2. European Association for the Study of the Liver (EASL). EASL Clinical Practice Guidelines on the management of benign liver tumours. J Hepatol. 2016;65:386–98.
3. Pol B, Disdier P, Le Treut YP, Campan P, Hardwigsen J, Weiller PJ. Inflammatory process complicating giant hemangioma of the liver: report of three cases. Liver Transpl Surg. 1998;4:204–7.
4. Khalid M, Ahmad M, Jain A, Rizvi I. Atypical giant haemangioma of liver with systemic inflammatory manifestations. BMJ Case Rep. 2013;2013, 10.1136/bcr-2012-007075.
5. Smyrniotis V, Kehagias D, Arkadopoulos N, Kostopanagiotou G, Labrou A, Kondi-Paphitis A. Liver hemangioma with systemic inflammatory manifestations. Am J Gastroenterol. 2000;95:830–2.

Berta Caballol[a,*], Pablo Ruiz[a], Joana Ferrer-Fàbrega[b], Alba Diaz[c], Alejandro Forner[a]

[a] *Servicio de Hepatología, Hospital Clínic de Barcelona, IDIBAPS, Universidad de Barcelona, Barcelona, Spain*
[b] *Servicio de Cirugía Hepato-Biliar, Hospital Clínic de Barcelona, IDIBAPS, Universidad de Barcelona, Barcelona, Spain*
[c] *Servicio de Anatomía Patológica, Hospital Clínic de Barcelona, IDIBAPS, Universidad de Barcelona, Barcelona, Spain*

* Corresponding author.
E-mail address: CABALLOL@clinic.cat (B. Caballol).
2444-3824/
Published by Elsevier España, S.L.U.

Bile duct obstruction secondary to a pseudoaneurysm of the gastroduodenal artery. An unusual presentation☆

Obstrucción biliar secundaria a seudoaneurisma de la arteria gastroduodenal. Una inusual forma de presentación

We present the case of a 74-year-old female patient with a prior history of atrial fibrillation and chronic pancreatitis who attended A&E due to a two-month history of recurrent syncope. She exhibited jaundice and mild diffuse abdominal pain. The examination revealed sinus tachycardia and melaena upon digital rectal examination. The blood tests revealed moderate microcytic anaemia (9.2 g/dl).

An emergency gastroscopy was performed, which showed a Forrest III gastric ulcer with inflammatory edges. The follow-up blood tests showed complete cholestasis (GGT = 1,303 U/l, AP 725 U/l, TBIL = 6.65 mg/dl), with elevated cytolytic enzymes (GOT = 190 U/l, GPT 137 U/l). Amylase and lipase were normal, as were all other values, except the complete blood count that continued to show anaemia (Hb = 9.3 g/dl), so intravenous iron therapy was started. The diffuse abdominal pain, although mild (3/10 on the VAS), persisted despite medical treatment.

Given the clinical suspicion of choledocholithiasis, an abdominal ultrasound was performed that revealed dilation of the extrahepatic and intrahepatic bile duct with a common bile duct of 14 mm and doubtful choledocholithiasis. An ERCP was performed, which confirmed dilation of the bile duct without any filling defects and without trawling any gallstones with a balloon. A protruding papilla was also observed with macroscopically normal mucosa, from which biopsies were taken.

Given the suspicion of ampullary neoplasm, an abdominal contrast CT scan was conducted that confirmed a diagnosis of a periampullary pseudoaneurysm of the superior pancreaticoduodenal artery measuring 2.4 cm in diameter, which compressed the distal bile duct and caused an impression on the ampullary region (Fig. 1).

Finally, the biopsies obtained confirmed nonspecific reactive inflammation and ruled out malignancy.

With the final diagnosis of pseudoaneurysm of the gastroduodenal artery with compression of the bile duct in the ampullary region, it was treated with coil embolisation combined with drainage placed by percutaneous cholangiography to alleviate the distension of the bile duct until the pseudoaneurysm had shrunk, with its removal scheduled for discharge (Fig. 1).

The patient's blood test results gradually improved, with peak bilirubin values of 10.5 mg/dl returning to normal. Haemoglobin levels also improved to 12.7 g/dl.

Although vascular complications secondary to chronic pancreatitis are very rare, their true incidence and prevalence are difficult to estimate and may be as high as 10%.[1] The incidence of bleeding following pseudoaneurysm rupture in chronic pancreatitis is estimated to be 3.2%. The arteries most commonly affected are the splenic artery and the left gastric artery, followed by the gastroduodenal artery, the superior mesenteric artery and the hepatic arteries themselves.[2] Pancreatic pseudoaneurysm is primarily associated with pancreatitis whose course involves the formation of pseudocysts. It has also been associated with hepatobiliary surgery.

The rupture of a pancreatic pseudoaneurysm is the most common cause of bleeding in acute and chronic pancreatitis, accounting for up to 61% of all cases.

Several aetiopathogenic theories to explain pseudoaneurysms secondary to chronic pancreatitis have been postulated, which include the activation of intrapancreatic

☆ Please cite this article as: Casas Deza D, Gotor Delso J, Gascón Ruiz M, Bernal Monterde V, Jimeno Ayllón C, Gracia Ruiz M, et al. Obstrucción biliar secundaria a seudoaneurisma de la arteria gastroduodenal. Una inusual forma de presentación. Gastroenterol Hepatol. 2019;42:384–386.

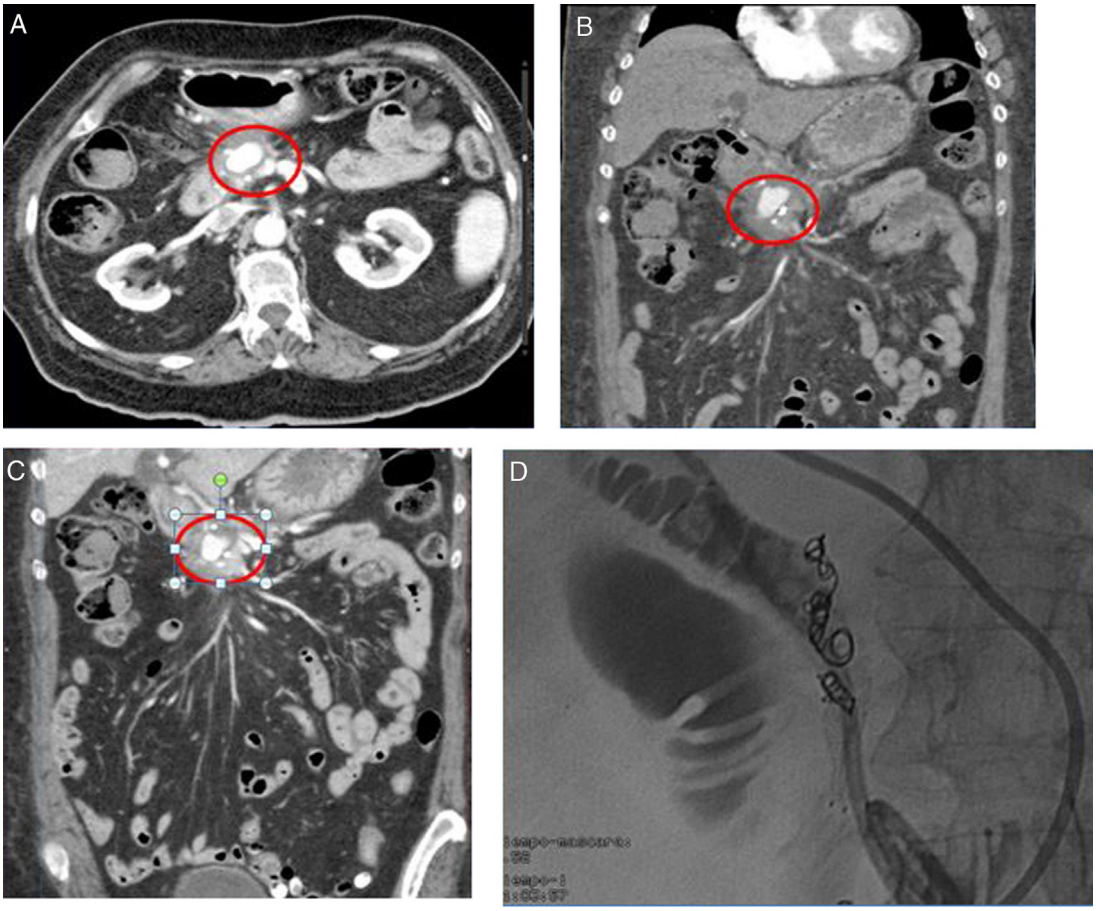

Figure 1 (A) Axial section showing the pseudoaneurysm (circle) and its anatomical relationships. (B and C) Coronal sections showing the pseudoaneurysm (circle) and its impression on the bile duct. (D) Implantation of the coils for embolisation.

enzymes and ischaemic processes leading to separation of the internal elastic lamina.[3]

Obstructive jaundice caused by the extrinsic compression of the bile duct is another form of presentation of pseudoaneurysms. However, this presentation is extremely rare. In the literature review we performed, we were only able to find isolated case reports.

Diagnosis should be confirmed by angiography or contrast CT scan to show arterial dilation and its association with neighbouring structures, in this case the bile duct.[4]

The recommended treatment is embolisation performed by an interventional radiologist. This may involve the use of coils or the placement of a stent, which isolates the pseudoaneurysm from the vessel lumen to reduce the pressure exerted on it. Studies that have assessed the efficacy of treatment with coils have found a high rate of resolution and an acceptable rate of complications, making it a less aggressive alternative to surgery.[5]

References

1. Pang TCY, Maher R, Gananadha S, Hugh TJ, Samra JS. Peripancreatic pseudoaneurysms: a management-based classification system. Surg Endosc Other Interv Tech. 2014;28:2027–38.
2. Balthazar EJ. Complications of acute pancreatitis: clinical and CT evaluation. Radiol Clin North Am. 2002;40:1211–27.
3. Volpi MA, Voliovici E, Pinato F, Sciuto F, Figoli L, Diamant M, et al. Pseudoaneurysm of the gastroduodenal artery secondary to chronic pancreatitis. Ann Vasc Surg. 2010;24:1136e7–11.
4. Hyare H, Desigan S, Nicholl H, Guiney MJ, Brookes JA, Lees WR. Multi-section CT angiography compared with digital subtraction angiography in diagnosing major arterial hemorrhage in inflammatory pancreatic disease. Eur J Radiol. 2006;59:295–300.
5. Saftoiu A, Iordache S, Ciurea T, Dumitrescu D, Popescu M, Stoica Z. Pancreatic pseudoaneurysm of the superior mesenteric artery complicated with obstructive jaundice. A case report. JOP. 2005;6:29–35. Available from: http://www.ncbi.nlm.nih.gov/pubmed/15650282 [accessed 18.03.18].

Diego Casas Deza[a,*], Jesús Gotor Delso[a], Marta Gascón Ruiz[b], Vanesa Bernal Monterde[a], Cristina Jimeno Ayllón[a], Marta Gracia Ruiz[a], Luis Sarria Octavia de Toledo[c], Joaquín Medrano Peña[d]

[a] *Servicio de Aparato Digestivo, Hospital Universitario Miguel Servet, Zaragoza, Spain*
[b] *Servicio de Oncología Médica, Hospital Clínico Universitario Lozano Blesa, Zaragoza, Spain*
[c] *Servicio de Radiodiagnóstico, Hospital Miguel Servet, Zaragoza, Spain*
[d] *Sección de Radiología Intervencionista, Servicio de Radiodiagnóstico, Hospital Miguel Servet, Zaragoza, Spain*

* Corresponding author.
E-mail address: diegocasas8@gmail.com (D. Casas Deza).
2444-3824/

Shadow cells in a cutaneous epidermoid cyst: Searching for a polyposis syndrome☆

Quiste epidermoide cutáneo con células sombra: a la búsqueda de un síndrome polipósico

We present the case of an asymptomatic 69-year-old man with a prior history of removed retroauricular epidermoid cyst. Histology revealed foci of intraluminal shadow cells, a finding consistent with Gardner's syndrome. He did not exhibit any other associated extraintestinal manifestations and had no family history of gastrointestinal disease. The panendoscopy revealed multiple millimetric oesophageal polyps, the histology of which did not indicate adenomatous changes. The colonoscopy showed six polyps of between 2 and 20 mm, which were resected. The histological study was consistent with tubular adenoma, with low-grade dysplasia observed in three of them and high-grade dysplasia in the other three. In addition, a 15-mm ulcerated neoformation was identified in the rectosigmoid junction, the biopsies of which were consistent with adenocarcinoma. Five days after the colonoscopy, the patient had low-grade fever and pain in the right iliac fossa. A CT scan was ordered that revealed findings suggestive of acute appendicitis, which was initially managed conservatively. Neoadjuvant radiotherapy was started and the follow-up CT scan revealed a focal hepatic injury. Surgery was performed, consisting of lower anterior rectal resection, liver metastasectomy and appendectomy, finding adenocarcinoma with lymph node involvement, adenocarcinoma liver metastasis and appendiceal goblet cell carcinoid tumour with free edges in the histological study, respectively. The genetic study for the *APC* mutation was negative. Adjuvant chemotherapy with fluorouracil and oxaliplatin was then administered, with no evidence of tumour recurrence to date.

Shadow or ghost cells are keratinised eosinophilic cells with an unstained central area at the site of the shadow of the lost nucleus. Their presence is an indication of an aborted attempt to form hair shafts (Fig. 1). They are a characteristic, albeit nonspecific, histological finding of pilomatricoma and pilomatrical carcinoma.[1] Familial adenomatous polyposis is an autosomal dominant disorder secondary to the mutation of the *APC* tumour suppressor gene and is characterised by the development of multiple premalignant adenomatous colon polyps. There are numerous variants of this disease, one of which is Gardner's syndrome, which is defined as the association of colonic polyposis with the onset of various extraintestinal manifestations.[2] Of these, the most common are epidermoid cysts and skin fibromas, osteoma of the face and long bones, hypertrophy of the retinal pigment epithelium, dental abnormalities and desmoid tumours of the abdominal and intra-abdominal wall, as well as the development of malignant tumours at various sites (thyroid, duodenal, ampullary, pancreatic, gastric, etc.).[3] Some of these extraintestinal manifestations may occur years before the onset of gastrointestinal symptoms.

Epidermoid cysts occur in 50–60% of all cases of Gardner's syndrome compared to 9–10% in the general population.[2] They generally develop before puberty, may be the first sign of the disease and carry no risk of malignancy. Accumulations of shadow cells and foci of matrix keratinisation inside these cells have been reported, similar to what occurs in pilomatricoma.[4] Although they constitute a common finding in the physical examination of patients

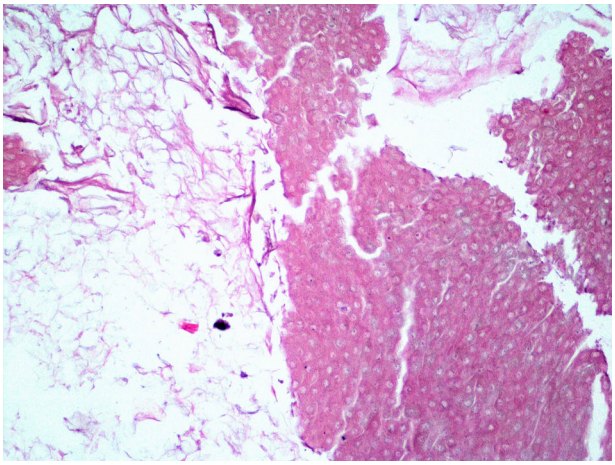

Figure 1 (H&E, ×40) Cyst contents comprising sheets of flaky keratin and masses of shadow cells.

☆ Please cite this article as: García de la Filia Molina I, Crespo Pérez L, Ríos León R, Barbado Cano A, Moreno García del Real C, Aburto Bernardo A, et al. Quiste epidermoide cutáneo con células sombra: a la búsqueda de un síndrome polipósico. Gastroenterol Hepatol. 2019;42:386–387.