain radiating to the right scapula (Boas’s sign) associated with dark-colored urine and pale stool. He was known to have gallstone disease as diagnosed two years back in the Philippines. He had no other significant past medical or surgical history. He denied any recent consumption of alcohol and no recent travel history. On examination, he was markedly jaundiced. His temperature was 36.6°C, pulse rate 83 beats per minute, blood pressure 115/72 mmHg, respiratory rate 16. Right upper quadrant tenderness was present but no sign of peritonitis. His abdomen was soft and Murphy’s sign was negative.

Full blood count revealed an elevated WBC 9.42×10⁹/L, Hb 12.7 g/dL, Platelet 427×10⁹/L. Serum sodium 137 mmol/L, serum potassium 4.6 mmol/L, urea 3.5 mmol/L, creatinine 76 mmol/L, bilirubin (total) 155 μmol/L, ALP 234 IU/L, α-GT 330 IU/L, ALT 91 IU/L, amylase 52 IU/L.

Ultrasound examination of the abdomen (Figure 7) indicated that there was a contracted gallbladder containing echogenic structures casting shadows consistent with gallstones. Bile ducts were of normal size with CBD measuring 5 mm. The MRCP examination (Figure 8) confirmed a large stone which was impacted at the level of a dilated cystic duct. There was marked intrahepatic duct dilatation. There was also an extensive column of stones in the common bile duct. The contracted gallbladder contained stones.

The patient subsequently underwent ERCP and sphincterotomy was also performed. He did not improve as expected and ERCP was repeated. The CBD was grossly dilated proximally and parallel to this was a grossly dilated cystic duct. There was a large stone

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**Figure 5:** Ultrasound examination of the abdomen shows a markedly dilated CBD measuring 9 mm (Case 2).

**Figure 6:** MRCP examination shows marked dilatation of the intrahepatic biliary tree (white-dashed arrow) with distended CBD (white arrow) (Case 2).

**Figure 7:** Ultrasound examination of the abdomen shows a normal CBD measuring 5 mm (Case 3).
DISCUSSION

Biliary strictures are uncommon. Their clinical presentation may, in fact, mimic the more common condition such as cholecystitis, choledocholithiasis, cancer of the bile duct and pancreatic cancer. Benign bile duct strictures may develop from chronic pancreatitis and iatrogenic injury to the bile duct after a laparoscopic cholecystectomy. The latter is not uncommon due to arising technical feasibility in the modern era of surgical advancement.

There are numerous causes for benign bile duct strictures. Causes of benign biliary stricture can be divided into: (a) Congenital—biliary atresia, (b) iatrogenic from bile duct injury at surgery—cholecystectomy, choledochotomy, gasterectomy, hepatic resection, transplantation and biliary-enteric anastomosis, (c) Inflammatory—CBD stones, cholangitis, chronic pancreatitis, parasitic, PSC and biliary obstruction due to cholecystolithiasis (Mirizzi syndrome), (d) Trauma and, (e) Idiopathic. These causes can also be further classified into benign or malignant. For example, cholangiocarcinoma and pancreatic cancer are associated with the malignant cause of biliary stricture. Therefore, it is important to rule out these in suspected groups of patients. Primary bile duct tumors appear at an average age of 60 years but may appear at any time between 20–80 years of age. There is no gender preference. Most malignant biliary tumors are adenocarcinomas located in the hepatic or CBDs.

Radiological imaging is important in the investigation and diagnosis of bile duct strictures. Imaging modalities include ultrasonography, MRCP, ERCP, transhepatic cholangiography and CT cholangiography. Both direct visualization of the biliary tract (diagnosis) and extraction of any stones present in the biliary tract (therapeutic) can be performed during ERCP procedure. Sensitivity and specificity in making a diagnosis of bile duct stricture is therefore higher with ERCP.

Management of benign stricture includes balloon dilatation and stent insertion. Complications of these procedures include cholangitis or further stricture formation. With an experienced endoscopist available, the outcome of such surgery is good, with 90% of patients having no further complications.

Both the cases (Case 1 and Case 2) of biliary stricture demonstrated may appear secondary to Mirizzi syndrome. This is an unusual variant of a rare condition. Case 3 demonstrated a classical Mirizzi syndrome due to external compression of extrahepatic duct secondary to an impacted large cystic duct stone. Balloon dilation and insertion of a stent were performed in all the three cases.

Case 1: This was a very interesting case. At the initial presentation, cholangiogram revealed 2.0 cm stricture at the level between the common hepatic ducts and upper common bile duct. A stent placement was carried out. She had delayed (interval, subsequent admission) cholecystectomy six weeks after her initial presentation of acute cholecystitis. The stent was removed post interval cholecystectomy. This patient was readmitted with a clinical evidence of gallstone pancreatitis five months post laparoscopic cholecystectomy. A repeat MRCP showed intraductal calculi in the distal CBD. At the repeat ERCP, stricture in the distal CBD was noted but no stones were seen. Balloon dilation was carried out without stent placement. This feature of stricture appears to be
benign. Intraductal stone that was seen on the repeat MRCP could have been passed from biliary tree systems.

In this case, we considered the following possibilities cause of biliary stricture: (a) Idiopathic benign common bile duct stricture, (b) Stricture secondary to Mirizzi syndrome and bile duct stone, (c) Iatrogenic stricture post ERCP, (d) Iatrogenic postoperative (cholecystectomy) stricture, (e) Intraductal retained stone (Post cholecystectomy Mirizzi syndrome).

Post cholecystectomy Mirizzi syndrome due to retained calculi in the remnant cystic duct following cholecystectomy [8, 9] is very uncommon. The presenting symptom may be dyspepsia or pain. Abnormal liver function study, jaundice and cholangitis are other manifestations that indicate residual biliary disease. Choledocholithiasis, biliary stricture, and chronic pancreatitis are the most common causes of symptoms. Patients with suspicious findings should be studied by ERCP or transhepatic cholangiogram (THC).

Case 2: In this case, a very old lady was admitted for obstructive jaundice secondary to biliary stricture. An ultrasound abdomen and MRCP revealed an impacted stone in the neck of the gallbladder, stricture of the common hepatic duct, and intrahepatic ductal distention. Malignant tumor of the bile duct is considered at presentation based on the clinical picture and epidemiological profile of the patient. Biliary brush cytology did not reveal any evidence of malignancy (i.e., cholangiocarcinoma) or inflammatory disease (i.e., PSC, ulcerative colitis). Association between Mirizzi syndrome and raised CA-199 is unknown [10, 11]. Some authors advocate that the elevated value of CA-199 is of no value in benign obstructive jaundice [12].

In the debilitated unfit patients at presentation, the only viable option may be percutaneous ultrasound-guided cholecystostomy. A temporary external biliary drainage may be achieved by passing a catheter percutaneously into intrahepatic duct. Stents may be passed through strictures at the time of ERCP and left to drain into the duodenum. When the general condition of the patients have improved, definitive surgery can be considered. Initial conservative management consists of intravenous fluid and electrolyte replacement, nasogastric suction, parenteral analgesia and systemic antibiotic is important to prevent further deterioration of seriously ill patients.

Case 3: Gallstone disease is uncommon among males. This patient presented with classical manifestation of obstructive jaundice. MRCP indicated that the cystic duct was impacted with a large gallstone causing extrahepatic and intrahepatic ductal dilation. This is a classical picture of Mirizzi syndrome. Neither MRCP nor ERCP shows evidence of biliary strictures. He eventually had a biliary stent inserted to bypass the narrowed part of the duct and allow bile drainage. He underwent open cholecystectomy and subsequently recovered well.

Association between biliary tree stricture and Mirizzi syndrome is not well documented in literature. Stricture secondary to inflammation process may be due to direct or indirect insult (e.g., pancreatitis), but the exact pathophysiology is unknown. The impacted stones, plus the associated chronic inflammation surrounding the biliary tree system, may cause a significant degree of narrowing and obstruction and subsequently strictures formation.

CONCLUSION

Biliary stricture is rarely associated with Mirizzi syndrome. At onset of presentation, the mainstay of treatment is to relieve the strictures by performing biliary stent insertion or stricture dilatation. Further investigations are necessary to determine the ‘true’ cause of these strictures.

Mirizzi syndrome is an uncommon spectrum of gallstone disease and diagnosis of this condition can be challenging. Surgery is the mainstay of therapy for the condition, therefore early recognition of its presence is essential to avoid bile duct injury. Interestingly, the syndrome itself can be manifested pre or post cholecystectomy treatment.

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Author Contributions
CS Wong – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
AK Al-Ajami – Analysis and interpretation of data, Drafting the article, Final approval of the version to be published
JM Crotty – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
SA Naqvi – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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REFERENCES


Heterotopic pregnancy in spontaneous conception: Report of three cases and review of literature
Zied Khediri, Chaouki Mbarki, Anis Ben Abdelaziz, Najeh Hsayoui, Mezghenni S, Hedhili Oueslati

ABSTRACT

Introduction: Heterotopic pregnancy is an uncommon clinical condition in which intrauterine and ectopic pregnancies occur at the same time. Case Series: We report three cases of heterotopic pregnancy in a natural conception that had different clinical presentation and surgical management. Conclusion: Although heterotopic pregnancy is a rare event in spontaneous conception, physicians must always keep in mind that confirming an intrauterine pregnancy does not exclude the coexistence of an ectopic pregnancy especially in women presenting a pelvic pain

Keywords: Heterotopic, Ectopic pregnancy, Spontaneous conception

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INTRODUCTION

Heterotopic pregnancy is diagnosed in the presence of simultaneous gestations at two or more implantation sites. It is commonly used to define a rare clinical condition in which intrauterine and ectopic pregnancies occur at the same time. It was first reported in the year 1708 as an autopsy finding. Its occurrence is rare in spontaneous conception with an incidence of 1:30,000 [1], and is much higher with assisted reproductive techniques (ART). The diagnosis can be easily missed, leading to life-threatening complications. We report three cases of heterotopic pregnancy in a natural conception.

CASE SERIES

Case 1: A 34-year-old primigravid woman was brought to our emergency room in the gynecology and obstetrics department, with a history of an acute pain in her abdomen, with nausea and vomiting for six hours. She was six weeks pregnant. It was a spontaneous conception and there was no past history of abortion, infertility, pelvic inflammatory disease or any history of abdominal surgery. The patient had never had contraception. The patient knew she was in a gravid state, has already consulted a midwife, and has had obstetrical ultrasonography that had shown an intrauterine pregnancy one week earlier. On examination, she was pale with a pulse rate of 145 per minute and blood pressure of 80/40 mmHg. Abdominal examination revealed diffuse lower abdominal
tenderness. Pelvic examination revealed an enlarged uterus corresponding to seven weeks of pregnancy, with no bleeding. Laboratory investigations showed an anemia with a 6.3 g/dL hemoglobin level. HCG dosage was positive. Despite an initial resuscitation with intravenous colloids, the patient had instable hemodynamics. She has been directly conducted to undergo an emergency exploration laparotomy. The laparotomy revealed a seven weeks gravid uterus and rupture of the left tube at its isthmic level and the presence of approximately 2.5 liters of hemoperitoneum. Left salpingectomy with removal of the hemoperitoneum and peritoneal lavage was performed. Dilatation and curettage (D&C) has immediately followed the laparotomy, which brought normal trophoblast that has been addressed to histological examination. The patient was transfused with two units of blood during the surgery and her postoperative period was uneventful. Pathology of the resected specimen confirmed the ruptured tubal pregnancy. Histology of the D&C product showed the presence of regressive chorionic villi confirming an intrauterine miscarriage. The patient was discharged on the fourth postoperative day. Follow-up was uneventful.

**Case 2:** A 29-year-old woman, who had a history of infertility for two years that has not been explored, consulted in our emergency for acute pelvic pain. She had eight weeks of amenorrhea. Physical examination found lower abdominal tenderness, with enlarged uterus corresponding to eight weeks of pregnancy, with no bleeding. Hemodynamics was stable. Ultrasonography found an intrauterine seven weeks ongoing pregnancy, and a left latero-uterine heterogeneous mass, with an extra-uterine embryo and positive cardiac activity. Routine laboratory tests were normal. Diagnosis of heterotopic pregnancy was confirmed and the patient underwent urgent exploratory laparoscopy. Laparoscopy confirmed the left tubal pregnancy, and found 200 mL of hemoperitoneum (Figure 1). The patient had left salpingectomy and we have decided to preserve the intrauterine pregnancy. Postoperative period was uneventful, and patient was discharged under progesterone tocolysis. The intrauterine pregnancy was monthly followed-up in our department until delivery on term by vaginal delivery.

**Case 3:** A 32-year-old woman, with a past history of late miscarriage, has consulted the emergency room for moderate pelvic pain with seven weeks of amenorrhea. Physical examination was normal. Ultrasonography examination found an intrauterine ongoing pregnancy of seven weeks, with a non-complicated ovarian cyst of 5 cm. The patient was discharged home with symptomatic treatment. The patient consulted the emergency department again after two days, with persistent pelvic pain. Clinical examination found pelvic tenderness with stable hemodynamics. Ultrasound found the ongoing intrauterine pregnancy, the ovarian cyst, and discovered a ruptured left tubal pregnancy with an extrauterine embryo and positive cardiac activity (Figure 2A). Diagnosis of heterotopic pregnancy with ruptured tubal pregnancy was confirmed. The patient immediately underwent laparoscopic surgery. Laparoscopy found a ruptured tubal pregnancy and 400 mL of hemoperitoneum (Figure 2B). The patient had left salpingectomy. In postoperative period, the patient had an ultrasound examination that checked the positive cardiac activity of the intrauterine pregnancy. Patient was discharged after three days under progesterone treatment. To date, the patient is 13 weeks pregnant and no complication occurred after surgery.

**DISCUSSION**

Heterotopic pregnancy is defined as the presence of multiple gestations, with one being present in the uterine cavity and the other outside the uterus [2, 3]. In natural conception cycles, heterotopic pregnancy is a rare event, it occurs in about 0.08% of all pregnancies [4]. With assisted reproduction techniques, however, this incidence increases to between 1/100 and 1/500, and occurs in 5% of pregnancies achieved after in vitro fertilization [5]. In our department, we have recorded three cases of confirmed heterotopic pregnancy that are

![Figure 1: Laparoscopic aspect of the heterotopic pregnancy: large uterus, left tubal pregnancy, hemoperitoneum.](image)

![Figure 2:](image)

![Figure 2: (A) Ultrasound aspect of the heterotopic pregnancy: ongoing intrauterine pregnancy (IUP), extra-uterine embryo (EUP), ovarian cyst (CYST), (B) Laparoscopic aspect of the heterotopic pregnancy: large uterus, left ruptured tubal pregnancy, hemoperitoneum.](image)
CONCLUSION

In natural conception cycles, heterotopic pregnancy is a rare event. Physicians must always keep in mind that confirming an intrauterine pregnancy clinically or by ultrasound does not exclude the coexistence of an ectopic pregnancy that should systematically be suspected in any woman presenting abdominal pain, genital bleeding and/or hypovolemic shock during pregnancy. Laparoscopic approach should always be preferred, and treatment should preserve the intrauterine ongoing pregnancy.

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REFERENCES

Isolated intracranial relapse of hodgkin lymphoma in a child

Harriet Holme, Niharendu Ghara, Thomas S Jacques, Paul Humphries, Stephen Daw, Ananth Shankar

ABSTRACT

Introduction: Although involvement of the central nervous system (CNS) is well described in non-Hodgkin lymphoma (NHL) at diagnosis and/or at relapse, there are few reports in the pediatric literature of primary or relapsed CNS Hodgkin lymphoma (HL). Case Report: We report dural disease as the only site of relapse of HL in an immunocompetent seven-year-old child, presenting with diplopia. Imaging demonstrated a well-circumscribed dural-based mass at the base of the skull involving the clivus and enveloping the pituitary. Restaging did not show disease elsewhere, with normal bone marrow trephine biopsies and cytological examination of the cerebrospinal fluid. Biopsy of the dural-based tumor confirmed classical HL. Eleven months before, he was diagnosed with stage IIIA HL nodular sclerosis subtype and underwent four courses of combination chemotherapy with a complete metabolic response on early response assessment. Forty-eight months after completing treatment for relapsed HL, he remains well in complete remission, although future relapse of HL cannot be excluded. Conclusion: There were no specific risk factors predictive of relapse in this child. He had responded well to standard chemotherapy as early response assessment after two courses showed complete metabolic response. Treatment of this child at relapse was challenging, since standard salvage chemotherapy regimens comprise drugs not well known to traverse the blood brain barrier. This case highlights the rarity of intracranial HL in children and difficulty with regards to the diagnosis, evidence based treatment and overall prognosis. We suggest that irrespective of any imaging abnormality, histological confirmation should be sought before commencement of definitive treatment.

Keywords: Lymphoma, CNS relapse, Pediatric hematology/oncology, Hodgkin lymphoma

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INTRODUCTION

Hodgkin lymphoma (HL) involving the central nervous system (CNS) is a rare. Involvement of the CNS
is well described in non-Hodgkin lymphoma (NHL) at both diagnosis and/or at relapse. However, there are few reports in the pediatric and adult literature of primary or relapsed CNS HL [1–5]. The management of this child including a review of literature is discussed.

CASE REPORT

A seven-year-old boy was presented with diplopia. Clinical examination was normal except for a left convergent squint. Magnetic resonance imaging (MRI) examination of the brain was normal. Eleven months before, he was diagnosed with stage IIIA HL nodular sclerosis subtype and underwent four courses of combination chemotherapy [Vincristine/Oncovin, Etoposide, Prednisolone, and Doxorubicin/Adriamycin (OEPA) x 2 and Cyclophosphamide, Vincristine/Oncovin, Prednisolone and Procarbazine (COPP) x 2]. Since early response assessment (ERA) with 18 fluorodeoxyglucose (FDG) positron emission tomography (PET) after two courses of OEPA showed a complete metabolic response, he did not receive radiotherapy. However, his diplopia progressed over two months, and a generalized seizure prompted another MRI brain scan; this showed a well-circumscribed dural-based mass at the base of the skull involving the clivus and enveloping the pituitary (Figure 1A). This lesion was also FDG-avid (Figure 1B). Restaging with whole body MRI and FDG PET did not show disease elsewhere. Bone marrow trephine biopsies and cytological examination of the cerebrospinal fluid were normal. Serologic testing for human immunodeficiency virus was negative. He had no family history of malignancy or immunodeficiency. Full evaluation of the integrity of the hypothalamic pituitary axis prior to the commencement of relapse treatment was normal.

Biopsy of the dural-based tumor confirmed classical HL. Figure 2 shows nodules containing Hodgkin cells and occasional Reed-Sternberg (RS) cells set against mixed inflammatory cell infiltrate. The Hodgkin cells and RS cells were positive for cluster of differentiation (CD) 30, CD15 and Epstein-Barr virus-encoded ribonucleic acid (EBER) but negative for CD3 and CD20. Histological examination showed clear evidence of residual epithelium, consistent with anterior pituitary gland, and glial tissue both infiltrated by HL as shown by cytokeratin immunohistochemistry (using MNF116), and Glial fibrillary acidic protein (GFAP) immunohistochemistry, respectively.

Clinical examination prior to the commencement of relapse chemotherapy showed left 6th and 7th cranial nerve palsies. He had no palpable lymphadenopathy or hepatosplenomegaly. He received two cycles of salvage chemotherapy comprising high dose cytarabine/Ara C, dexamethasone and cisplatin/platinol (DHAP). An ERA was performed 14 days after the 2nd DHAP course and included whole body MRI and PET scans; MRI scan showed almost complete resolution of the dural lesion (Figure 3A) while the PET scan showed complete metabolic response (Figure 3B). He had 2 further courses of DHAP followed by 30 Gy radiotherapy to the clivus and pituitary region.

Forty-eight months after completing the treatment for relapsed HL, he remains well in complete remission, although future relapse of HL cannot be excluded.

DISCUSSION

Hodgkin lymphoma unlike NHL rarely involves the CNS and occurrence of intracranial disease is usually

Figure 1: MRI, PET and CT scan of a seven-year-old boy, (A) Sagital T1 weighted image (TR 14 ms, TE 769 ms) demonstrating a mass lesion centred on the skull base, involving the pituitary (long arrow) and clivus (arrowhead), with an adjacent extra-osseous soft tissue dural based mass (short arrow), (B) Coronal T2 weighted image (TR 5140 ms, TE 133 ms) demonstrating extension of the lesion into the left cavernous sinus (short arrow), partially surrounding the left internal carotid artery (arrowhead). Note patchy clivus signal (long arrow).

Figure 2: Histopathology report of biopsy, (A) and (B) Hematoxylin and eosin stain of sphenoid mucosa, (C) CD30 stain of dura and pituitary, (D) CD15 stain of dura and pituitary Arrows indicate Hodgkin and RS cells. Scale bar – 50 μm.
with disseminated relapse [2, 6, 7]. We report dural disease as the only site of relapse of HL in an immunocompetent child.

There were no specific risk factors predictive of relapse in this case report; the child had responded well to OPEA chemotherapy as the ERA by FDG PET after two courses showed a complete metabolic response [8]. It is arguable that without radiotherapy, his initial treatment was sub-optimal but this approach was according to the European pediatric Hodgkin treatment strategy and there are no suggestions that non-irradiated patients with intermediate risk HL and good early response have a worse outcome [9, 10]. Although the striking feature of the MRI scan at relapse was the dural-based lesion involving the pituitary, there was also abnormal signal within the clivus. The mechanism of leptomeningeal involvement is unknown, but thought to originate from systemic hematogenous spread [11–13]. However, in our patient the dural lesion was the only site of relapse. It is difficult to know whether the clivus was the site of actual relapse with dural involvement as an extension from the clivus or vice versa (Figure 1). Review of whole body MRI and FDG PET scans at first presentation did not show any evidence of bone involvement.

Since standard salvage chemotherapy regimens comprise drugs not well known to traverse the blood brain barrier (BBB), treatment of this child at relapse was challenging. We chose chemotherapy that included high dose cytarabine and cisplatin, both reported to penetrate the CNS [14, 15]. Success of the relapse treatment strategy is reflected in the ERA MRI and FDG PET scans (Figure 3A–B). It is possible initial biopsy was beneficial, with surgery disrupting the BBB helping with CNS penetration of chemotherapy [16].

CONCLUSION

This case highlights the rarity of intracranial HL in children and difficulty with regards to the diagnosis, evidence based treatment or overall prognosis. Considering this case report, we suggested that irrespective of any imaging abnormality, histological confirmation should be sought before commencement of definitive treatment.

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Author Contributions

Harriet Holme – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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Thomas S Jacques – Acquisition of data, Analysis and interpretation of data, Final approval of the version to be published
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REFERENCES

Primary serous carcinoma of peritoneum: A case report

Shelly Sehgal, Reena Agarwal, Prashant Goyal, Sompal Singh, Vinita Kumar, Ruchika Gupta

ABSTRACT

Introduction: Primary serous papillary carcinoma of the peritoneum (PSPCP) is a rare malignant epithelial tumor that is histologically indistinguishable from papillary serous carcinoma of the ovary (PSCO). It is defined as primary tumor of peritoneum that diffusely involves the peritoneal surface but spares or only superficially invades the ovaries. Better recognition of this entity in recent years has contributed to an increasing diagnostic frequency. Case Report: A case of 50-year-old female who was presented with abdominal distension and pain is reported. Ascitic fluid cytology showed malignant cells favoring papillary serous adenocarcinoma. Preoperative serum CA-125 was markedly elevated. CECT scan showed omental thickening with normal uterus and ovaries. Exploratory laparotomy revealed massive ascites with extensive peritoneal deposits and normal sized ovaries.

Histopathology confirmed diagnosis of PSPCP with surface involvement of both the ovaries. Conclusion: PSPCP is a rare neoplasm, histologically indistinguishable from PCSO. We presented this case report to emphasise that peritoneum, can also be a primary site of malignancy and that it presents and is managed just like primary ovarian cancer. Pre-op diagnosis of this entity is difficult. Histopathology is mandatory to confirm the diagnosis.

Keywords: Papillary, Serous, Peritoneum, Ovary, CA-125

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INTRODUCTION

Primary serous papillary carcinoma of the peritoneum (PSPCP) is a rare malignant epithelial tumor that is histologically indistinguishable from papillary serous carcinoma of the ovary (PSCO). It is considered to originate from embryonic nests of mullerian cells in the peritoneum. This case report describes the clinical, radiological, pathological findings of the entity and to emphasize that peritoneum can also be primary site of malignancy and should be considered in the differential diagnosis of peritoneal carcinomatosis.
CASE REPORT

Case History

A 50-year-old postmenopausal female who was presented with abdominal distension and pain. On examination, abdomen was distended tensely with ascites, but there were no lymph node enlargement, or that of liver and spleen. On pelvic examination, cervix was normal with no mass palpable in fornices but some nodularity and thickening was palpable in pouch of douglas. Preoperative serum CA-125 was elevated to 1850 U/mL (reference range 0–30 U/mL), while CEA had normal value of 0.479 ng/mL. Ascitic fluid cytology was reported as having malignant cells favoring papillary serous adenocarcinoma. Upper and lower gastrointestinal endoscopy were normal. CECT scan showed omental thickening with normal uterus and ovaries (Figure 1). Preoperative diagnosis of PSPCP was made on basis of cytologic and radiologic findings and no evidence of primary disease elsewhere. Exploratory laparotomy revealed massive 3–4 L of ascitic fluid. Extensive peritoneal deposits were seen all over the abdominal and pelvic peritoneum, including large confluent deposits on under surface of diaphragm. Uterus and both ovaries were normal in size. Omentum showed small deposits though there was no caking. Tumor nodules were seen in pelvic peritoneum, pouch of douglas, and urovesical peritoneum. She underwent panhysterectomy along with infragastric omentectomy and removal of other deposits. Residual disease was multiple small deposits all over parietal peritoneum, and over undersurface of diaphragm. Postoperative CA-125 was 127 U/mL. Histopathology revealed PSPCP with surface involvement of both the ovaries. Final diagnosis was stage 3C primary serous papillary adenocarcinoma of peritoneum. Postoperatively, she was placed on three weekly regime of paclitaxel, carboplatin and ifosfamide with mesna as part of adjuvant treatment. She has received three cycles and her latest CA-125, six months postoperative was 23 U/mL. Patient was doing well at the time of last follow up.

Pathological findings

Ascitic fluid cytology showed moderately cellular smears showing predominantly singly dispersed and few 3-dimensional clusters (Figure 2A) of tumor cells. These cells had peripherally placed nuclei, at places showing glandular arrangement (Figure 2B). Tumor cells showed marked pleomorphism with high N/C ratio. The cells had irregular nuclear margin, coarse chromatin, prominent nucleoli, moderate cytoplasm, often showing cytoplasmic vacuolation and cytoplasmic protrusions. Many bizarre and multinucleated tumor giant cells were seen. Few cells with atypical mitosis were also observed. The findings were consistent with diagnosis of malignant cytology favoring serous adenocarcinoma.

Subsequently, panhysterectomy specimen along with omentum was received. Grossly, specimen of uterus with cervix along with bilateral fallopian tubes was unremarkable and both the ovaries were normal in size. Omentum was received as fibrofatty tissue measuring 45x30x5 cm and serial sectioning showed many tiny nodules. Extensive sampling was done from these areas and showed an invasive tumor arranged in solid and complex glandular structures which were partly papillary (Figure 3A). Intervening areas showed extensive desmoplastic response along with proliferation of tumor cells over the omentum surface (Figure 3B). Focal areas of comedo necrosis were also identified (Figure 3C). The papillary structures were lined by several layers of cells with nuclear crowding and high N/C ratio. Cytologically, nuclei were of high grade with vesicular chromatin and prominent nucleoli with frequent mitosis (6-8/10 hpf). The surface of both the ovaries showed tumor deposits involving only ovarian surface epithelium (Figure 3D). The hysterectomy specimen along with both the fallopian tubes did not show any significant pathology. A panel of immunostains was applied comprising CA-125, CK-7, EMA, Vimentin, S-100. Tumor cells showed positivity for CA-125, CK-7 and EMA and rest of the markers were negative (Figure 4A-D). Final diagnosis of PSPCP was made.

DISCUSSION

PSPCP is a rare malignant epithelial tumor that is histologically indistinguishable from PSCO. It is defined
as primary tumor of peritoneum that diffusely involves the peritoneal surface but spares or only superficially invades the ovaries [1]. This entity was first described by Swerdlow [2] in his case report in 1959 as mesothelioma of pelvic peritoneum. Since then several studies [3–5] have established PSPCP as a separate entity and has been reported under different names. The true incidence of PSPCP remains unknown although an estimated relative frequency to ovarian cancer is 1:10 [3]. Better recognition of this entity in recent years has contributed to an increasing diagnostic frequency approaching 18% of laparotomies performed for ovarian carcinoma [3]. We presented this case report to emphasise that peritoneum, can also be a primary site of malignancy and that it presents and is managed just like primary ovarian cancer. Preoperative diagnosis of this entity is difficult but normal sized ovaries radiologically and positive serous cytology in ascitic fluid can be helpful to recognize this lesion. However, histopathology is still mandatory to confirm the diagnosis.

Since, Lauchlan [6] first included the female peritoneum in the definition of secondary mullerian system in 1972; PSPCP is better understood as neoplasm that arises from mesothelial cells under mullerian influence [1]. This theory, therefore, explains why PSPCP behaves like PSCCO in many ways with similar clinical, radiological and immunohistochemical findings with similar sensitivity to platinum based chemotherapy.

PSPCP is mostly reported in elderly females. Like its ovarian counterpart the tumor often presents with abdominal distention and pain [5]. CT and MRI scans suggest omental caking, peritoneal nodules or enhancement with ascitis without ovarian enlargement in most of the patients [1]. CA-125 is markedly elevated in ovarian malignancy and can be used for monitoring the efficacy of therapy and for early detection of recurrence [1, 3].

The diagnostic inclusion criteria of PSPCP were defined by gynecologic oncology group [7] in 1993 to differentiate it from PSCCO as (1) both ovaries must be either physiologically normal in size or enlarged by a benign process, (2) involvement at extra-ovarian sites must be greater than the involvement on the surface of either ovary, (3) the ovarian component must be non-existent or confined to surface epithelium or less than 5x5 mm within the stroma, and (4) the histological and cytological characteristics of the tumor must be predominantly of serous type that is similar or identical to any grade of ovarian serous papillary adenocarcinoma.

Cytologically, closest differential of papillary serous cytology in ascitic fluid is malignant mesothelioma (MM) (Table 1) and poorly differentiated squamous cell carcinoma. Squamous cell carcinoma is relatively rare in effusions and shows dispersed cells with well demarcated cytoplasm as compared to adenocarcinoma. Histologically, PSPCP must be differentiated from MM, benign papillary mesothelioma, metastatic peritoneal carcinomatosis, borderline primary peritoneal serous tumor, endosalpingiosis, psammocarcinoma of peritoneum and pseudomyxoma peritonei [5, 8]. MM is closely related to long term asbestos exposure, has a male predominance, frequent spindle cell component, cytoplasmic eosinophilia and sometimes extensive cell vacuolization. Benign papillary mesothelioma has well formed papillae, mostly lined by one layer of single cell type showing little or no anaplasia or mitosis and absence of invasion into the peritoneum. Morphologically, PSPCP cannot be differentiated from metastatic peritoneal carcinomatosis; diagnosis of later rests on recognizing a
Table 1: Cytological differentiael feature between malignant mesothelioma and serous adenocarcinoma.

<table>
<thead>
<tr>
<th>Features</th>
<th>Malignant mesothelioma</th>
<th>Serous adenocarcinoma</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Cell arrangement</td>
<td>Singly scattered, 3-D clusters, morulae</td>
<td>Singly scattered, 3-D clusters, papillary and acinar</td>
</tr>
<tr>
<td>2. Cell shape</td>
<td>Cuboidal to polygonal</td>
<td>Cuboidal to columnar</td>
</tr>
<tr>
<td>3. Nuclear location</td>
<td>Central</td>
<td>Peripheral</td>
</tr>
<tr>
<td>4. N/C ratio</td>
<td>Unchanged</td>
<td>Increased</td>
</tr>
<tr>
<td>5. Nuclear pleomorphism</td>
<td>-/+</td>
<td>+++++</td>
</tr>
<tr>
<td>6. Nucleoli</td>
<td>Irregular and angular</td>
<td>Conspicuous to prominent</td>
</tr>
<tr>
<td>7. Two tone cytoplasm</td>
<td>Present</td>
<td>Absent</td>
</tr>
<tr>
<td>8. Cytoplasmic vacuolation</td>
<td>-/+</td>
<td>+++++</td>
</tr>
<tr>
<td>9. Psammoma bodies</td>
<td>Absent</td>
<td>Occasionally</td>
</tr>
</tbody>
</table>

primary tumor usually in ovary, fallopian tube or endometrium. Serous psammocarcinoma of peritoneum is a less virulent variation of PSPCP that has been described. It has proportionately larger number of psammoma bodies and less aggressive cytologic appearance.

The treatment of PSPCP consists of debulking surgery including hysterectomy, salpingo-oopherectomy, omentectomy followed by platinum based chemotherapy [9]. Surgery remains critically important for both diagnosis and treatment of PSPCP. Once diagnosis is established and extend of disease documented, maximum cytoreduction becomes primary goal of management. Prognosis of PSPCP is poor. Medial survival time varies between 7 and 28 months while 5 year survival rate ranges from 0 to 26.5% [1]. In few of the case matched control studies, prognosis of PSPCP was similar to that of stage III or IV serous ovarian papillary carcinoma when the same treatment was performed [10].

CONCLUSION

PSPCP is a rare neoplasm, histologically indistinguishable from papillary serous carcinoma of the ovary, which diffusely involves the peritoneum but spares or minimally invades the ovaries. The clinical presentation and behavior is also similar to its ovarian counterpart. Ascitic fluid cytology combined with radiological and clinical data allows a presumptive preoperative diagnosis of PSPCP. However, one needs to histologically study the ovaries before rendering a final diagnosis. We suggest that PSPCP should be included in differential diagnosis when ascites, omental caking, and peritoneal nodules or enhancement are observed in a postmenopausal women with or without ovarian enlargement.

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**Guarantor**
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REFERENCES


Traumatic brachiocephalic artery pseudoaneurysm following penetrating injury on the contralateral side

Victor Kong, Damon Jeetoo, Grant Laing, Damian Clarke

ABSTRACT

Introduction: Trauma to the brachiocephalic artery is uncommon, but is associated with significant morbidity and mortality. The development of pseudoaneurysm following both penetrating and blunt trauma have been well documented, mostly in the setting of blunt trauma. Case Report: A highly unusual case of a patient who developed a pseudoaneurysm of the brachiocephalic artery on the side contralateral to the side of the original penetrating injury has been reported. Conclusion: Pseudoaneurysm of the brachiocephalic artery following penetrating neck injury in the contralateral side is exceeding rare. It may remain symptomatic and present long after the initial injury. Clinician must have a high index of suspicion for such potentially serious injury.

Keywords: Penetrating, Trauma, Brachiocephalic, Pseudoaneurysm

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INTRODUCTION

We present the case of a 21-year-old male with a penetrating neck injury who subsequently developed a large pseudoaneurysm of the brachiocephalic artery on the contralateral side of the original injury.

CASE REPORT

A 21-year-old male was referred to the trauma unit with a single penetrating wound caused by a screwdriver, and located in the lateral, infraclavicular region on the left side. He had no other significant medical history. His blood pressure was normal and equal in both arms. No pulsatile mass was noted. Three weeks before, he initially assessed at another hospital, and a confirmed hemothorax had been treated with an intercostal chest drain. He was subsequently discharged; however, he presented again with an infected wound in the drain-insertion site, with pus discharge. He was hemodynamically stable, with tachycardia (heart rate 90/min), low-grade pyrexia (temperature, 37.6°C), leucocytosis (white cell count, 16x10^9) and a chest radiograph showing a marked opacity at the left base. An empyema thoracis secondary to a retained hemothorax was suspected and treatment with intravenous antibiotics was initiated. A contrast CT scan of the thorax obtained on the following day and
revealed a consolidation in the left lower lobe of the lung, with adjacent air collection. A large round mass was also noted on the right side of the neck, adjacent to the brachiocephalic trunk suspicious of a pseudoaneurysm. Subsequent CT Angiography (64-slice MDCT) of the neck vessels confirmed a large pseudoaneurysm (38.5 mm sagittal, 31.6 mm transverse and 33.2 mm longitudinal), located on the right brachiocephalic trunk, just prior to the bifurcation into the right subclavian and common carotid arteries (Figures 1 and 2). A decision was made to manage the empyema non-operatively. Once it has subsided, he underwent an open repair of the pseudoaneurysm and made an uneventful recovery.

**DISCUSSION**

Trauma to the brachiocephalic artery is uncommon. The exact incidence of this injury is unknown, as most cases with significant injury tend to exsanguinate prior to reaching the hospital [1]. Isolated pseudoaneurysm of the brachiocephalic artery as a result of penetrating and blunt trauma is a well-documented complication [2], but still relatively rare. Many cases reported in the literature were related to blunt trauma, although penetrating trauma is relatively more common [3].

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**Figure 1:** 3D Reconstruction (CTA), with background subtracted for clarity. Large pseudoaneurysm (red arrow) of the brachiocephalic artery.

**Figure 2(A–B):** 3D Reconstruction (CTA), depicting a large pseudoaneurysm (red arrow). The green arrow represents the side of the original penetrating injury.
A wide range of presentations is seen, but many can remain asymptomatic and present many years after the initial injury [4]. Mortality in treated patients can be as high as 38%, and the further increases if multiple vessels are involved [5].

Once the diagnosis is confirmed, patients must undergo definitive management. Traditionally, open surgical repair via median sternotomy and occasionally with cardiopulmonary bypass [6], has been practiced. Recent advances in endovascular techniques have enabled treatment of selected cases with excellent results [7].

The anatomical location of the pseudoaneurysm was highly unusual (as an incidental finding on thorax CT), because it was located on the contralateral side of the original injury. The patient had no other previous injuries and was asymptomatic after the injury. To our knowledge, this is the first case in literature where presentation of the pseudoaneurysm was present on the side contralateral to the injury. The only other case with a similar site of injury was reported by Erkut et al. [8], where a pseudoaneurysm of the right common carotid artery was observed. The trajectory of penetration from the left infraclavicular area to the brachiocephalic artery on the right with no other associated injury can be difficult to appreciate.

Anatomically, the most likely explanation was that the penetrating object followed an anterior course in the upper mediastinum. The common carotid artery and left subclavian artery tend to lie slightly posterior in the sagittal plane to the brachiocephalic trunk. Thus it possible for it to miss the left side vessels and injury the brachiocephalic artery. The alternative was the presence of the pseudoaneurysm prior to injury, but in view of the clear history, seems unlikely.

CONCLUSION

Although pseudoaneurysm of brachiocephalic artery resulting from penetrating trauma is a rare presentation, it remains an important entity because of its implication in subsequent management. Clinicians must remain vigilant of such injury, particularly with delayed presentations. Thorough clinical assessment is mandatory, with judicious use of imaging studies to identify such an injury that has the potential of causing serious adverse outcomes.

********

Author Contributions
Victor Kong – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Damon Jeetoo – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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Damian Clarke – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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REFERENCES
Incidental finding in a patient with trauma and hematuria

Shahnawaz Amdani, Apeksha Sathyaprasad, Aurea P Thomas, Magda Mendez, Elita Gose-Balakrishnan, Shefali Khanna

ABSTRACT

Introduction: Schistosomiasis is the second (to malaria) most important human parasitic disease in tropical and subtropical regions infecting more than 100 million people in sub-Saharan Africa alone. Case Report: We report a case of 17-year-old male who was brought to the Emergency room after being assaulted by his classmates. Apart from the facial injuries, patient had hematuria, which prompted the physicians to undertake additional testing revealing that the patient had *Schistosoma hematobium* infection. Discussion: Microscopic examination of urine and stool samples for parasite eggs are currently the ‘gold’ standard field method of diagnosing schistosomiasis. Praziquantel (PZQ) has been available as an effective treatment of schistosomiasis for nearly 30 years. To date, there is no convincing clinical evidence for schistosome resistance to PZQ. Conclusion: Despite the public health prominence of schistosomiasis in Africa and the availability of a cheap and efficacious drug to treat infected people, less than 5% of the infected population is receiving treatment. An integrated strategy, which emphasizes health education, access to clean water and adequate sanitation, mechanization of agriculture, and fencing of water buffaloes, along with mass chemotherapy for both human and livestock, have been suggested to be carried out in parallel to control the infection sources and to stop schistosome transmission.

Keywords: Schistosome, Hematuria, Praziquantel, Calcification

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INTRODUCTION

Schistosomiasis, commonly known as bilharzia or snail fever, is the second (to malaria) most important human parasitic disease in tropical and subtropical regions [1] infecting more than 100 million people in sub-Saharan Africa alone [2]. In Africa, *Schistosoma haematobium*, the causative agent of urogenital schistosomiasis, is the most prevalent species causing human disease and is responsible for most of the schistosoma-related disease in the region [1]. In affected populations, children carry the heaviest burden of infection; urogenital schistosomiasis causes hematuria, dysuria, nutritional deficiencies, anemia, growth retardation, decreased physical performance and impaired memory and cognition [3].
**S. haematobium** has a complex lifecycle, which includes free-living stages in the environment and asexual reproduction in intermediate freshwater snail hosts as well as maturation and sexual reproduction within mammalian hosts. Humans acquire infection through contact with water bodies containing the infective stages (cercariae) [2].

**CASE REPORT**

A 17-year-old male was brought to the emergency room (ER) after being assaulted by two of his classmates. He was reported to have been struck on the face with fists. On arrival to the ER, patient was hemodynamically stable. He was alert and oriented x 3. He had a Glasgow Coma Scale (GCS) of 15. He had one episode of vomiting of blood in the triage. Eye examination revealed right periorbital edema and ecchymosis. Also right subconjunctival hemorrhage and 1 cm horizontal complex laceration to the right eyelid without involvement of the eyebrow was noticed. There was tenderness to palpation on the site of swelling. There was no diplopia or loss of vision. CN II through XII was grossly intact. On examination of the nose, there was mild tenderness to palpation with bilateral epistaxis. CBC, BMP, LFT, Amylase, Lipase, UA, Brain CT, CXR and Facial bone CT were done. CBC showed eosinophils of 8.6%. LFT, Amylase, Lipase were normal. BMP was significant for high glucose (likely stress hyperglycemia). UA showed blood 3+ and RBC of 50–100. Further workup of hematuria revealed the cause.

**Diagnosis:** Brain CT and CXR were normal. Facial CT revealed right medial orbital wall fracture and right orbital floor fracture. Also a minimally displaced fracture of the left nasal bone was seen. Since the patient had hematuria, the decision was made to get an Abdominal CT with contrast and Bilateral Renal USG to rule out trauma to the kidney. The Bilateral Renal USG was normal. An Abdominal CT revealed 5 mm calcifications in the right hepatic lobe and also in the bladder wall (Figures 1–4).

Serial UAs done during the hospital day # 1 and 2 revealed blood 3+ and RBC 50–100. Considering the patient had hematuria, the decision was made to get Serum Calcium and PTH which were normal. Since the patient was a native of the Ivory Coast in Africa, further workup of urine for ova and parasites were done which revealed that the patient had **Schistosoma haematobium** infestation (Figure 5). Patient was treated with Praziquantel 40 mg/kg two times a day for two days. During the day of discharge UA of the patient showed blood 1+ and RBC of 15–30. No schistosome ova or parasites were detected in the urine.

**DISCUSSION**

The microscopic examination of urine and stool samples for parasite eggs are currently the ‘gold’
standard field method of diagnosing schistosomiasis [3]. It is unable to detect prepatent or single sex infections or reliably detect low levels of infection characteristically found in young children. In the case of *Schistosoma haematobium*, even these low infection levels are clinically relevant since the relationship between infection level and morbidity is not linear but a complex interaction between infection levels and the host’s immune system. Thus, being able to accurately measure this is an important public health aspect [1]. Praziquantel has been available as an effective treatment for schistosome infection for nearly 30 years [4]. PZQ exhibits stage-specific functions in killing adult worms. Multiple doses of 40/60 mg/kg PZQ provide an enhanced efficacy in treating schistosomiasis compared to a single dose. To date, there is no convincing clinical evidence for schistosome resistance to PZQ used for human schistosomiasis treatment, although worrying low-cure rates have been reported in some studies. Adverse effects were dizziness, stomach discomfort or stomach ache, headache, nausea, debility, muscular and joint soreness, and diarrhea, which disappeared shortly after drug withdrawal [5].

**CONCLUSION**

Despite the public health prominence of schistosomiasis in Africa and the availability of a cheap and efficacious drug to treat infected people, less than 5% of the infected population are receiving treatment [3]. An integrated strategy, which emphasizes health education, access to clean water and adequate sanitation, mechanization of agriculture, and fencing of water buffaloes, along with mass chemotherapy for both human and livestock, have been suggested to be carried out in parallel to control the infection sources and to stop schistosome transmission [5]. Also, it is very important to ask about travel history in a patient with hematuria.

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The corresponding author is the guarantor of submission.
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REFERENCES

A case of myocardial metastasis from lung adenocarcinoma presenting as cerebral infarction

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ABSTRACT

Introduction: Myocardial metastasis from lung cancer is rarely found in antemortem settings. Herein we present a case of myocardial metastasis from lung adenocarcinoma presenting as cerebral infarction. Case Report: A 78-year-old Japanese female (non-smoker) who had been diagnosed with poorly differentiated lung adenocarcinoma suddenly complained of weakness in the left hand on a regular follow-up visit. Brain non-contrast-enhanced computed tomography (CT) scan showed no abnormality. Electrocardiography (ECG) showed abnormal Q waves, ST-segment elevation, and inverted T waves in leads II, III, and aVF that had not been demonstrated previously. The results of brain CT scan and symptoms were strongly suggestive of cerebral infarction. Brain magnetic resonance imaging (MRI) with diffusion-weighted imaging performed 5 days after admission showed a small, acute infarct on a knob on the precentral gyrus. Transthoracic echocardiography performed to detect cardiac sources of embolism revealed an intracardiac mass. Cardiac 18F-fluoro-deoxyglucose positron emission tomography (18F-FDG PET)/CT scan demonstrated increased uptake in the mass. After the patient died, autopsy revealed many metastatic lesions of various sizes accompanied by thrombi on the surface of the ventricular myocardium. Conclusion: Cardiac 18F-FDG PET/CT was useful for diagnosing myocardial metastasis in our patient. The possibility of cardiac metastases should be considered in patients with lung cancer in the event of sudden cerebral infarction.

Keywords: Myocardial metastasis, Lung cancer, Cerebral infarction, Cardiac PET/CT


INTRODUCTION

Myocardial metastasis from lung cancer is often clinically invisible, and is thus very difficult to diagnose antemortem. Positron emission tomography (PET)/computed tomography (CT) scan offer additional metabolic information resulting in increased sensitivity for the detection of malignant tumor compared to purely morphological modalities [1]. We present a case
of myocardial metastasis from lung adenocarcinoma presenting as cerebral infarction.

**CASE REPORT**

Our patient was a 78-year-old Japanese female (non-smoker) who had been diagnosed with poorly differentiated lung adenocarcinoma staged as cT2aN2M0, stage IIIA, in October 2008. She had been treated with chemoradiotherapy comprising cisplatin and docetaxel, followed by gefitinib. In February 2009, gefitinib treatment was stopped after the appearance of ground glass opacities in the lung. In June 2009, stereotactic radiotherapy was performed for local recurrence. Follow-up screening for metastasis with CT of the whole body, magnetic resonance imaging (MRI) of the brain, and bone scintigraphy in September 2009 showed negative results.

The patient suddenly complained of weakness in the left hand during a regular follow-up visit in October 2009. Neurological examination showed a positive Barré sign in the left upper extremity. No abnormalities were apparent on physical examination other than coarse crackles in the right lower lung field. Brain non-contrast-enhanced CT scan showed no abnormalities.

Blood testing showed normocytic anemia with a hemoglobin level of 9.8 g/dL. Serum creatinine level was 1.07 mg/dL and C-reactive protein concentration was 0.20 mg/L. Congestive heart failure was suspected based on the increased cardiac brain natriuretic peptide (BNP) level of 257.0 pg/mL. Levels of fibrin degradation products (FDP) and D-dimer were 43.6 µg/mL and 13.7 µg/mL, respectively. All other results of biochemical testing were normal.

Electrocardiography showed abnormal Q wave, ST-segment elevation, and inverted T waves in leads II, III, and aVF that were not demonstrated in September 2009, but the patient reported no cardiac symptoms (Figure 1A).

Results of brain CT and patient symptoms were strongly suggestive of cerebral infarction. Treatment was started with edaravon and argatroban and neurological symptoms gradually improved. Brain MRI with diffusion-weighted imaging performed five days after admission showed a small, acute infarct on a knob on the precentral gyrus (Figure 1B–C). Magnetic resonance angiography of the circle of Willis yielded normal results.

Transsthoracic echocardiography performed to detect cardiac sources of embolism revealed an intracardiac mass arising from the ventricular septum close to the apex (Figure 2A). Cardiac CT demonstrated a low-density mass located in the wall of the left ventricle (Figure 2B). Cardiac 18F-fluoro-deoxy-glucose (FDG) PET/CT demonstrated increased uptake in the mass in the inferior wall of the left ventricle close to the apex (Figure 2C–E). These findings were strongly suggestive of myocardial metastasis from lung adenocarcinoma.

On the other hand, brain MRI with gadolinium-enhanced T1-weighted imaging performed 32 days after...
admission showed enhancement at the same site demonstrated previously on MRI.

The patient was treated with erlotinib for recurrent lung adenocarcinoma, but died soon afterwards. Autopsy was performed, revealing many metastatic lesions of various sizes accompanied by thrombi on the surface of the ventricular myocardium (Figure 3).

![Figure 3: (A) Gross and (B) microscopic findings of cardiac metastases at autopsy. (A) Multiple metastases of various sizes in the myocardium were demonstrated in the resected heart. (B) Adenocarcinoma cells were demonstrated not only in myocardium but also in thrombus on the myocardial surface. (HE, x100 magnification).]

DISCUSSION

Metastasis to the pericardium or heart is reportedly present in 30-45% of autopsies for lung cancer patients [2, 3]. Pericardium, including epicardium, is the most common location of cardiac involvement by secondary tumors [4], with myocardial metastasis reportedly comprising only about 10% of metastases to the heart [3]. Myocardial metastasis from malignant tumor is often clinically invisible, and can be very difficult to diagnose antemortem. Indications of metastases to the heart are often described as a rapid increase in heart size due to pericardial effusion, new signs of heart failure or valve disease, conduction defects, and atrial or ventricular heart rhythm disturbances [5]. In our patient, abnormalities were seen on ECG but the patient reported no cardiac symptoms.

Regarding the mechanisms of cardiac metastasis, it is reported that metastatic cells can reach the heart via the lymphatic or hematogenous route, or by direct or transvenous extension. In our patient, tumor cells seem to have attained to the heart mainly via hematogenous route considering the fact that hematogenous spread preferentially gives rise to myocardial metastasis [5].

Cerebral infarction has also been reported to result from obvious thromboembolism, probably from the surface of a metastatic tumor [6]. In this patient, cerebral infarction was the first presentation of cardiac metastasis from lung adenocarcinoma. Such embolic phenomena may be due to either tumor emboli or bland emboli that have dislodged from the tumor thrombus on the surface of the myocardial metastasis. The possibility of cardiac metastasis should be kept in mind for patients with lung cancer who suddenly present with cerebral infarction.

To explain the mechanisms of unexpected cerebral infarction in a patient with malignant tumor, the possible presence of Troussseau’s syndrome should be considered. Troussseau’s syndrome represents unexplained thrombotic events that precede the diagnosis of an occult visceral malignancy or appear concomitantly with the tumor. This syndrome is considered to be mediated by multiple mechanisms such as tissue factors, tumor-associated cysteine proteinase, tumor hypoxia, carcinoma mucins, or an overlap of these as a cause of prothrombotic condition [7]. Besides the presence of myocardial metastasis, the influence of Troussseau’s syndrome might be one cause of cerebral infarction.

For the diagnosis of metastasis to the heart, echocardiography, enhanced CT, enhanced MRI, and the combination of these modalities may prove helpful [5]. The advantage of cardiac PET/CT scan for detecting myocardial metastasis has recently been reported [8]. Compared to purely morphological modalities, PET/CT scan offers additional metabolic information resulting in increased sensitivity for the detection of regional tumor growth and/or distant metastases, facilitating staging and follow-up of patients with malignancies [1]. In our patient, distinguishing metastatic cardiac tumor and thrombus was difficult using only echocardiography and contrast-enhanced CT scan. On the other hand, cardiac PET/CT scan demonstrated focally abnormal increases in the uptake of FDG in myocardium and correctly suggested metastatic tumor. Cardiac PET/CT scan is usually used for the detection of coronary artery disease (CAD), and is rapidly gaining popularity as a powerful and efficient alternative to conventional single-photon emission CT to evaluate regional myocardial perfusion and metabolism in patients with CAD [9]. With respect to cardiac tumors, cardiac PET/CT scan is useful not only for the detection of myocardial metastasis but also for specifying the exact location of the tumor compared with conventional PET/CT.

In contrast, a case report has described a patient with underlying malignancy who was misdiagnosed with cardiac metastasis due to an imaging pitfall in PET/CT scan. Efforts should therefore always be made to rule out false-positive interpretations [10].

CONCLUSION

We report here on a case of myocardial metastasis from lung adenocarcinoma that presented as cerebral infarction. Cardiac PET/CT was a useful modality for diagnosing myocardial metastasis in our patient. The possibility of cardiac metastasis should be kept in mind for patients with lung cancer who develop sudden cerebral infarction.

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Guarantor
The corresponding author is the guarantor of submission.

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Authors declare no conflict of interest.

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REFERENCES

Synchronous nasal metastasis from pulmonary squamous cell carcinoma

Liling Zhang, Junli Liu, Tao Wang, Gang Wu

ABSTRACT

Introduction: Squamous cell carcinoma spread from lung to the nasal cavity is extremely rare. Because of the identical appearance under light microscope, making a definitive diagnosis is a challenge when lesions are from both the nasal cavity and lung. Case report: We present a case of a 56-year-old patient with simultaneous malignancies in the lung and nasal cavity. On the basis of histologic analysis and radiographic features, a diagnosis of primary lung cancer with nasal metastasis was established. Since the histological appearance of the lung tumor and the nasal lesion is similar, the differential diagnosis between second primary and metastasis mainly depends on clinical criteria. However, the accuracy of this approach remains in question. Conclusion: It is a challenge to discern the true relationship of lung squamous cell carcinomas and nasal squamous cell carcinomas. The criteria currently employed in the distinction are mainly dependent on clinical, radiographic, and histologic grounds. Recent studies demonstrated that molecular genetic analysis can be a promising approach to solving this diagnosis dilemma.

Keywords: Squamous cell carcinoma, Lung cancer, Cancer of nasal cavity, Metastasis, Differential diagnosis

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INTRODUCTION

Nasal metastasis from primary lung cancer is extremely rare. The frequency of nasal metastasis in lung cancer has never been evaluated, since very few cases have been reported to date, and most common histologic subtype is adenocarcinoma [1]. The diagnosis of metastasis can be easily made when the patient had a history of previous malignancy, and the histologic features are distinctive and characterize the organ of the primary lesion as in renal cell carcinoma metastatic to the nasal cavity [2]. However, when nasal lesions simultaneously present with primary lung cancer, particularly both histologic types are squamous cell carcinomas, distinguishing metastasis from second primary will be a challenge. We present a case of a 56-year-old patient with simultaneous malignancies in the lung and nasal cavity. The diagnosis of primary lung cancer with nasal metastasis was established on the basis of histologic analysis and radiographic features. The difficulty encountered in differential diagnosis is discussed and approaches to solve this diagnostic dilemma are reviewed.
CASE REPORT

A 56-year-old male patient was presented with an enlarging mass on the left side of his nasal root, accompanied with occasional epistaxis, nasal obstruction and numbness over the left cheek for one and half month duration. Physical examination revealed a 2×3 cm firm, fixed mass on the left side of the nasal root. Paranasal sinuses CT scan showed a soft tissue mass surrounding the left frontal process, with irregular osteolysis of the frontal process and anterior wall of maxillary sinus (Figure 1A). The mass infiltrated the left nasal cavity, left maxillary sinus, alveolar process, and subcutaneous tissues overlaying it. Biopsy of the mass demonstrated the nasal neoplasm was a squamous cell carcinoma (Figure 1B). To further evaluate the staging of the patient, a chest CT scan was performed. The result showed a lesion in the right middle lobe near the right hilum (Figure 2A). The lesion was sized 2×3.5 cm with uneven density, irregular shape and blurry margin. No hilar and mediastinal lymphadenopathy was observed. Fibrebronchoscopy revealed that a 2×3.5 cm cauliflower-like neoplasm located in the right middle lobar bronchus and biopsy of the lesion demonstrated a squamous cell carcinoma too (Figure 2B). Except serum CA19-9 was slightly increased, other auxiliary examinations of the patient were normal, which including abdominal CT, pelvic CT and bone scans. Thus, on the basis of combined analysis of histologic and radiographic features, the clinical diagnosis was established as primary lung cancer with nasal metastasis (stage IV).

The patient underwent two cycles of chemotherapy with gemcitabine (1000 mg/m², day 1 and 8) and cisplatin (75 mg/m², day 1), and the palpable mass on the left nasal root shrank. Further plans for four such cycles abandoned due to financial constraints. The patient occurred cough and dyspnea and died from respiratory failure six months after initial diagnosis.

DISCUSSION

Tumors arising in two different organs, such as lung and nose, can represent either a primary tumor with a metastasis or two primary tumors. Diagnosis of a metastatic disease may be easily made when the histologic features are distinctive and characterize the organ of the primary lesion. As reviewed by Huang et al. [1], among 16 reported cases to 2009, the most common histologic subtype of the lung is adenocarcinoma, which resulting in an easy definitive diagnosis, and only one case with squamous cell carcinoma [3] was included in the review. As in our case, when both nasal and pulmonary lesions are squamous cell histology, the differential diagnosis between metastasis and second primary tumor is extremely difficult. In such case, the differential diagnosis includes primary nasal tumor with lung metastasis, primary lung cancer with nasal metastasis, and synchronous nasal and lung cancer.

There is no gold standard in the differential diagnosis of metastasis versus second primary tumor in patients with a squamous cell carcinoma of the lung and a squamous cell lesion of the nasal cavity. The criteria currently employed in the distinction are mainly dependent on clinical, radiographic, and histologic grounds. Such parameters as tumor stage, time interval between two tumors detected, histologic grade, and radiographic presentation are frequently mentioned in literature [4]. Since both are squamous cell carcinomas and have identical features under light microscopy, histologic classification is of negligible help in this case, while radiographic features may be an important aid in the differential diagnosis.

The fundamental distinction between a lung metastasis and a primary lung cancer is usually straightforward on radiographic grounds [5, 6]. A smooth nodal margin can be regarded as an indication of a metastasis, which often peripherally located. However, a primary lesion often presents with irregular or blurry margin and can be peripherally or centrally located. Squamous cell carcinoma of lung is more often centrally located within the lung, whereas adenocarcinoma is typically peripherally located. CT features of this patient’s pulmonary lesion, including irregular shape, blurry margin and central location, suggested a primary cancer. Further fiber bronchoscope biopsy confirmed the diagnosis of primary lung cancer.

Although radiographic features of metastatic nasal lesions are similar to those of primary nasal neoplasms, localizing the epicenter of the tumor may aid in determining its origin [7]. In our case, the mass epicenter is located at the frontal process with invasion
of nasal cavity and facial subcutis, but the histology is squamous cell carcinoma which is seldom seen in bone tumors. Therefore, the nasal mass was regarded as a metastatic lesion from lung squamous cell carcinoma.

Although we had made the diagnosis based on the currently employed clinical criteria, the accuracy of this approach remains in question. Since making correct diagnosis has great influence on patient prognosis and could rationally guide therapeutic strategies, some molecular genetic methods have been recently developed for discerning the true relationship of lung squamous cell carcinomas and head and neck malignancies.

Leong et al. [6] and Geurts et al. [4] performed loss of heterozygosity (LOH) analysis for paired tumors from head and neck squamous cell carcinoma and solitary lung nodules to assess the origin of the tumor. Those studies suggested that comparison of genetic alterations in the tumors can be very helpful in distinguish metastasis and second primary tumor. Moreover, the use of gene expression profiling has demonstrated the potential to resolve this diagnostic dilemma. Vachani et al. [8] identified a panel of 10 genes (CXCL13, COL6A2, SFTPBI, KRT14, TSPYL5, TMP3, KLK10, MMP1, GAS1, and MYH2) that accurately distinguished these two tumor types. This 10-gene classifier showed a high accuracy of 96% on the samples from the Talbot et al. study [9].

CONCLUSION

It is a challenge to discern the true relationship of lung squamous cell carcinomas and nasal squamous cell carcinomas. They could be metastasis from either, or both are primary. The criteria currently employed in the distinction are mainly dependent on clinical, radiographic, and histologic grounds. As in our case, the final diagnosis was made on the basis of these criteria, although the accuracy of these criteria remains in question. Recent studies demonstrated that molecular genetic analysis, such as LOH and gene expression profiling, can be a promising approach to solving this diagnosis dilemma.

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Author Contributions

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Junli Liu – Acquisition of data, Critical revision of the article, Final approval of the version to be published
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REFERENCES

Giant cellular neurilemmoma, a rare mesenchymal tumor of the oesophagus: A case report

Mitali Singhal, Vatsala Misra, Vishal Dhingra, Sri Prakash Misra

ABSTRACT

Introduction: Gastrointestinal schwannomas are classified as mesenchymal or neuroectodermal neoplasms. Most common site is stomach followed by rectum. Schwannoma of oesophagus is very rare. It can present with dysphagia and odynophagia. Case Report: A 30-year-old female was presented with progressive dysphagia, mild odynophagia, retrosternal heartburn and vomiting for last six months. On radiological investigations a rounded radio opaque shadow in right paratracheal region was seen. Endoscopy showed ulcerated mucosa that bled on touch. Lumen was narrowed. Endoscopic biopsy showed only mild dysplasia in squamous epithelial lining. Segmental oesophagectomy was done to remove the mass. On gross examination an already cut open segment of oesophagus of about 6 cm in length was received. An irregular, lobulated, firm, grey white growth, of 7x5x8.5 cm, having smooth external surface was observed towards serosal surface. Cut surface was homogenous white. Histology showed monomorphic spindle shaped cells arranged in fascicular and whorled pattern with nuclear palisading at places. Peri tumoral lymphoid aggregates were also seen.

Conclusion: Oesophageal schwannomas are benign tumors having excellent prognosis following surgical resection. A case of oesophageal schwannoma is documented here.

Keywords: Oesophagus, Schwannomas

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INTRODUCTION

Schwannoma or neurilemmoma is a benign tumor of the peripheral nerves. Most common sites are flexor aspects of extremities, neck, mediastinum, retroperitoneum, posterior spinal roots and cerebellopontine angle. Schwannomas of the gastrointestinal tract are rare and distinctively different neoplasms from conventional schwannomas that arise in soft tissue or the central nervous system [1]. Gastrointestinal schwannomas occur most commonly in the stomach (60–70% of cases), followed by the colon and rectum [2–5]. Esophageal and small-intestinal schwannomas have been rarely reported [2–4]. The most common mesenchymal tumor occurring in oesophagus is leiomyoma [6]. Esophageal schwannoma is a rare and different neoplasm from conventional schwannomas arising in soft tissue or the central nervous system. Peak incidence is in the third and fourth decade of life. These are more common in females as compared to males. Esophageal schwannoma like other gastrointestinal schwannomas
show distinctive histological features that separate them from conventional schwannomas. On microscopic examination, Oesophageal schwannomas show spindle cells with a microtrabecular pattern, peripheral lymphoid cuffing with occasional germinal centers. Cells show S-100 positivity [1–3]. They do not show nuclear palisading that is usually present in conventional schwannomas. Gastrointestinal schwannomas lack neurofibromatosis-2 genetic alterations supporting the theory that gastrointestinal schwannomas are unique tumors that are distinct from conventional schwannomas [7]. To date about 30 cases of oesophageal schwannoma have been documented in literature [8–15]. A case of oesophageal schwannoma is documented here due to its rarity and unusual presentation.

CASE REPORT

A 30-year-old female was presented with progressive dysphagia, mild odynophagia, retrosternal heartburn and vomiting for last six months. On radiological investigations, a round radio opaque shadow in the right paratracheal region was seen. CT scan revealed a large (7x5x8.5 cm) lobulated homogenous mildly enhancing soft tissue mass in tracheoesophageal groove extending in right paratracheal and subcarinal region. Endoscopy showed ulcerated mucosa which bled on touch. Lumen was narrowed. Endoscopic biopsy showed only mild dysplasia in squamous epithelial lining. Segmental oesophagectomy was done to remove the mass.

On gross examination an already cut open segment of oesophagus of about 6 cm in length was received (Figure 1A). An irregular, lobulated, firm, grey white growth, of 7x5x8.5 cm, having smooth external surface was observed towards serosal surface. Cut surface was homogenous white (Figure 1B).

Multiple sections were processed and showed variable picture on histopathological examination. Sections processed from mucosal surface showed hyperplastic stratified squamous epithelial lining underneath which areas of hemorrhage and fibrocollagenous tissue were seen (Figure 1C). Fibrocollagenous tissue was compressed and surrounded by lymphocytes and plasma cells extending into the underlying mass (Figure 1D). Sections from tumor area showed monomorphic spindle shaped cells surrounded by fibrocollagenous tissue. Cells were arranged in fascicular and whorled pattern. Cells had poorly defined eosinophilic cytoplasm and pointed basophilic nuclei with nuclear palisading at places (Figure 2A–C). Some of them showed large wavy nuclei. Occasional mitotic figures were seen. Thickened blood vessels and perivasculare hyalinization was present along with moderate amount of inflammatory infiltrate mainly (lymphocytes and plasma cells). Peritumoral lymphoid aggregates were also seen.

Immunohistochemistry for S-100, Desmin and CD 117 was done. Tumor was strongly positive for S-100 (Figure 2D) and negative for CD 117 and desmin.

![Figure 1](image1.png)

Figure 1: (A) Lobulated oesophageal mass with attached mucosa, (B) Cut surface is homogenous white with overlying ulcerated and degenerated mucosa, (C) Hyperplastic stratified squamous epithelial lining with areas of hemorrhage and a submucosal growth, and (D) Peritumoral lymphoid aggregates are seen.

![Figure 2](image2.png)

Figure 2: (A) Section showing spindle shaped cells arranged in whorled pattern, (B) Area showing spindle shaped cells arranged in palisading pattern, (C) Higher magnification of Fig2B showing palisading arrangement of spindle shaped cells with ovoid to spindle nuclei and mild lymphocytic infiltrate, and (D) Immunohistochemistry for s-100 showing strong positivity.

DISCUSSION

Oesophageal schwannomas are benign tumors having excellent prognosis following surgical resection. Benign tumors of oesophagus are more common in men than women in the oesophagus most common soft tissue tumors are leiomyomas [6, 16]. Others are leiomyosarcomas and GIST. It is difficult to distinguish schwannoma from leiomyoma, and GIST. So preoperative diagnosis is difficult and can be confirmed.
only by histopathological and immunohistological examination. Oesophageal schwannomas like other gastrointestinal schwannomas are not encapsulated, a feature that distinguishes them from schwannomas in peripheral nervous system. On histopathological examination, these schwannomas have a lymphoid cuff with germinal centre. They are composed of interlacing bundles of spindle cells which show only loose palisading. They may resemble GISTs but the presence of lymphoid cuff helped in diagnosing it as schwannomas in this case. The tumor cells of oesophageal schwannoma are positive for S-100 protein and negative for smooth muscle markers, such as actin and desmin, which are positive in myogenic tumors. Oesophageal schwannoma tumor cells are also negative for CD34 and CD117, which are positive in GIST [1, 4]. In present case, mass showed spindle shaped cells having wavy nuclei, arranged in fascicular and whorled pattern with evidence of nuclear palisading at places. Peritumoral lymphoid aggregates were also seen suggesting a diagnosis of Schwannomas. No significant difference in oesophageal schwannoma and other gastrointestinal schwannomas has been documented in literature. Leiomyomas are benign tumour showing perpendicularly oriented fascicles of brightly eosinophilic spindle cells with blunt ended cigar shaped nuclei and sometimes Para nuclear vacuole [16]. They are positive for desmin, smooth muscle actin, calponin and caldesmon. Gastrointestinal stromal tumors show differentiation along interstitial cells of Cajal and CD117/ c-kit positivity on immunohistochemistry. They may behave in benign and malignant fashion. On microscopic examination, they are spindle cell or epithelioid type, sometimes with skeneoid fibers in stroma [17]. Though oesophageal schwannomas can be treated by enucleation [18], Partial oesophagectomy has been used to treat Large benign schwannomas [19].

CONCLUSION

Oesophageal schwannomas are benign tumors having excellent prognosis following surgical resection. A case is documented here due to its rarity and unusual presentation.

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Author Contributions

Mitali Singhal – Acquisition of data, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published.

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The corresponding author is the guarantor of submission.

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Pneumoperitoneum is not always an indication for laparotomy: A case report

Oleg Ponomarenko, Ohn Sibirsky, Sergio Susmallian

ABSTRACT

Introduction: Pneumoperitoneum is a radiological term that is used to describe an abnormal collection of free gas in the peritoneal cavity but outside the viscera. In the majority of cases (>90%), it is a result of perforation of intra-abdominal viscus with serious consequences and it needs for emergency surgical management. Pneumoperitoneum reflex to the surgeons to proceed to an exploration. Laparotomy and general anesthesia are associated with significant morbidity, therefore it is important to recognize idiopathic spontaneous pneumoperitoneum and treat it appropriately. Case Report: A case of 67-year-old male patient with a massive idiopathic spontaneous pneumoperitoneum that was operated without any cause of free air with a benign evolution during the postoperative period is reported. Conclusion: The absence of clear signs of peritonitis and sepsis allow the possibility to perform more studies as computed tomography scan and not to follow the suggestion of ‘any pneumoperitoneum require exploration’. Correct management of those cases can be treated conservatively avoiding unnecessary operations.

Keywords: Pneumoperitoneum, Pneumatosis, Laparotomy

INTRODUCTION

Pneumoperitoneum is a radiological term that is used to describe an abnormal collection of free gas in the peritoneal cavity but outside the hollow viscera. In the majority of cases (>90%), it is a result of perforation of intra-abdominal viscus with serious consequences and it needs for emergency surgical management [1]. Pneumoperitoneum reflex to the surgeons to proceed to an exploration. Laparotomy and general anesthesia are associated with significant morbidity, therefore it is important to recognize idiopathic spontaneous pneumoperitoneum and treat it appropriately.

We present a case of a patient with bloody stool and abdominal distention with radiological pneumoperitoneum.

CASE REPORT

A 67-year-old male with a history of umbilical hernia and underlay mesh repair twenty years ago, without any medication, presented to the emergency department with complaints on single fresh bloody stool passing and increasing abdominal distension for the last day. It was a first episode in his life. From anamnesis was known about mild constipation without weight loss history. There were no associated gastrointestinal or systemic symptoms. The patient was hemodynamically
stable. Respiratory rate 16/min. On physical examination his abdomen was markedly distended, tympanic to percussion but not tender and without any peritoneal signs. The surgical scar from umbilical hernia repair was normal without signs of recurrence and without tenderness. A left inguinal hernia was found without signs of incarceration. Plain radiographs of the chest (Figure 1) and abdomen (Figure 2) revealed a large pneumoperitoneum with small bowel and colon distention. Blood count, biochemical screening and blood gas analyses were within normal limits. Rectoscopy was subsequently performed and this also showed normal mucosal pattern. He was admitted to the hospital. An exploratory laparotomy was performed with large quantities of entirely odorless gas rushed out of the abdomen, all the peritoneal cavity and retroperitoneum was minousciously checked. There was no evidence of perforation or fluids. We found distention of small and large bowel looked like non-mechanical paralytic ileus. During the exploration, we found mild sigmoid diverticulosis and gallbladder stone without any signs of inflammation. The computed tomography scan was performed on the second day after operation and it showed small amount of free gas and fluid, distention of small bowel without signs of leakage of contrast and no inflammation. The patient was managed of broad-spectrum intravenous antibiotics and intravenous hydration with pain killer. On the third day after operation, the patient was started to pass gases and on the fourth day, he passed diarrhea bowel movement. The patient took a regular diet and was discharged on the sixth day after operation.

DISCUSSION

The first mention of pneumoperitoneum in literature was by Kelling [2] in 1902 who suggested its induction for diagnostic purposes. Popper in August 1915 first called attention to the possibility of pneumoperitoneum in ruptured peptic ulcer [3]. Vaughan and Brams demonstrated the presents of subphrenic free gas in 26 of 29 cases of acute perforation of peptic ulcer [4]. Although perforated colon or small bowel may also present with pneumoperitoneum.

Pneumoperitoneum without evidence of visceral perforation has been reported in 5% to 14% of all occurrences [5]. In April 1915, Weiland had found a similar sign in a patient with perforated ulcer, but necropsy showed that this radiolucent zone above the liver was due to the presence of transverse colon in the zone [6]. An alternative radiological sign suggesting intra peritoneal free gas was described by Rigler in 1941, as the ability to visualize the outer as well as the inner wall of the bowel on plan X-ray in the supine position [7].

About 10% of all cases of pneumoperitoneum are caused by physiologic processes that do not require surgical management. Chandler et al. were the first to doubt on the relevance of this sign when they reported 11 of 29 patients having pneumoperitoneum in the absence of peritonitis [8]. Hinkel in 1940 reported a case of spontaneous pneumoperitoneum without peritonitis, demonstrable visceral perforation or exogenous origin [9]. Since then there have been

Figure 1: Chest X-ray showing free peritoneal air.

Figure 2: Plain abdomen showing small bowel and colon distention and signs of pneumatosis cystoides intestinalis.
sporadic reports in literature highlighting various non-surgical conditions which predispose to spontaneous pneumoperitoneum, where laparotomy is unnecessary. There is some kind of classification of non-surgical causes of spontaneous pneumoperitoneum as thoracic causes of non-surgical pneumoperitoneum, abdominal causes, gynecological causes and miscellaneous causes that includes use of cocain, dental extraction, diving with decompression, scleroderma and idiopathic [10]. But almost all of them have any sources like some disease or manipulation. Occasionally, as in this case, the diagnosis was never established and one may only speculate as to the underlying etiology.

The most common abdominal cause of non-surgical spontaneous pneumoperitoneum is pneumatosis cystoides intestinalis, also referred as lymphomatosis or enteromesenteric emphysema [11]. John Hunter first recognized this condition and contributed two specimens from hog intestine to the museum of the Royal College of Surgeons that demonstrated multiple gas-filled cysts beneath the serosal layer, the first pathologic description is attributed to DuVernoi from a cadaver dissection in 1730 [12]. Pneumatosis cystoides intestinalis is characterized by multiple intramural gas-filled cysts that may be throughout the gastrointestinal tract but are most commonly found at the terminal ileum. The condition is generally benign and asymptomatic. It is generally considered to be a primary idiopathic phenomenon or secondary to another clinical condition such as chronic obstructive pulmonary disease, connective tissue disease, asthma, inflammatory bowel disease and intestinal obstruction. Of 213 cases reviewed, Koss found that 85% were secondary to other underlying causes [13]. Theories of etiology include a mechanical theory in which air originates from the chest and reaches the abdomen via a perivascular plane, supporting the association with chronic obstructive airways disease [14], a bacterial theory in which gas-producing organisms are thought to penetrate the intestinal mucosa producing cysts, an inflammatory process, and several others. Rarely, spontaneous pneumoperitoneum is reported following tracheostomy, adenotonsillectomy, dental extraction, aerophagia, scleroderma, amyloidosis and in cocaine addicts [15]. When the cysts rupture, a pneumoperitoneum may be produced with a spectrum of symptoms ranging from asymptomatic to acute abdominal pain. The condition generally resolved spontaneously but may be indolent and recurrent. Treatment with hyperbaric oxygen and antibiotics has demonstrated some efficacy in reducing long-term symptoms from this disease [16].

CONCLUSION

Spontaneous or non-surgical pneumoperitoneum is an uncommon pathology but it is important to identify patients with this condition from among the larger group with intraperitoneal free gas and prevent unnecessary laparotomy. Generally, most of the patients with pneumoperitoneum have peritonitis and require immediate abdominal exploration and treatment. The absence of clear signs of peritonitis and sepsis allow the possibility to perform more studies as computed tomography scan and not to follow the suggestion of ‘any pneumoperitoneum require exploration’.

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Author Contributions
Oleg Ponomarenko – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Ohn Sibirsky – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
Sergio Susmallian – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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Conflict of Interest
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Ear damage by high voltage electrification in a Mexican worker

Guadalupe Aguilar-Madrid, Arturo Torres-Valenzuela, Yvette de la Fuente-Rivera, Vanessa Crystal Sánchez-Escalante, Luis Cuauhtémoc Haro-García, Cuauhtémoc Arturo Juárez-Pérez

ABSTRACT

Introduction: Altered hearing has been reported in persons who received electrical discharge by lightning. Case report: The case of a Mexican worker electrified by a high voltage (23 kV) electrical current is described as having hearing loss, absent otoacoustic emissions, and increased latency of all waves and interwave intervals of auditory evoked potentials of the brainstem. The worker showed permanent hearing loss post high voltage electrocution (23 kV), with progressive sensorineural hearing loss and perturbed neural conduction of the auditory nerve, and disturbed neuroconduction of the auditory nerve following to a high voltage electrical discharge. Conclusion: Given the implications in rehabilitation and the social and legal consequences patients should be fully evaluated, including a complete hearing study.

Keywords: Hearing loss, Electrification, High voltage, Work accident

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INTRODUCTION

Electricity generation has increased the risk of occupational accidents due to high voltage electrical discharge (HVED) >1000 volts (electrification) [1]. In the United States there have been reports of low voltage electrification <1000 volts in 20% and with high voltage electrification in 38% of the general population. Half of these cases are of occupational origin [1, 2]. Due to the seriousness of the injuries, medical care focuses on the patient’s survival. However, in the HVED survivors have been reported osteomuscular, cardiovascular, neurological, renal, visual, and other types of damage [2, 3]. Hearing loss and tinnitus were reported in one case of HVED, but were reversible [4]. Experimentally, alterations in the posterior cochlea following HVED in guinea pigs have also been reported [5].

Moreover, these alterations were observed in patients who received an atmospheric electric discharge (lightning) [6–9]. These cases included perforation of the tympanic membrane, conductive hearing loss,
sensorineural hearing loss, ossicular disruption, and perilymphatic fistula in the oval window with mixed type hearing loss [6–9]. The present report describes the case of a worker who underwent HVED (23 kV) and survived; however, hearing damage was irreversible.

CASE REPORT

A 38-year-old male working at an electrical supply company installing and repairing electrical service (ISCO-088-7245) who dealt with 6, 13.2, and 23 kV electricity supply wires [10] and had been held the workplace for 12 years with no significant personal or family background of auditory symptoms and no previous exposure to organic solvents. He underwent fluctuating exposure to environmental noise due to urban vehicle traffic. On 11 September 2006, he suffered a HVED with direct 23 kV current in both hands, while working. He lost the alert status and was hospitalized. Four reconstructive surgeries were required due to the deep second degree burns. Five weeks after the accident he regained consciousness although failed to remember events prior to, during, or after the work accident. Once discharged from the hospital, the worker reported hearing loss and tinnitus, and thus was evaluated at the audiology ward of the Specialty Hospital Siglo XXI at National Medical Center of the Mexican Social Security Institute (IMSS).

Since discharge from the hospital, patient underwent five audiological evaluations: at three, six, twelve, eighteen, and twenty months so as to identify a specific pattern of cochlear damage as well as any fluctuation in the hearing threshold. The first three hearing evaluations were performed using a Beltone 2000 audiometer; in the fourth and fifth evaluations a Madsen Orbiter audiometer was used.

The fifth evaluation assessed otoacoustic emissions (OAE) with a Madsen Capella analyzer, in the modes of distortion product (DPOAE), transient (TOAE), and spontaneous (SOAE). Auditory brainstem response (ABR) were also obtained using multimodal evoked potentials equipment (Nicolet Viking Quest).

Otoscopy showed normal tympanic membranes and tuning forks with symmetrical decrease in the hearing acuity. Pure Tone Average (PTA) was determined for each ear. Audiometry evidenced right superficial sensorial hearing loss which evolved to severe sensorial hearing loss, while the left ear had superficial sensorial hearing loss that evolved into moderate sensorial hearing loss (Figure 1). Phonemic discrimination also showed progressive bilateral degeneration at the same period. Otoacoustic emissions by distortion products were absent in both the ears.

Twenty months after the accident, ABR were performed under physiological sleep, using 2000 clicks of alternate polarity at an intensity of 100 dB SPL and with mask at 80 dB SPL, at a stimulation rate of 33.1 clicks/second and 10 ms analysis duration. The ABR showed adequate of I–III and V waves, prolonged absolute latency of I, III and V waves, and bilateral elongation of absolute latency waves (I, III, and V). Moreover, interwave elongation of latencies was observed (I–III, III–V, and I–V) with respect to the equipment’s reference parameters (Table 1).

DISCUSSION

Audiological evaluation of the case revealed the presence of bilateral sensorineural progressive hearing loss; this finding suggests irreversible hearing damage. Jindal et al. [4] published a similar HVED case, in which they have also observed normal tympanic membranes and bilateral sensorineural hearing loss. However, this case was reversible.

Otoacoustic emissions were absent in the present case due to detriment of the hearing threshold, a difference with Jindal’s [4] case, where the emissions were also absent in both the ears, but they were recovered in a month. As regards ABR, absolute latencies of components I, III, and V and interwave latencies (I–III, III–V, I–V) were prolonged compared to reference parameters (Table 1). In the study by Jindal et al. [4], prolonged absolute latencies were documented but came back to normal one month after the event, where the interwave intervals were normal.

Despite similar results between this study and that of Jindal et al. [4], the present case report displayed progressive hearing loss, absence of otoacoustic emissions, and prolonged absolute latencies as well as interwave intervals.

A probable explanation for the differences between this and report of Jindal et al. [4] could be the severity of electrification, due to the difference in electrical tension, or the type of electrical current. In the present study the damage was caused by a direct current, whereas Jindal have not mentioned the type of current. This is an important piece of information because experimentally in guinea pigs [5] caused a decrease in otoacoustic emissions by administering alternate and direct current stimuli throughout ten days. To explain hearing damage, Jindal et al. proposed that the HVED causes functional
excitement that leads to temporary—or permanent, as in the present case—hearing problems. Apparently, damage depends on various factors: the energy given off or the amount of electricity that goes through tissues, type of current, tension, resistance, time, frequency, and waveform [1]. Alterations in ABR may be explained by cochlear changes, disturbances in the cochlear microphonic, damage to nerve structures in the auditory tract caused by the energy flux through the internal ear and auditory tract, and vascular disturbances with probable hemorrhages that caused cochlear damage [4].

**CONCLUSION**

Our results show permanent hearing alterations including progressive neurosensorial hearing loss, disturbed neuroconduction of the auditory nerve following HVED. The most relevant aspect in this type of accident is prevention. But when such accidents do occur, it is vital to attain ensure survival. Nevertheless, given the implications in rehabilitation and the social and legal consequences patients should be fully evaluated, including a complete hearing study.

The results of the present study pave the way for further research in larger groups of workers who suffered HVED; therefore future research should elucidate the mechanism and physiopathology of hearing damage and its relationship with the nature, gravity, type of current (DC or AC), voltage, resistance, path, and contact mechanism (direct, arch or ignition) [1].

**Guarantor**

The corresponding author is the guarantor of submission.

**Conflict of Interest**

Authors declare no conflict of interest.

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**REFERENCES**


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Table 1: Comparison of PEATC latencies of the case with reference values in milliseconds (ms)

<table>
<thead>
<tr>
<th></th>
<th>Wave I</th>
<th>Wave III</th>
<th>Wave V</th>
<th>Interwave I – III</th>
<th>Interwave III – V</th>
<th>Interwave I – V</th>
</tr>
</thead>
<tbody>
<tr>
<td>Reference value</td>
<td>1.65</td>
<td>3.79</td>
<td>5.82</td>
<td>2.14</td>
<td>2.02</td>
<td>4.16</td>
</tr>
<tr>
<td>Right ear</td>
<td>1.74</td>
<td>4.24</td>
<td>6.42</td>
<td>2.50</td>
<td>2.18</td>
<td>4.68</td>
</tr>
<tr>
<td>Left ear</td>
<td>1.78</td>
<td>4.28</td>
<td>6.56</td>
<td>2.50</td>
<td>2.28</td>
<td>4.78</td>
</tr>
<tr>
<td>Differential Right ear</td>
<td>0.09</td>
<td>0.45</td>
<td>0.6</td>
<td>0.36</td>
<td>0.16</td>
<td>0.52</td>
</tr>
<tr>
<td>Differential Left ear</td>
<td>0.13</td>
<td>0.49</td>
<td>0.74</td>
<td>0.36</td>
<td>0.26</td>
<td>0.62</td>
</tr>
</tbody>
</table>

Abbreviations: ms- milliseconds

Yvette De la Fuente-Rivera – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published

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**Author Contributions**

Guadalupe Aguilar-Madrid – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Arturo Torres-Valenzuela – Acquisition of data, Revising it critically for important intellectual content, Final approval of the version to be published

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Unsuspected wooden foreign body of lung parenchyma masquerading pulmonary tuberculosis: A rare surgical entity

Mohammad Sadik Akhtar, Mohammad Hanif Beg, Manoj Khurana

ABSTRACT

Introduction: Parenchymal foreign bodies after chest trauma are rare. These foreign bodies have no specific sign and symptoms and X-rays are unhelpful. The detection of wood is especially important because it may serve as a nidus for infection and masquerade pulmonary tuberculosis. Case Report: We report a rare case of retained wooden foreign body in the lung parenchyma that was suspected on computed tomography after prolonged treatment and repeated X-rays. Thoracotomy was performed and two wooden pieces were removed. Review of literature shows that presence of wooden foreign bodies in the lung parenchyma is quite rare and may present with a wide variety of symptoms. Conclusion: We conclude that foreign body should be considered in the differential diagnosis when patient presents with a history of trauma and patient fails to improve despite continued treatment, and it must be removed on an urgent basis due to the risk of recurrent infection.

Keywords: Foreign body, Lung, Wood, Parenchyma, Tuberculosis, Pulmonary disease

INTRODUCTION

Pulmonary parenchymal foreign bodies are a rare cause of chronic lung disease and infrequently considered in a differential diagnosis of pulmonary opacities on the chest radiographs [1]. Foreign bodies can penetrate soft tissues through open wounds and lacerations during trauma or by direct impact [2]. Such wounds harboring foreign bodies may appear to be deceptively minor and may not be accompanied by any major symptoms. However, if these foreign bodies are left undetected in the tissues they can result in serious sequelae like abscess, fistula formation [3] and hemoptysis [4, 5, 6], days, months or even years after the initial trauma. Although wooden foreign body is very common in soft tissue and orbital traumas [7], pulmonary parenchyma foreign bodies are a rare cause of pulmonary disease and are a rare differential diagnosis of lung opacity on the chest radiographs [8]. Only a limited number of case reports about retained pulmonary foreign body have been published in medical journals so far [9, 10]. The purpose of reporting this unusual case of recurrent pneumonia and hemoptysis caused by a retained wooden foreign body is to highlight the difficulties in detection of foreign bodies and need for their prompt removal as they are always a cause for recurrent chest infection. We report a rare case of a pulmonary parenchymal wooden foreign body diagnosed five years after the chest trauma. In our
opinion, wooden foreign body must be kept in
differential diagnosis of chest trauma when patient fails
to improve after prolonged treatment and removed as
soon as possible.

CASE REPORT

A 12-year-old boy (from a rural area) was presented
to our hospital with shortness of breath, cough, purulent
sputum and hemoptysis. He had a history of recurrent
respiratory tract infections that were resolved with
antibiotics. He had a history of chest trauma, after
falling from a tree five years back (Figure 1). His
treatment was done in a private hospital at the time of
trauma and chest drainage tube was placed and
improved. But patient had episodes of recurrent fever,
purulent sputum and later on hemoptysis. Repeated
chest X-rays showed opacity in right lower lung fields
(Figure 2). He was given multiple courses of antibiotics
but failed to improve. He was then started on ATT
suspecting pulmonary tuberculosis. But he had
recurrent infections and hemoptysis despite anti
tubercular treatment. Patient was referred to
cardiothoracic unit of our institute for further
management. He was investigated and repeat chest
X-rays revealed an area of hyperdensity in right lower
lobe lung fields. CECT Thorax was done which was
suggestive of consolidation with multiple cavities
formation in right lower lobe with right sided pleural
collection with single linear hyper-dense focus with CT
value of 190 HU(bony fragment/foreign body) with
adjacent pleural thickening (Figure 3). A thoracotomy
with removal of two wooden foreign bodies was done.
Peroperatively two wooden foreign bodies were
removed—one from anterior basal and another from
posterior basal segment of right lower lobe, each
measuring 4x2 cm (Figures 4–6). The lung parenchyma
was having two pieces of wood, which were enveloped in
granulation tissue and fibrosis although CECT reported
only a single foreign body. Postoperatively patient
improved uneventfully and discharged. He was doing
well till the time of last follow up.

DISCUSSION

Although aspiration of a wooden foreign body into
the tracheobronchial tree is not uncommon, [11]
pulmonary parenchymal wooden foreign bodies are
quite rare. Most parenchymal wooden foreign bodies are a result of trauma, and the diagnosis is made on the basis of the history and physical examination at the time of presentation [2]. Pulmonary wooden parenchymal foreign bodies are a rare cause of chronic lung disease and infrequently are considered in a differential diagnosis of pulmonary opacities on chest radiographs [1]. Despite advances in imaging techniques, the detection of retained wooden foreign bodies remains a difficult and challenging task [12]. The detection of wood is especially important because it may serve as an unrecognized nidus for infection [8]. Wood, with its porous consistency and organic nature, is an excellent medium for microorganisms. The retained foreign bodies may result in abscess, fistula formation [3] and hemoptysis [4, 5, 6]. Our patient was unaware of his pulmonary wooden foreign body and the treating physicians also failed to keep a possibility of a foreign body in the chest even when the patient did not improve after prolonged treatment including anti-tubercular therapy until the CECT was done five years after the trauma. The injury most likely occurred while the patient fell from a tree on the ground and a wooden fragment impaled her chest. In patients with recurrent uni-focal pneumonia, an underlying problem such as a foreign body should be considered [8]. Wooden fragments account for the largest proportion of retained foreign bodies after trauma to the human body [13]. Radiographs have been reported to revealed wooden foreign body in only 15% of patients [13]. CECT scan showed that the abnormality had consistency which could be a single piece of bone or foreign body. CT scan has been proved to be useful in the evaluation of suspected wooden matter. Brewer and Leonard [14] stated that CT scans are the most sensitive tool available for the detection of wooden foreign bodies in lacerations on puncture wounds. The attenuation of a retained wooden foreign body varies in relation to the content of air and fluid in the interstices of the wood. Within approximately one week, the wood absorbs blood products and exudates and increases its attenuation [15]. Dry wood, with high air content, has been reported to mimic a gas collection [7]. Bodne et al. [16] cited three cases of wooden foreign bodies with various attenuation values, ranging from close to air in acute cases to high (near to calcium) in chronic cases. In our case, the attenuation value of the wood particle was 190 HU, which is a high density near to calcification.

CONCLUSION

In our opinion, pulmonary wooden foreign bodies must not be treated conservatively and operated as soon...
as possible because they can serve as a nidus for recurrent infection. Also one should look for multiple foreign bodies although CT scan may report just single foreign body.

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Author Contributions
Mohammad Sadik Akhtar – Substantial contributions to conception and design, Acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published
Mohammad Hanif Beg – Substantial contributions to conception and design, Analysis and interpretation of data, Drafting the article, Final approval of the version to be published
Manoj Khurana – Substantial contributions to conception and design, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

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REFERENCES
Free floating right atrial thrombus leading to occlusion of the tricuspid valve in a patient with cirrhosis

Turgut Karabag, Sait Mesut Dogan, Muhammet Rasit Sayin, Cem Çıl, Mustafa Aydin

ABSTRACT

Introduction: Right atrial thrombi are rare conditions in structurally normal hearts, except for special conditions such as hypercoagulable states, malignant tumors and some systemic diseases. Case Report: We present a case of giant free-floating atrial thrombus extending from the inferior vena cava into the right atrium and leading to occlusion of the tricuspid valve, in a 55-year-old female patient being followed up for cirrhosis. Conclusion: Free-floating right heart thrombus is an extremely rare and dangerous phenomenon which can result in highly dangerous complications. Transthoracic echocardiography, being easily accessible and practical, is a valuable tool for rapid diagnosis. Free-floating thrombi, is an extremely dangerous phenomenon that requires immediate diagnosis and treatment. An immediate decision should be made in these patients for whom treatment options include thrombolytic therapy, medical treatment and surgical intervention in appropriate patients.

Keywords: Free-floating thrombus, Tricuspid valve occlusion, Cirrhosis, Hypercoagulable state


INTRODUCTION

Free-floating right heart thrombi are a rare phenomenon leading to severe complications such as occlusion of the tricuspid valve and severe pulmonary embolism [1]. We hereby present a case of giant free-floating atrial thrombus leading to occlusion of the tricuspid valve, which is considered to have migrated from the portal vein to the right atrium in a 55-year-old female patient being followed up for cirrhosis secondary to hepatitis C and discuss the treatment options.

CASE REPORT

A 55-year-old female patient was admitted to our hospital with complaints of swelling of the abdomen and legs. It was learnt that the patient had been followed up for cirrhosis secondary to hepatitis C for ten years and had received ribavirin treatment until last year. She was on amiodipine for hypertension and ursodeoxycholic acid. She had no history of drug abuse. Physical examination revealed blood pressure of 120/80 mmHg, pulse of 88/beat per minute and body temperature of 36.3°C. Cardiovascular examination revealed diastolic murmur in the tricuspid focus. Bilateral leg edema was also noted and there were findings consistent with ascites in the abdomen. Other system examinations were normal. Electrocardiography and teleradiography were also normal. Two dimensional echocardiography
revealed a 62×35 mm mobile thrombus extending from the inferior vena cava into the right atrium (Figure 1). Thrombus was moving to and fro into the right ventricle with each cycle (Figure 2). Thrombus was found to have caused occlusion of the tricuspid valve, with a maximum gradient of 10 mmHg and a mean gradient of 5 mmHg by continuous wave Doppler (Figure 3). The patient had a normal ejection fraction with dilated left atrium and mild mitral regurgitation. An abdominal ultrasound showed ascites in the abdomen, enlargement of the hepatic vein and portal vein and a 29 mm-long thrombus in the portal vein. The thrombus in the right atrium was considered to have originated from the portal vein. Levels of protein C, protein S and antithrombin III were 73%, 54% and 97%, respectively. Protein C and antithrombin III levels were near, protein S was under to the lower limit. Levels of D-dimer and ANA were normal. Deep venous thrombus was not detected in bilateral lower-extremity Doppler. The patient was advised for surgery, with a joint decision by gastroenterology and cardiovascular surgery. The patient refused surgical intervention and was placed under follow-up with heparin infusion followed by warfarin administration. After six months, no complication was occured and the thrombus was completely resolved.

DISCUSSION

Free-floating right heart thrombus is an extremely rare phenomenon which can result in highly dangerous complications. The condition manifests mainly through its symptoms, which result from the occlusion of the tricuspid valve or from a pulmonary embolism [1]. The mortality rate is approximately 40% in cases of right heart thrombi with the potential to cause severe pulmonary embolism [2]. Thus, transthoracic echocardiography, being easily accessible and practical, is a valuable tool for rapid diagnosis. Right atrial thrombi are rare in structurally normal hearts, except for catheter-related thrombi. However, it can occur in association with hypercoagulable states, malignant tumors [3] and some systemic diseases [1]. Right atrial thrombus can also be seen in low output states, cardiomypathies and cardiac arrhythmias [4]. Although its mechanism in cirrhosis has not been fully elucidated, a hypercoagulable state can occur due to various local and systemic mechanisms. It often leads to a tendency for thrombosis due to decreased portal venous flow and decreased natural anticoagulants such as, protein C, S and antithrombin III [5] which were detected near or under the normal limits in our patient. Besides, one study reported that up to 70% of all patients with portal vein thrombosis and cirrhosis have an underlying inherited hypercoagulable state (such as Factor V Leiden mutation and prothrombin 20210 gene mutation) [6]. In our case, thrombus in the right atrium was considered to have migrated from the portal vein thrombosis through inferior vena cava into the right atrium. Our patient had no systemic disease or
malignancy other than cirrhosis. The patient was in sinus rhythm and had no structural heart disease such as cardiomyopathy.

Right atrial thrombus, detected by two-dimensional echocardiography, may take different configurations during the cardiac cycle, reflecting, as seen in our case, the coiling and uncoiling of the elongated clot as it moves back and forth through the tricuspid valve, thus even leading to occlusion of the tricuspid valve [7]. Even though the treatment of right heart thrombi remains controversial, recommended treatment options include surgery, thrombolytic therapy and medical follow-up. Surgical intervention is recommended as soon as the diagnosis is established in patients with thrombus in the right atrial cavity [8], whereas thrombolytic therapy is another treatment of choice. The possibility of a pulmonary embolism caused by a fragmented thrombus due to thrombolytic therapy can cause unfavorable complications [9]. In addition, anticoagulation with heparin followed by warfarin administration is another treatment option in patients at high risk for surgery [10]. Surgery was recommended for this patient due to the size of the thrombus and high risk for pulmonary embolism. However, the patient refused surgical intervention and was placed under follow up with heparin infusion followed by warfarin administration.

CONCLUSION

Right heart thrombi, particularly free-floating thrombi, is an extremely dangerous phenomenon that requires immediate diagnosis and treatment. Transthoracic echocardiography may prove useful in emergency diagnosis. An immediate decision should be made in these patients for whom treatment options include thrombolytic therapy, medical treatment and surgical intervention in appropriate patients.

Author Contributions

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REFERENCES

Gallstone ileus
Sherif Monib, Ahmed Farghaly, Andrew Ritchie, Mustafa Halawa

ABSTRACT
Introduction: Gallstone ileus is an uncommon complication of cholelithiasis but an established cause of mechanical bowel obstruction in the elderly. Perforation of the small intestine proximal to the obstructing gallstone is rare, and only a handful of cases have been reported.
Case Report: We report the case of a 74-year-old man who presented with a clinical picture of small bowel obstruction secondary to a gall stone impacted in the jejunum. Conclusion: The diagnosis of gallstone ileus should be always kept in mind when dealing with any elderly patient presenting with abdominal pain, distension and a previous history of gall bladder stones.

Keywords: Gallstone, Ileus, Bowel obstruction

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INTRODUCTION
Gallstone ileus is an uncommon condition that may result when a gallbladder stone enters into the intestinal tract. The pathogenesis in the majority of cases is a cholecystoduodenal fistula, but rarely, cholecystogastric and cholecystocolonic fistulas can form and produce a gallstone ileus. Gallstone ileus is a rare cause of intestinal obstruction, accounting for only 1-4% of all cases [1]. Whilst only 0.3-0.5% of patients with cholelithiasis will go on to develop gallstone ileus, the mortality ranges from 12-18% [2, 3]. Mortality increases in those with multiple co-morbidities and the elderly.

CASE REPORT
This is a case of a 74-year-old male seen in the Accident and Emergency department with a two day history of vomiting, dehydration, abdominal distension and pain. Following an early assessment by the on call medical team, a surgical opinion was sought as his clinical picture was consistent with an intestinal obstruction. His medical history included type II diabetes mellitus but there was no history of previous abdominal surgery. Two months prior to admission, he attended the same accident and emergency department with abdominal pain. On that occasion he underwent a computed tomography (CT) scan and was diagnosed with acute calculic cholecystitis (Figures 1 and 2). On this occasion, he was treated conservatively with plans to carry out an elective laparoscopic cholecystectomy three months later.

On physical examination, the patient appeared generally well. He was afebrile and his observations were all stable. On abdominal examination he had marked abdominal distension (mainly central) with tenderness in the right upper quadrant and epigastric
region. There was no rebound tenderness and Murphy’s sign was negative. Digital rectal examination revealed an empty rectum.

Initial laboratory investigations revealed a mild hyponatraemia and hyperglycaemia. Liver function tests were not deranged and amylase was normal: Sodium 128 mmol/L and Potassium 3.8 mmol/L, Hb 16.2g/dL, white blood cells (WBCs) 7.0x10^9 /L, Platelets 141x10^9/L, PT 10.4 s, APTT 23.9 s, Amylase 30 U/L, T. Bil 20 mg/dL, ALT 30 U/L, ALP 94 U/L, Total Calcium 2.27 mmol/L, Alb 42 g/L, random blood sugar 15.2 mmol/L.

Plain abdominal radiograph of the patient demonstrated distended loops of small bowel (Figure 3). Abdominopelvic CT scan showed that a very large gallstone had migrated and impacted in the jejunum with proximally dilated and distally collapsed jejunal loops (Figures 4 and 5).

Based on the history, physical examination and CT scan findings, our nominal diagnosis was of gallstone ileus. After fluid resuscitation the patient underwent an exploratory laparotomy, approximately six hours after confirming mechanical small bowel obstruction. At laparotomy, there was a transition zone at the point of impaction, however, the bowel segment appeared healthy with no perforation. The gallbladder was adherent to the duodenum and no attempt was made to separate the two. A longitudinal enterotomy followed by successful extraction of the stone and primary transverse closure of the enterotomy was carried out without complications. Postoperatively, the patient was transferred to a general surgical ward where he recovered well. On the second postoperative day, the patient was able to tolerate fluids and a light diet. He mobilized well and was discharged home the following day.

After one month, the patient was seen in the outpatient department. He had continued to recover well and an elective cholecystectomy was arranged for three months later.
As with the pattern of incidence in cholelithiasis, gallstone ileus occurs three to five times more frequently in women than in men [2].

The gallstone enters the intestinal tract through a fistula formed between the gallbladder and the duodenum, stomach or colon. The terminal ileum is the most frequent site of obstruction [1]. However, as with our patient, there are other obstruction points, including jejunum (30%), colon (2.5%) and duodenum which is eponymously known as Bouveret syndrome [2].

Gallstone ileus remains a diagnostic challenge despite advances in imaging techniques, and pre-operative diagnosis is often delayed. Plain abdominal radiographs may reveal signs of small bowel obstruction and concomitant aerobilia to suggest the diagnosis [7, 8]. Whilst the classic radiological triad, or Rigler triad, of pneumobilia, small bowel obstruction and ectopic gallstone is specific for the disease, it is unfortunately only present in 9–14% of cases [7]. Therefore, the current definitive investigation of choice is CT scan.

The principal goal in management of gallstone ileus is a quick effective relief of mechanical bowel obstruction. When appropriate, it is possible to avoid surgery, for example, if the stone is in within reach of an endoscope, either in the proximal small bowel or in the colon, there is the option to treat by lithotripsy and removal of the fragments [2]. Spontaneous passage of gallstones large enough to cause impaction has been reported, but most patients require surgical intervention and the prominent risk of perforation underlines why a conservative approach is rarely adopted. Extracorporeal shockwave lithotripsy can be successful, but this method is limited by the presence of bowel gas.

The majority of patients require surgery either for bowel obstruction or perforation. Surgical options include enterotomy and removal of the stones (enterolithotomy) with subsequent elective cholecystectomy or enterolithotomy plus cholecystectomy and repair of the fistula in the same sitting [3].

Whilst the most appropriate surgical intervention remains unclear, authors mostly favor enterolithotomy to relieve the ileus, followed by subsequent elective Cholecystectomy. Reports suggest this method of management causes fewer complication and there is a 50% chance of spontaneous fistula closure [7]. In addition, enterolithotomy alone minimises operating time in the emergency setting and is technically less demanding [9]. This is especially advantageous in the high risk surgical candidates. For the frail patient, enterolithotomy alone is often adequate treatment and subsequent Cholecystectomy is not mandatory [10].

Zuegal et al. (1997) propose that the one stage procedure significantly decreases morbidity and mortality because removing the gallbladder and biliary-enteric fistula prevents future recurrence of gallstone ileus and recurrent biliary symptoms [11]. It also avoids the need for a second operation.

In cases of perforation secondary to pressure necrosis of impaction or a perforated jejunal diverticulum, due to increased intra-luminal pressure

DISCUSSION

Gallstone ileus is an uncommon cause of small bowel obstruction that occurs almost exclusively in the elderly [1–6]. In the population over 65-years-old, gallstone ileus is thought to account for up to 25% of cases of mechanical small-bowel obstruction. In this subsection of the general public it carries a mortality of 12-50% [4].
proximal of the obstructing gallstone, the only way of management is segmental bowel resection and primary anastomosis [12].

CONCLUSION

Although it is a rare cause of bowel obstruction, gallstone ileus should be kept in mind when dealing with a case of small bowel obstruction, especially in elderly patient as early surgical intervention is the mainstay of treatment.

SUMMARY

Gallstone ileus poses a management challenge as the pre-operative diagnosis is often delayed, despite advances in imaging techniques. Although intestinal perforation in gallstone ileus is extremely rare, the condition still results in both high morbidity and mortality rates, which are partly due to the fact that the condition affects mainly the elderly population.

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Sherif Monib – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published
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REFERENCES

Double axillary vein: A case detected by CT

Takaaki Fujii, Reina Yajima, Satoru Yamaguchi, Hiroyuki Kuwano

CASE REPORT

A 50-year-old Japanese woman was presented with right breast cancer with clinically negative axillary nodes. A preoperative contrast-enhanced computed tomography scan showed that the contrast medium admitted flowing in lower part of the right axillary vein, suggesting an anomaly of axillary vein known as double axillary vein (Figure 1). Mastectomy and sentinel lymph node (SLN) biopsy were performed. The intraoperative diagnosis of the axillary SLN was positive for metastasis, and thus additional conventional axillary lymph node dissection was performed. At axillary dissection, a double axillary vein was detected (Figure 2). In this case, the thoracodorsal vein originated from the lower vein (Figure 2, arrowhead). In our case, none of the non-SLN was metastatic at final histology.

DISCUSSION

The axillary vein is the continuation of the basilic vein at the lower border of the teres major muscle. It continues as the subclavian vein at the outer border of the first rib. A few reports describe the variations of axillary veins [1–3], however, variations in axillary

Figure 1: A preoperative contrast-enhanced CT showed that the contrast medium admitted flowing in lower part of the right axillary vein, suggesting an anomaly of axillary vein such as a double axillary vein (arrow).

Figure 2: At axillary dissection, a double axillary vein was detected (arrow). The thoracodorsal vein originated from the lower vein (arrowhead).
anatomy have been rarely described in most text of operative surgery or anatomy [1]. The knowledge of anatomical variations of the axilla is necessary for the axillary dissection. Kutiyana wala et al., reported the anatomical variants during axillary dissection, including double axillary vein [1]. In 10% of patients with breast cancer cases, double axillary vein was observed during axillary dissection. Double axillary vein itself is not rare, however, double axillary vein is diagnosed during the axillary dissection. If the variation of axillary vein is detected preoperatively, the axillary dissection may be performed more safely. This is the first report that double axillary vein is preoperatively diagnosed by computed tomography (CT). A preoperative contrast-enhanced CT is thought to be useful for the diagnosis of double axillary vein.

CONCLUSION

The knowledge of axillary vein variations is important in axillary surgery in case of breast cancer or brachial plexus anesthesia. With the use of enhanced CT scan, the risk of damage to the axillary vein during axillary dissection can be reduced in cases with double axillary vein.

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REFERENCES


Polycystic kidneys occupying entire abdominal cavity

Vinay S Gundlapalli, Carlo Ramirez

CASE REPORT

A 54-year-old female patient, pharmacist by profession, met her family doctor for annual health check-up. She was in her usual state of health. She is a known hypertensive, well controlled with anti-hypertensives. Patient had no other past medical or surgical history. Her family history was significant for polycystic kidney disease. Patient is a non-smoker denied alcohol consumption and drug abuse. The physical examination was unremarkable. Routine blood tests demonstrated elevated serum creatinine level of 2 mg/dL (normal 0.7–1.4 mg/dL). An ultrasound examination was performed which showed bilateral very large cystic kidneys consistent with polycystic kidneys. This was followed by a CT scan of the abdomen which showed that both kidneys are replaced by numerous cysts typical for polycystic kidney disease. The right kidney measures 22x17.6x24 cm, and left kidney measures 17x29x24 cm. The entire bowel was displaced anteriorly by the large kidneys. There was a 1 cm cyst seen in the right lobe of liver. The CT scan confirmed the diagnosis of very large polycystic kidneys occupying almost the entire abdomen and pelvis (Figures 1 and 2). Over the next one year the patient’s renal function deteriorated to GFR of 6, serum creatinine level 7 mg/dL (normal 0.7–1.4 mg/dL), BUN 42 mg/dL (normal 7–26 mg/dL), serum potassium level 6.2 mmol/L (normal 3.5–5 mmol/L). Hemodialysis was initiated through a dialysis catheter then via a dialysis access AV fistula. She also started having early satiety. She underwent bilateral nephrectomy making room for the renal transplant and also to relieve the compressive gastrointestinal symptoms. Currently, she is on renal transplantation list and getting hemodialysis three times a week.

DISCUSSION

Autosomal dominant polycystic kidney disease (ADPKD) is a hereditary disorder characterized by multiple renal cysts and various systemic manifestations [1]. Massively enlarged kidneys with innumerable cysts characterize it. Grossly the kidneys are enlarged; the cysts are well defined round or oval with thin imperceptible or calcified wall. Cysts are filled with clear, serous, turbid, or hemorrhagic fluid. Microscopically; the cysts are lined by simple flattened or cuboidal epithelium with or without calcification [2]. ADPKD or adult PKD is one of the most common inherited disorders [1]. Abnormality in genes located on chromosome 16 and 4 leads to proliferation of renal tubular cells leading to formation of diverticulae of nephrons (collecting ducts) ultimately causing cystogenesis [1, 2]. It is a multisystemic disorder. The cystic manifestations of ADPKD are seen in kidneys (100%), liver (75%), pancreas (10%), spleen, ovaries, testis, and seminal vesicals [3]. Noncystic manifestations of ADPKD are cardiac valvular disorders (26%), hernias (25%), colonic diverticulae, cerebral “berry” aneurysms (5–10%) aorta and coronary arteries aneurysms [3].

It can present in any age group. It has equal preponderance in males and females. The incidence in white population is 1 in 400 to 1 in 1000. Patients can be asymptomatic or present with flank pain, hematuria, hypertension or renal failure. The disease process can be complicated with hemorrhage, infection, rupture, renal calculi and renal failure [1].

Ultrasound shows multiple, well-defined, round, anechoic areas in both enlarged kidneys. Ultrasound has
The treatment is usually symptomatic and managing complications like hypertension, pain, and renal infections. Once patients develop end stage renal failure dialysis or renal transplantation is necessary. The prognosis is fair, following renal transplantation like any other chronic renal failure [1]. The indications for pre transplant nephrectomy in ADPKD are for making space for the renal transplant, uncontrollable symptoms like pain, massive hematuria, recurrent infections and gastrointestinal compressive symptoms [5].

CONCLUSION

ADPKD causing massively enlarged kidneys occupying the entire abdominal cavity compressing adjacent visceral organs is rare. We should be aware that bilateral nephrectomy is indicated if the patient is symptomatic. The long-term prognosis for such patients is still good with renal replacement therapy, initially with dialysis and ultimately renal transplantation.

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REFERENCES

Iodide mumps

Ping Chen

CASE REPORT

A 74-year-old normally fit and well gentleman was presented with hemoptysis and altered mental status, and underwent a chest and brain CT scan with contrast. The patient received total of 75 mL of Ultravist 370 during the procedure. After a few hours undergoing CT scan, the patient complained of swelling and pain in the upper neck, and slight shortness of breath. He was apyrexial, respiratory rate 15/min, regular heart rate of 76 bpm and blood pressure 146/67 mmHg. Bilateral, diffuse submandibular enlarged glands were noted (Figure 1). They were tender to touch. His thyroid function test was normal, thyroid peroxidase antibody was negative. Contrast induced acute sialadenitis (iodide mumps) was suspected on the basis of clinical presentation. The patient was treated with simple analgesics. The submandibular gland swelling was completely resolved after 24 hours.

DISCUSSION

Acute sialadenitis features swelling of saliva glands after exposure to contrast media. It is a very rare adverse reaction. Onset time can be from minutes to five days. The condition is usually self-limiting. Other associated adverse reactions include facial nerve paralysis, enlarged thyroid/lacrimal glands [1]. There are no life threatening cases reported. In this case, the patient was given Ultravist 370, which is iodinated contrast containing mainly organically-bound iodine, and a tiny amount of inorganic iodide. 1 mL Ultravist contains 769 mg of iopromide, which is equivalent to 370 mg iodine. The exact causes of iodide mumps are unclear. They are thought to be idiosyncratic. Possible toxic accumulation in the ductal system and high concentration of non-organic iodine induce inflammation changes leading to obstruction [2].

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Figure 1: Diffuse, bilateral, symmetrical submandibular gland enlargement four hours after CT scan.
CONCLUSION

Contrast media are frequently used, estimated at 70 million administrations worldwide per year [3]. Often, large volumes are required, so reactions are an important problem. Exposure to iodinated contrast media may elicit a variety of adverse reactions. Iodide mumps or acute sialadenitis after contrast media imaging is a rare adverse effect to iodine.

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REFERENCES

Varied presentations of epithelial cysts

Ashok Kumar Bolbandi, Pramod Krishnappa, Arunchandra Bennehalli

CASE REPORT

**Case 1:** A 55-year-old male patient presented with a swelling over the scalp since three years gradually increased in size (Figure 1). On examination, a nontender fluctuant swelling of size 7x6 cm was found over the scalp with smooth surface and well defined borders. Plain radiograph of the skull was normal. Elliptical skin incision was made and the entire cyst and its wall was excised. Histopathological diagnosis was epidermal inclusion cyst (Figure 2).

**Case 2:** A 60-year-old male patient presented to us with a swelling protruding from the umbilicus since four years gradually increasing in length. On examination, 8x1 cm swelling was found arising from the inferior aspect of umbilicus and lying over the skin surface (Figure 3). A diagnosis of sebaceous horn was made and excision was done. Microscopically the horn consisted of a mixture of squamous epithelial cells and keratinized debris.

**Case 3:** A 47-year-old male patient presented with multiple painless swellings over the anterior, lateral and posterior wall of the scrotum gradually increasing in number and size since 20 years (Figure 4). There was no lymphadenopathy. Serum and urinary levels of calcium and phosphates were normal. Subtotal excision of the scrotal wall containing the nodules was done, and primary closure of the scrotum was done which could accommodate both the testes without tension. Histopathological examination revealed idiopathic scrotal calcinosis (Figure 5).

![Epithelial cyst over the scalp.](image1)

![Epidermal inclusion cyst: Cyst wall identical to the epidermis and filled with keratin.](image2)
DISCUSSION

Epithelial cysts are common lesions formed by the invagination and cystic expansion of the epidermis or of the epithelium forming the hair follicle. The epidermal inclusion cyst, pilar cyst (trichilemmal cyst), dermoid cyst and steatocystoma are the different types of epithelial cysts. Each has a wide range of presentation varying from a size of a pea to a very large mass. We have described a few rare presentations of epithelial cysts.

Epithelial cysts are divided into several types according to the structural components of their walls. They are epidermal inclusion cyst, pilar or trichilemmal cyst, dermoid cyst and steatocystoma.

The epidermal inclusion cyst has a wall nearly identical to the epidermis and is filled with laminated strands of keratin. Pilar cysts have a wall that resembles follicular epithelium, without a granular cell layer and filled by a more homogenous mixture of keratin and lipid.

The dermoid cyst is similar to the epidermal inclusion cyst, but it also shows multiple appendages (such as small hair follicles) budding outward from its wall. Finally, steatocystoma are the true sebaceous cysts with a wall resembling the sebaceous gland duct, if multiple, called steatocystoma multiplex [1].

Complications of longstanding epithelial cysts are infection, abscess formation, cutaneous horn, cock’s peculiar tumour and squamous cell carcinoma.

Cock’s peculiar tumour was described by Cock in 1852. When the sebaceous cyst of the scalp ulcerates, excessive granulation tissue forms resembling fungating epithelioma. This is called the Cock’s peculiar tumour [2].

Total excision of the cyst is the treatment of choice. Different techniques have been proposed for excision of a epithelial cyst: (i) Conventional elliptical incision and dissection technique, (ii) Squeeze eversion technique, and (iii) Minimal excision technique/Punch biopsy technique.

The conventional elliptical incision and dissection technique includes making an elliptical incision on the skin. The cyst is gradually dissected using artery forceps from the surrounding skin till the entire cyst can be removed intact. It must be remembered that the whole of the cyst wall must be removed, otherwise recurrence is common.

Squeeze eversion technique can be applied successfully to small cysts (<2 cm) without much fibrous attachment to the capsule. Make an elliptical incision, then dissection is carried out with curved scissors. The capsule should not be picked up with instruments. One should lift the skin edges rather than the capsule to facilitate dissection. Once the superficial aspect of the cyst is freed, then gently squeeze the cyst from the sides and base to evert the cyst. The main idea is to avoid handling the capsule [3].

The punch technique allows for extrusion of the cyst contents through a 3–4 mm skin and cyst opening created by a punch biopsy tool. The deflated cyst can
then be pulled through the skin aperture and excised; this technique carries a recurrence rate of approximately 6% [4]. The minimal excision technique utilizes a 2–3 mm incision to excise the cyst following extrusion of contents in a similar manner to the punch technique [5, 6].

The pathogenesis of scrotal calcinosis (SC) is still controversial. Some authors think that SC is the result of dystrophic calcification of pre-existing structures such as sebaceous cysts. It may also be due to the degeneration of the dartoic muscles. Some authors did not find any evidence of pre-existing cystic structures and think that this condition is idiopathic [7]. Scrotal calcinosis may require partial/total scrotectomy and placing the testes in the thigh pouch.

Cutaneous horn is a clinical diagnosis referring to a conical projection above the surface of the skin that resembles a miniature horn. Although often benign, they can also be malignant or premalignant [8]. The horn is composed of compacted keratin. Various histologic lesions have been documented at the base of the keratin mound, and histologic confirmation is often necessary to rule out malignant changes. Malignant lesions at the base of the horn usually are squamous cell carcinoma, although basal cell carcinoma has been rarely reported. Benign idiopathic causes are frequent and include seborrheic keratosis, epidermal nevus, trichilemmal cyst, trichilemmoma, prurigo nodule, and intradermal nevus.

CONCLUSION

Epithelial cysts have various presentations in different parts of the body. Other conditions simulating an epithelial cyst should be excluded before the surgery to avoid complications. Postoperatively, the diagnosis should be confirmed by histopathology to exclude malignancy.

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The authors declare no conflict of interest.

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Author Contributions

Ashok Kumar Bolbandi – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Pramod Krishnappa – Substantial contributions to conception and design, Acquisition of data, Drafting the article, Revising it critically for important intellectual content, final approval of the version to be published
Collision tumors of ovary: A rare phenomenon

Shaista Choudhary, Shankar Adisesha

To the Editors,

Collision tumors represent a coexistence of two adjacent but histologically distinct tumors without admixture in the same tissue or organ. Though such tumors have been reported often in various organs, their occurrence in ovary is rare.

We report here a rare case of collision tumor of ovary comprising serous cystadenoma and mature cystic teratoma. A 55-year-old woman was presented with palpable mass in the lower abdomen associated with pelvic pain and dysfunctional uterine bleeding. Patient underwent right salpingo-oophorectomy following ultrasound diagnosis of ovarian tumor. Grossly the tumor measured 6 cm in diameter. Cut surface revealed a trilocular cyst with two locules containing clear fluid. Inner wall of one of the locules exhibited few tiny polypoidal excrences (Figure 1). The other locules had a gelatinous appearance.

Microscopy revealed a combination of morphologic features. Sections from two locules showed the cyst wall lined by unlayer of low cuboidal epithelium which at places exhibited presence of cilia. A few papillary processes lined by ciliated/cuboidal epithelium were also noted. Sections from locule with gelatinous appearance revealed cyst wall lined by well differentiated stratified squamous epithelium (Figure 2).

Figure 1: Microphotograph showing a trilocular ovarian cyst with papillary excresences. H&E stain, 40x.

Figure 2: Microphotograph showing serous cystadenoma and teratomatous component. H&E stain 10x.

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beneath which was seen adipose tissue, sebaceous glands, sweat glands, hair follicles, ganglion cells (Figure 3) and a focus of colloid filled area. Based on the microscopy, a diagnosis of collision tumor of ovary was made.

![Microphotograph showing a gland lined by cuboidal epithelium with adjacent ganglion cells H&E stain 40x.](image)

Figure 3: Microphotograph showing a gland lined by cuboidal epithelium with adjacent ganglion cells H&E stain 40x.

We present this case because collision tumors in ovary are a rare entity and combination of serous cystadenoma with teratoma is rarer [1]. Though collision tumors have been reported earlier, combined serous cystadenoma with mature cystic teratoma is rarely reported. With the available literature, there is just one similar case reported in a non child bearing young woman [2]. There are instances of collision tumors consisting of teratoma with serous cystadenocarcinoma, mucinous cystadenocarcinoma and/or granulosa cell tumor [3]. In a study conducted at Seoul national University college of medicine, the authors reviewed seven pathologically proven cases of collision tumors of ovary associated with teratoma. Ovarian teratomas were co-existent with mucinous cystadenoma (4 cases), borderline mucinous tumor (1 case), mucinous cystadenocarcinoma (1 case) and dysgerminoma (1 case) [4]. There is a single case report of collision tumor composed of a colonic adenocarcinoma arising in a sigmoid diverticulosis coexisting with recurrent ovarian granulosa cell tumor [5]. Though mature cystic teratoma of ovary contains derivatives of all three embryonic germ cell layers, it rarely presents with ovarian epithelial or sex cord stromal tumors. Rare cases of ovarian cystic teratoma in association with surface epithelial tumors have been reported in literature and occurrence of serous cystadenomas with mature cystic teratoma is even rarer [3]. Collision tumors have been described in various organs including oesophagus, stomach, liver, bone, kidney, brain and lung. Such tumors involving ovary are rare.

In conclusion, we would like to emphasize upon the fact that multiloculated cysts have to be extensively examined, so as not to miss any component which might have a bearing on prognosis of the patient. Such cases need to be documented for academic as well as prognostic purpose.

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Shankar Adisesha – Analysis and interpretation of data, Critical revision of the article, Final approval of the version to be published

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The corresponding author is the guarantor of submission.

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A rare formation of renal calculi like a mushroom

Takashi Kawahara, Hiroki Ito, Hideyuki Terao, Yoshinobu Kubota, Junichi Matsuzaki

To the Editors,

A 65-year-old male patient was referred to our institute for the treatment of his left ureteral and renal calculi (Figure 1A–B). After two months, he was admitted to our department for treating his left ureteral and renal calculi with ureteroscopic lithotripsy using a holmium yttrium aluminum garnet (Ho: YAG) laser. After inserting ureteral access sheath, we firstly made stone fragment and retracted them in the ureter using rigid ureteroscope (URS) with Ho: YAG laser lithotripsy. Thereafter, we observed the renal collecting system using flexible URS. In the view of flexible URS findings, renal stones were formed like mushrooms (Figure 2). Because of large stone volumes, we made stone fragments and retracted them in the second session of URS one month after initial treatment.

In general, ureteral and renal stones were usually formed as a round ball, except for the large renal staghorn calculi. Our institute is a large referral stone disease center in Japan, more than 400 shockwave lithotripsy (SWL), 300 URS and 80 percutaneous nephrolithotomy (PCNL) were performed per year.

![Figure 1: (A) Kidney Ureter Bladder film and (B) CT scan, showed left renal stone, but showed no detailed stone formation.](image1)

![Figure 2: Renal stones were formed like a mushroom.](image2)

However, it is our first time to see these ridiculously formed renal stones like mushrooms. A total of nine stones were successfully removed and chemical composition showed calcium oxalate monohydrate.
It was the first report of strange formed renal stone. For gallbladder stone, stone formation was sometimes a clue for suspecting the chemical composition [1, 2]. In the previous report of ureteral stones, stone surface formation might be a clue to be easily broken or not for SWL in cystine stone [3]. However, the correlation between renal stone formation and chemical stone composition was not widely accepted. Further reports about stone formation will be needed.

We herein report the first case of unfamiliar formation of renal stone.

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