moderate facial swelling, minimal ankle edema, but no pleural effusions. Spirometry showed FEV1 of 1.7 and FVC of 2.7 L (BTPS). She had been a regular heavy smoker.

Bilateral conductive deafness led to a diagnosis of keratosis obturans of the external auditory meatus. This has been removed at regular intervals since with hearing improvement each time.

In 1981, when she was 66 years old, bilateral pleural effusions were noted. These were tapped but they reaccumulated rapidly. Cytologic study of the pleural fluid showed no malignant cells, culture was negative, and the protein content was 5.4 g/dL. Other investigations were normal except for a raised erythrocyte sedimentation rate (ESR) at 69 mm/h. Spirometry in the presence of the effusions gave an FEV1 of 1.02 L and FVC of 1.60 L (57 percent and 66 percent predicted, respectively). Because of persistent dyspnea due to recurrent pleural effusions, she underwent successful tetracycline pleurodesis. Her condition remains stable three years later with bilateral basal pleural thickening but no obvious effusions. Most recent spirometry in 1989 gave an FEV1 of 1.5 L and FVC of 2.5 L (predicted, 1.7 L and 2.4 L).

**DISCUSSION**

The yellow nail syndrome has now been well characterized and consists of yellow nails, lymphedema, and pleural effusions. Bronchiectasis and sinusitis are often associated.

The etiology is unknown, but lymphangiography has shown very variable lymphatic hypoplasia. Fluid kinetics of the pleural effusions suggest that the accumulation is due to ineffective drainage rather than excess production of pleural fluid. Pleural effusions often occur late in life and are sometimes preceded by infection.

Moran and Larkworthy speculated that keratosis obturans was a manifestation of the yellow nail syndrome in the external auditory meatus. However, in an early description of the association between bronchiectasis and keratosis obturans, a secretomotor reflex mediated through vagal channels in the auditory branch of the vagal nerve was postulated. An active cough reflex induced by stimulation of that nerve was cited as evidence; also, the bronchiectasis and the ear problems were usually ipsilateral. In patients without associated bronchiectasis, an underlying hyperemia and chronic inflammation have been observed in the external auditory meatus with keratosis obturans and consequent increased desquamination suggested as the mechanism for the keratin plug.

If keratosis obturans is a manifestation of the yellow nail syndrome, our patient represents the first report in which all the different features coexist. It is now 20 years since the diagnosis was made and she remains well, emphasizing the good prognosis in these patients.

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**Bilious Pleural Effusion following Liver Biopsy**

Richard J. Pisani, M.D.; and Frederick A. Zeller, M.D.

Pleural effusions in patients with chronic liver disease are common and usually are of little consequence. Bilious pleural effusion can occur following percutaneous biopsy or cholangiography procedures if the pleura is traversed. This report emphasizes the role of biliary tract obstruction in the development of a bilious effusion and the importance of biliary tract decompression in treatment. We discuss the laboratory evidence supporting the diagnosis of bilious effusion and review the reported experience with this complication. (Chest 1990; 98:1535-37)

Extravasation of bile into the thoracic cavity is an unusual cause of pleural effusion which tends to occur in a limited number of clinical settings. Bilious pleural effusion was initially described as a complication of echinococcal and amebic, pyogenic, and tuberculosis subphrenic abscesses. It is a rare complication of abdominal trauma which involves the liver, and can also occur in acute duodenal perforation in the presence of a congenital defect in one of the hemidiaphragms. The extensive use of percutaneous transhepatic biliary tract decompression in recent years has been associated with numerous case reports of bilious pleural effusion complicating this procedure.** Herein we report a case of bilious pleural effusion complicating orthotopic liver transplantation and subsequent liver needle biopsy and percutaneous transhepatic cholangiography.

**CASE REPORT**

A 59-year-old man with hepatic failure secondary to chronic active hepatitis underwent liver transplantation. Postoperative liver function tests remained elevated though there was no biopsy evidence of rejection nor was there any evidence of biliary tract obstruction. Nineteen months later, he was referred back to the Mayo Clinic for evaluation of jaundice. At the time of admission, his prednisone dosage was 20 mg twice a day. Examination revealed a thin, icteric man with mild peripheral edema, but who was otherwise normal. Laboratory evaluation revealed a total bilirubin of 8.8 mg/dl; direct, 6.6 mg/dl; AST, 312 U/L; ALT, 101 U/L; and alkaline phosphatase, 1,628 U/L. On the day of admission, ultrasound of the liver showed no evidence of intrahepatic or extrahepatic ductal ectasia and the portal vein and hepatic artery were patent by Doppler echocardiography. A transhepatic cholangiogram showed normal sized intrahepatic ducts, mild dilatation of the native common bile duct (which contained some debris), but no evidence of obstruction. A percutaneous liver biopsy under ultrasonic guidance suggested rejection, and a steroid pulse was given. Due to a rising bilirubin and suspicion of biliary obstruction, endoscopic retrograde cholangiopancreatography (ERCP) was performed on the 4th hospital day. A common duct stone was removed endoscopically. The next day the patient complained of mild shortness of breath, and the chest radiograph showed a moderate-sized right pleural effusion. Thoracentesis yielded 1,100 ml of fluid, and the patient's dyspnea resolved. A post-thoracentesis radiograph showed

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minimal pleural fluid. On the 9th hospital day, the patient had hypotension and respiratory insufficiency requiring pressors and mechanical ventilation. Blood, urine, and sputum cultures grew Candida albicans. Multiorgan failure ensued. Reaccumulation of the right pleural effusion prompted thoracentesis on two additional occasions (a summary of laboratory results appears in Table 1). Repeat ERCP was performed on the 18th hospital day, and another common duct stone was removed. The patient expired on the 24th hospital day. Autopsy limited to the liver demonstrated "shock liver," mild rejection, and a patent common bile duct without residual calculi. No evidence of a biliary pleural fistula was found, and no evidence of direct hepatic involvement with Candida was present.

**DISCUSSION**

Needles passed between the 9th and 10th intercostal space frequently transgress the pleural sulcus en route to intrahepatic bile ducts. Nonetheless, pleural complications are rarely associated with percutaneous transhepatic cholangiography, and in one large series of 2,006 procedures, only two patients developed pneumothoraces. Previous reports of bilious effusion complicating percutaneous transhepatic biliary cannulation pertain primarily to patients requiring prolonged biliary decompression.\(^{1,8}\) The risk of fistula increases with (1) inadvertent removal of drainage tubes; (2) prolonged drainage; (3) tubes placed intercostally between the 9th and 10th ribs in the midaxillary line; (4) persistent biliary tract obstruction; and (5) biliary tube dysfunction. The common feature in these cases is the presence of bile under increased pressure which can leak backward along a path created by the drainage tube. Decompression occurs in the pleural space and is facilitated by negative intrathoracic pressure.

Thoracic complications following percutaneous liver biopsy are unusual. Pneumothorax occurs in less than 1 percent of biopsies and most are small and asymptomatic. Right-sided pleural fluid collection also occurs in less than 1 percent of procedures by one of several mechanisms. Laceration of intrathoracic vasculature, presumably of the lung, can lead to hemothorax which may require drainage. Perforation of the diaphragm with attendant leakage of ascitic fluid into the chest probably accounts for the majority of pleural effusions developing after biopsy. Lastly, biliary pleural fistula can occur. However, there are only two cases reported in the world literature.\(^{6,9}\) Although biliary obstruction was thought to play a permissive role in one case;\(^6\) in the second case, no evidence of obstruction was present.\(^7\) Several risk factors for thoracic complication of liver biopsy have been reported and include transthoracic approach, larger needle size, multiple passes of the biopsy needle, and right lower lobe pulmonary disease. The latter has been noted as a relative contraindication for transthoracic percutaneous liver biopsy by several authors; however, it is based on anecdotal experience.\(^{6,9}\)

It is conceivable that small bilious effusions may commonly occur after percutaneous liver biopsy or PTC (even when drainage tubes are not left in place), but resolve rapidly if the biliary tree is not obstructed.

Bilious pleural effusion is diagnosed by finding a pleural/serum total bilirubin greater than one. In an animal model of bilious pleural effusion,\(^4\) bilirubin clearance was found to be rapid. It has been suggested that if thoracentesis is delayed, the pleural fluid biochemistry may be nondiagnostic. This may explain the failure to see an elevated pleural/serum bilirubin ratio on the second thoracentesis. Only with recurrent biliary leakage were the laboratory results confirmatory of a bilious effusion. In our patient, only one pass was required to perform the cholangiogram, and it was done with a small (22-gauge) needle through the 10th and 11th interspace. On the other hand, the liver biopsy was performed with a larger (15-gauge) needle, and therefore, may have been more likely to leave a significant track. Increased intrabiliary pressure, which in part may have been caused by contrast injections during ERCP, may have maintained tract patency. Treatment of bilious pleural effusion is aimed at securing adequate biliary decompression. The recurrence of a bilious effusion may suggest ongoing biliary tract obstruction. Such was likely the case in our patient. When associated with subphrenic abscess, biliopleural fistula develops as a consequence of local tissue necrosis. In some cases, empyema may spread to involve the lung parenchyma and bronchial tree resulting in bronchopleuralbiliary fistula and expectoration of bile. When bilious effusion is associated with empyema, subdiaphragmatic abscess, or a broncho-pleurobiliary fistula, surgery may be required.

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Permanent Venous Access Via Subcutaneous Infusion Port in Severe Asthma*

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A subcutaneous infusion port was implanted in a 34-year-old patient with frequent and severe asthma attacks to ensure prompt and reliable venous access. Difficulties with peripheral venous access were possible cofactors necessitating mechanical ventilation on two occasions before this implantation. The method described is simple and seems useful for asthmatics in need of frequent parenteral medication.

(SIP = subcutaneous infusion port)

The use of a subcutaneous infusion port for chronic venous access in cancer chemotherapy, parenteral nutrition, and antibiotic therapy in certain patients with recurrent respiratory tract infections has become well established during the last ten years.1-4 We report SIP used in a patient with unstable asthma where peripheral venous access was poor due to frequent infusions and obliterated veins.

CASE REPORT AND METHODS

A 34-year-old man suffered from unstable asthma with several acute severe attacks which required mechanical ventilation on two occasions (May 1986 and October 1987). He did not tolerate systemic steroid treatment because of gastrointestinal and psychologic side effects. On several occasions, he experienced psychotic symptoms even with minor doses of systemic prednisolone, and he refused further treatment with systemic steroids. He received local treatment with salbutamol and beclomethasone and systemic treatment with salbutamol and theophylline at optimal doses. The plasma-theophylline level varied greatly, probably due to poor compliance concerning tablet intake.

From January 1986 to October 1987, he had 13 admissions to the hospital because of severe asthma attacks. Due to frequent infusions, peripheral veins obliterated, and to ensure a quick and safe venous access in emergency, a subcutaneous infusion port was placed in October 1987 (Norport, Norfolk Medical Products, Skokie, IL). The SIP consisted of a conical chamber with a self-sealing rubber septum connected to a central venous catheter. All parts of this device were placed subcutaneously. For injection, a special needle (Huber point) was inserted through the skin into the chamber. When using a 22 G (0.7 mm) needle, the septum withstands at least 2,000 perforations (manufacturers unpublished data).

The implantation was done using local anesthesia, with a percutaneous technique for introduction of the central venous catheter. The right internal jugular vein was used, and the SIP placed in the right infraclavicular region.

The patient had follow-up visits as an outpatient once or twice a week. Theophylline in plasma was controlled in blood drawn from the SIP, and additional short-time infusions of 100 to 200 mg theophylline were given if the plasma-theophylline level so permitted. On several occasions, he also received infusions with beta-agonist drugs. The system was flushed with 10 ml of saline solution containing 0.5 IU heparin/ml milliliter prior to and at the completion of infusion, and filled with 5 ml heparinized saline solution (100 IU/ml) between infusions. This will eliminate the risk of coagulation in the catheter for at least one month.5

There has been no complication with the use of SIP. The patient tolerated the device well and has had no limitation to normal activities. The catheter has presently functioned for 730 days. The SIP allows frequent blood sampling, which is of great importance in the monitoring of plasma-theophylline at the follow-up visits.

DISCUSSION

The application of SIP in asthma is exceptional. In this particular patient, the intolerance to systemic steroid treatment and sudden, severe asthma attacks indicated the need for reliable venous access. The patient had two previous periods with ventilator treatment. He now feels confident with the SIP and has been able to be at full time work most of the last two years.

The most important indications for the use of SIP have been cancer chemotherapy and total parenteral nutrition.1,3 Complications have mostly been minor, and major complications are rare.1,3 Our patient has had infusions once or twice weekly for two years and no complications have occurred so far. Some patients with SIP are taught self-administration of drugs, in particular, patients receiving parenteral nutrition or antibiotic therapy. We would not consider self-administration in our patient because of the potential hazards of the drugs used, and the possibility of overdosage in a critical situation.

We believe that SIP can be a useful alternative in brittle asthmatics with need for frequent infusions where peripheral veins are poor and a safe venous access is critical.

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