Hepatectomy Cures a Cough: Giant Cavernous Hemangioma in a Patient With Persistent Cough

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Cavernous hemangiomas are the most common type of benign liver tumor. Although these tumors are often asymptomatic, they can occur with an array of symptoms. The authors describe a case of a 51-year-old man who presented to the emergency department with a relentless cough, nausea, and abdominal pain. Results of a computed tomography scan suggested the patient had a giant cavernous hemangioma on his liver; microscopic evaluation confirmed this diagnosis. The hemangioma was initially deemed unresectable and the patient was treated with one session of hepatic artery embolization. The embolization was unsuccessful at easing the patient’s symptoms, however, and a hepatic lobectomy and resection was performed. After surgical intervention, the patient’s symptoms resolved. The present case illustrates an unusual instance in which chronic cough was cured through hepatectomy for giant cavernous hemangioma. To our knowledge, no reports of coughing as a primary symptom of giant cavernous hemangioma have been previously reported in the literature.

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Cavernous hemangiomas (CHs) are the most common type of benign liver tumor. Studies have cited prevalence rates for CHs ranging from 1.4% to 52%.2 However, based on autopsy data, most major texts cite a 7% occurrence rate for CHs.3,4

Cavernous hemangiomas consist of multiple large vascular channels filled with slowly moving blood. The channels are lined by a single layer of endothelial cells and supported by collagenous walls.4 The margins of the tumors are usually well defined. Cavernous hemangiomas greater than 5 cm in diameter are termed giant cavernous hemangiomas (GCHs) and may contain areas of hemorrhage, thrombosis, extensive hyalinization, fibrosis, and calcifications. Ectasia is believed to contribute to lesion enlargement.5,6

Cavernous hemangiomas are 3 times more common in women than in men.7 The vast majority of CHs are asymptomatic.7 Prospective trials have shown that while CHs are often found in asymptomatic patients, GCHs occur with symptoms in 47% of patients.5,7 Symptoms associated with GCHs include pain in the right upper quadrant of the abdomen, fever, jaundice, and cholestasis.6 Large CH lesions can create pressure on the stomach and duodenum and cause abdominal pain, early satiety, nausea, and vomiting.5 Compression of the inferior vena cava may result in Budd-Chiari syndrome.1 Acute thrombosis in patients with CH can lead to fever, abdominal pain, and abnormal results in liver function tests.8 Spontaneous or posttraumatic thrombotic rupture occurs in 1% to 4% of patients with CH; this condition has a mortality rate as high as 60%.9

Sometimes, patients with hemangioma will present with anemia or low blood platelet counts. Known as Kasabach-Merritt syndrome, this condition occurs when red blood cells or platelets are sequestered or destroyed in the tumor.

We report a case of a man who presented with persistent cough as a primary symptom of GCH. To our knowledge, no reports of this kind have been previously reported in the literature.

Report of Case
A 51-year-old man presented to the emergency department with complaints of an unrelenting cough, nausea, and constant, nonspecific pain in the right-upper quadrant of his abdomen. The patient’s cough worsened at night, causing insomnia. The patient also complained of night sweats and a 60-pound weight loss from anorexia in the past year. His abdominal pain and anorexia began at the same time. His past medical history was notable for hypertension, hypercholesterolemia, anemia, renal lithiasis, and impaired coagulation. The patient had no history of alcohol or tobacco abuse and no family history of liver disease. The patient also had no history of skin rashes, hypoglycemia, gastrointestinal bleeding, or vitiligo. While he had no personal history of cancer, his family history was remarkable for his maternal grandmother having...
cancer of the jaw, ear, and parotid gland. The patient was taking colesevelam hydrochloride for his hyperlipidemia. Despite the use of acid suppression, the patient’s pain did not improve. In addition, he could not elicit reflux during positional therapy.

The patient’s physical examination revealed an enlarged liver and marked tenderness in the right-upper quadrant and epigastrium regions of his abdomen. The examination did not show evidence of scleral icterus, splenomegaly, ascites, spider angiomata, or palmar erythema. Results of the patient’s ear, nose, and throat and lung examinations were normal. Laboratory findings (reference range) were as follows: hemoglobin level, 9.2 g/dL (13.5-16.7 g/dL); blood platelet count, 472 × 10^3/µL (150-400 × 10^3/µL); prothrombin time, 16.4 seconds (11.5-14.3 seconds); albumin, 3 g/dL (3.9-4.8 g/dL); and alkaline phosphatase, 214 U/L (39-117 U/L) with normal transaminase levels. Viral serologies, autoimmune markers, iron levels, and alpha fetoprotein levels were unremarkable. Genetic testing was negative for von Hippel-Lindau disease. A computed tomography (CT) scan of the chest, abdomen, and pelvis was completed with intravenous and oral contrast. The results of the CT scan revealed several small masses in the right lobe of the liver and complete replacement of the left lobe of the liver by a mass that was approximately 16 × 11 × 12 cm³ (Figure 1 and Figure 2). Results of a tagged red blood cell scan confirmed uptake in three homogenous areas of the right lobe of the liver, peripheral uptake in a large region of the left lobe of the liver, and a decreased central uptake consistent with CH. The presence of CH was confirmed with a microscopic evaluation.

After an exploratory laparotomy, the CH in the patient’s liver was initially deemed unresectable and the patient was treated with one session of left-sided hepatic artery chemoembolization. While the patient’s symptoms initially improved, they gradually returned over the next 6 months. At a follow-up examination 6 months after the hepatic artery embolization, a CT scan revealed no change in the size of the GCH. Results of the follow-up examination also showed new findings including a small cervical CH at the C7 vertebrae, an 8 × 6 × 8 mm³ intracanalicular acoustic neuroma, a bony tumor of the left-sided zygomatic bone, and a 5 × 5 × 9 mm³ prolactinoma.

Nine months after embolization, the patient underwent excision and reconstruction of the vascular lesion on his left-sided zygomatic bone because of progressive pain in the area. Three months after the zygomatic bone resection, serial imaging showed little change in the acoustic neuroma, C7 hemangioma, and prolactinoma. The patient’s symptoms of cough with insomnia and abdominal pain continued to persist.

Based on these findings, the liver transplant team reconsidered the patient for surgery. A left-sided hepatic lobectomy and resection of a 20 × 10 × 12 cm³ GCH was performed. The smaller CHs in the remainder of the patient’s liver were not resected. The patient’s postoperative course was uncomplicated, and he was discharged after a 3-day hospital stay. After the surgery, the patient’s cough resolved within 1 week and the rest of his symptoms resolved within 2 weeks. He has remained asymptomatic for more than 1 year.

**Comment**

The present case is notable because the patient with GCH presented with persistent cough as a primary symptom. Giant cavernous hemangiomas have been known to cause an array of symptoms, but, to our knowledge, no reports of coughing as a primary symptom of GCH have been previously reported in the literature.

Cavernous hemangiomas are typically diagnosed by identifying classic patterns on imaging studies. For example, ultrasonography will show an isoechic lesion with a hyperechoic rim in the abdomen of a patient with CH. However, ultrasonography is limiting when used as the single tool for diagnosis because, with this method, images of malignant tumors can often have appearances similar to CHs. Technetium 99m–labeled red blood cell imaging has an 89% sensitivity and almost 100% specificity for CHs larger than 2 cm. However, rare false-positive results from hypervascular malignancies have been reported. A CT scan with intravenous contrast will often show a peripheral ring of enhancement, with central filling on delayed images. Magnetic resonance imaging of the abdomen shows similar peripheral enhancement followed by central enhancement of the CT. T2-weighted imaging has a 100% sensitivity and a 92% specificity when used to differentiate CH from malignant hepatic tumors.

In asymptomatic patients with CHs smaller than 10 cm, observation is recommended. Intervention is recommended for CHs that occur with symptoms. If embolization fails, as in our case, surgery is considered a viable treatment option. Current surgical options for the treatment of patients with CH include: liver resection, enucleation, hepatic artery ligation, and liver transplantation. Of these, enucleation is the preferred treatment option because it is associated with the lowest risk for complications. Complication rates for enucleation range from 11% to 16%; mortality rates range from 0% to 2.5%. By comparison, complication rates for hepatectomy have been reported to be as high as 44%. In our patient, enucleation was not possible without lobectomy because the entire left lobe was replaced with GCH.

**Conclusion**

Considering chronic cough is a common symptom accounting for 30 million primary care physician visits per year, it is important for physicians to consider atypical diagnoses. In the present case, we believe the GCH irritated the patient’s diaphragm, causing the persistent cough. The patient’s cough...
went away after the removal of the GCH, leading us to believe the hepatic lobectomy and resection was responsible for resolution of his symptoms. More than 1 year after the surgery, the patient’s symptoms of cough, fevers, anemia, and abdominal pain had not returned.

References