Hemoptysis due to pulmonary pseudosequestration secondary to gastro-pulmonary fistula after a revisional bariatric operation

Carlos Cerdán Santacruz, María Conde Rodríguez¹, Andrés Sánchez-Pernaute¹, Antonio José Torres García¹

We report the case of a patient with a history of a complicated revisional bariatric operation who developed

a lung pseudosequestration secondary to a gastro-pulmonary fistula. As the patient presented with recurrent hemoptysis, she was initially submitted to embolization of the aberrant vessels and later to a definite operation,

which consisted on a diversion of the gastric fistula into a Roux-en-Y intestinal loop. It is an exceptional case

about late complications of bariatric surgery, and it underlines the importance of discarding these complications

even when the clinical manifestations affect another anatomic region different from the operated abdomen.

Bariatric surgery complications, gastrobronchial fistula, gastric mucocele, lung pseudosequestration

Abstract:

Key words:

Departments of General Surgery, Santa Elena Clinic, Madrid, ¹General Surgery, Esophago-Gastric and Bariatric Surgery, San Carlos Hospital, Madrid, Spain

Address for correspondence:

Dr. Carlos Cerdán Santacruz, Department of General Surgery, Santa Elena Clinic, C/La Granja N°8, Madrid - 28003, Spain. E-mail: carloscerdansantacruz@ hotmail.com

> Submitted: 28-03-2014 Accepted: 19-05-2014



Website: www.thoracicmedicine.org DOI: 10.4103/1817-1737.140141 Over the last 15 years there has been an impressive increase in the number of bariatric operations performed worldwide. Most patients obtain a definite solution for their obesity and metabolic problems; however, changes in the intestinal anatomy performed by the operation might predispose to strange complications with difficult solutions. Herein we report the case of a patient with life-threatening hemoptysis after a revisional bariatric surgery.

Case Report

A 61-year-old female had been submitted in the past to a vertical banded gastroplasty for morbid obesity and later was revised to a duodenal switch for continuous vomiting. In the second surgery, the crossing of staple lines left an excluded portion of the stomach developing a gastric mucocele which opened into the left subphrenic space.^[1] The resulting abscess was satisfactorily drained through an indwelling catheter inserted intercostally and left in place for 5 months. As a consequence, the patient developed chronic costal pain and left lower lobe athelectasis. At least 4 years after the second operation, she complained of irritative cough and occasional hemoptysis, which progressively increased in frequency, intensity, and clinical significance. Bronchoscopy was performed, but it resulted with unremarkable findings. A computed tomography (CT) revealed the gastric mucocele in the upper abdomen and athelectasis in the left lower lobe. An arteriogram was performed,

and a systemic-pulmonary arterial fistula was found, communicating the left phrenic artery with the pulmonary circulation. The left phrenic artery was embolized, but hemoptysis recurred. A second arteriogram revealed communication between the left mammary artery and some intercostal branches with the pulmonary circulation [Figures 1a and b], and embolization was repeated. Thereafter hemoptysis changed to a blood stained bronchorrhea. To rule out the implication of the gastric mucocele in the recurrent hemopthysis, the stomach wall was punctured and ditrizoate meglumine (Gastrographin®) introduced in the mucocele's lumen through endoscopic ultrasonography; an immediately performed CT revealed fistulization from the gastric mucocele into the left lower bronchus [Figures 2a and b].

The patient was submitted to surgery through a median laparotomy. The upper left quadrant of the abdomen was "blocked" with the spleen, pancreas, and gastric remnant intimately adherent to the diaphragmatic dome. The residual mucocele was found completely opened to the peritoneal cavity with a normalappearing mucosa [Figure 3a]. A track was seen communicating the subphrenic space with the lung parenchyma through a hole in the diaphragm [Figure 3b]. The orifice was sutured, and the gastric mucocele diverted into a Roux-en-Y intestinal loop. Apart from a severe pneumonia which needed ventilatory support, the evolution was finally satisfactory, with total



Figure 1: Arteriograms showing systemic inferior left lobe support and pseudosequestration. (a) Left phrenic artery. (b) Intercostal branches after coils for the embolization of internal mammary artery and left phrenic artery



Figure 2: CT scan images. (a) Gastric mucocele filled in with oral contrast. (b) Left lower bronchii filled in with oral contrast secondary to gastro-bronchial fistula



Figure 3: Intraoperatory pictures. (a) Opened to the abdominal cavity gastric mucocele. (b) Diaphragmatic hole communicating the gastric mucocele effusion with the left lower lobe

remission of cough, hemoptysis and bronchorrhea three years after the procedure.

Discussion

Two significant aspects have to be emphasized. First, a differential diagnosis between lung sequestration and pseudosequestration should be completed, in order to accomplish the correct surgical therapy; and secondly, the results of the previous abdominal operation should be thoroughly reviewed to discard a long-term complication from a previously complicated bariatric surgery.

Lung sequestration consists on a congenital systemicto-pulmonary arterial supply that might course asymptomatically, and manifest in the adulthood. Pseudosequestration is an acquired disease in which the systemic arterial supply to the lungs develops secondary to the neovascularization generated due to an inflammatory chronic stimulus, mainly bronchiectasis.^[2]Systemic arteries supplying the lung parenchyma are usually bronchial arteries, but sometimes transpleural systemic-pulmonary artery anastomosis are found. The systemic vessels can be the intercostal arteries, internal mammary arteries, inferior phrenic arteries, or other regional arteries. Radiological appearance and clinical evolution are useful to differentiate pseudosequestration from sequestration. In lung sequestration one anomalous vessel is identified supplying the mass, while in pseudosequestration there are many small hypertrophied vessels more prominent in the surface of the mass; a pleural blush and a peripheral enhancement are also typical of pseudosequestration. Pseudosequestration usually responds to systemic medical treatment but if only embolization is performed, it generally recurs, as has been previously described.^[2]

A mucocele of the digestive tract is a cystic structure with a mucosal lining that secretes its mucoid content. Although frequently oligosymptomatic, occasionally it causes severe intraabdominal infections.^[1,3] This case illustrates a potentially lethal late-onset thoracic complication, lung pseudosequestration and hemoptysis, originated in the abdomen as a sequel of a revisional bariatric surgery.

The cornerstone of this case was to stablish the relationship between the bariatric operations and the pulmonary pathology. The residual mucocele opening to the subphrenic space was the origin of the pseudosequestration through an established mucocele-pulmonary fistula facilitated by the indwelling catheter employed to treat the subphrenic abscess. It is not unfrequent to see cardiac, mediastinal or pleural complications after gastric surgery, usually as a result of an anastomotic leakage.^[4,5] Both operations on this patient were performed in the upper part of the stomach, close to the esophagogastric junction; an extremely delicate anatomic region. Leaks from the upper stomach after a sleeve resection are difficult to treat, and generally take long to heal despite adequate treatment under total parenteral nutrition, appropriate drainage, or intragastric covered stents.^[6-8] In this case, the leak was from a gastric mucocele resulting from the crossing of staple lines in the second operation.^[1] The subphrenic abscess derived from the mucocele was transdiaphragmatically drained, giving rise to a gastro-pulmonary fistula and avoiding the natural evolution of the mucocele to an asymptomatic cyst with atrophied mucosa. The fistula originated a chronic and limited lung infection, which generated neovasculature from the systemic circulation and secondary hemoptysis. The performance of the CT after the introduction of contrast medium into the stomach was crucial for the optimal surgical treatment.

Mucocele exclusion through a Roux-en-Y diversion was performed, a safe and effective technique, widely employed in abdominal surgery after organ resections or to drain visceral fistulae and cysts.^[9] A transthoracic approach leaving an untouched gastric mucocele would have probably lead to the persistence of thoracic symptoms, in this case hemoptysis, as previously reported.^[10]

Bariatric operations, though routinely performed, remain complex operations. It is mandatory to discard complications of these techniques in patients presenting with abdominal or thoracic symptoms, as there might lay the key to solve the clinical problem.

References

- Sánchez-Pernaute A, Pérez-Aguirre E, Talavera P, Robin A, Díez-Valladares L, Cabrerizo L, *et al.* Mucocele of the gastric tube after conversion of vertical banded gastroplasty to duodenal switch: Not just a radiological image. Obes Surg 2006;16:524-7.
- Do KH, Goo JM, Im JG, Kim KW, Chung JW, Park JH. Systemic arterial supply to the lungs in adults: Spiral CT findings. Radio Graphics 2001;21:387-402.
- Haddad R, Lima RT, Boasquevisque CH, Marsico GA. Symptomatic mucocele after esophageal exclusion. Interact Cardio Vasc Thorac Surg 2008;7:742-4.
- Kim WJ, Choi EJ, Oh YW, Kim KT, Kim CW. Gastropericardial fistula-induced pyopneumopericardium after esophagectomy with esophagogastrectomy. Ann Thorac Surg 2011;91:e10-1.
- Fuks D, Dumont F, Berna P, Verhaeghe P, Sinna R, Sabbagh C, et al. Case report - Complex management of a postoperative bronchogastric fistula after laparoscopic sleeve gastrectomy. Obes Surg 2009;19:261-4.
- 6. Blackmon SH, Santora R, Schwartz P, Barroso A, Dunkin BJ. Utility

of removable esophageal covered self-expanding metal stents for leak and fistula management. Ann Thorac Surg 2010;89:931-7.

- Campos JM, Pereira EF, Evangelista LF, Siqueira L, Neto MG, Dib V, *et al.* Gastrobronchial fistula after sleeve gastrectomy and gastric bypass: Endoscopic management and prevention. Obes Surg 2011;21:1520-9.
- 8. Alharbi SR. Gastrobronchial fistula a rare complication post laparoscopic sleeve gastrectomy. Ann Thorac Med 2013;8:179-80.
- Baltasar A, Serra C, Bengochea M, Bou R, Andreo L. Use of Roux limb as remedial surgery for sleeve gastrectomy fistulas. Surg Obes Relat Dis 2008;4:759-63.
- 10. Darbari A, Tandon S, Singh GP. Gastropleural fistula: Rare entity with unusual etiology. Ann Thorac Med 2007;2:64-5.

How to cite this article: Santacruz CC, Rodríguez MC, Sánchez-Pernaute A, García AJ. Hemoptysis due to pulmonary pseudosequestration secondary to gastro-pulmonary fistula after a revisional bariatric operation. Ann Thorac Med 2014;9:242-4.

Source of Support: Nil, Conflict of Interest: None declared.