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SCIENTIFIC LETTER

A challenging alpha-fetoprotein after liver transplantation

Un diagnóstico desafiante de alfafetoproteína elevada en paciente trasplantado hepático

To the Editor,

The most common causes for elevation of serum alpha-fetoprotein are hepatocellular carcinoma and germ cell tumors.[1] Less commonly, it may be associated with alpha-fetoprotein producing gastric cancer, a rare subtype of gastric adenocarcinoma (2–15%) that is usually diagnosed in advanced stage and typically associated with poor prognosis and higher rates of lymphatic and venous invasion, lymph node metastasis and liver metastasis than conventional gastric cancer. Accordingly, it is demonstrated that increased alpha-fetoprotein is associated with overall poorer prognosis in gastric adenocarcinoma.[2]

We report the case of a 39-year-old female with previous medical history of liver transplantation for alcoholic liver cirrhosis performed 4 years earlier, and abstinent since then, who was referred to our hospital for investigation of a progressive rise of serum alpha-fetoprotein (peak: 1100 mg/dL). Since liver transplantation, she had been under immunosuppression with tacrolimus without any signs of graft rejection. She referred asthenia, post-prandial fullness, nausea and vomiting. Abdominal computed tomography scan revealed gastric parietal thickening suggestive of malignancy (Fig. 1A) associated with peri-gastric lymphadenopathy and two hypodense liver nodules with 10 and 32 mm, both located in segment II and demonstrating peripheral hyperenhancement during arterial phase without evident washout during portal phase, suggestive of liver metastasis. Liver allograft did not display signs of cirrhotic transformation. Therefore, she undergone upper digestive endoscopy that showed a large ulcerated polypoid lesion involving the smaller curvature along body and antrum (Fig. 1B). Biopsies of the lesion were performed and histopathological examination revealed adenocarcinoma (Fig. 1C), with strong immunohistochemical positivity for alpha-fetoprotein (Fig. 1D), consistent with alpha-fetoprotein producing gastric cancer (stage IV). After multidisciplinary discussion, palliative chemother-

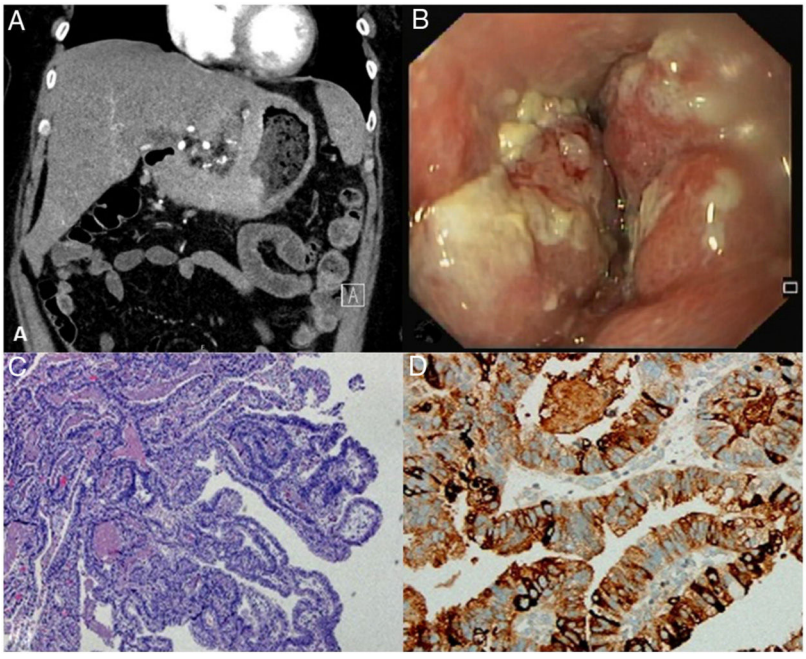

Figure 1 (A) Abdominal computed tomography scan revealed gastric parietal thickening and a luminal protrusion suggestive of malignancy. (B) Esophagogastroduodenoscopy revealed a large protruding ulcerated lesion extending along the smaller curvature of antrum and body. (C) Histopathological examination revealed a solid neoplasia forming glandular structures consistent with adenocarcinoma. (D) Immunohistochemistry showed strong positivity for alpha-fetoprotein in neoplastic cells.

apy was started. Additionally, because of food intolerance and vomiting, a palliative endoscopic stent was placed. However, the patient evolved unfavorably and died after 4 months.

Alpha-fetoprotein producing gastric cancer has been previously described in patients with chronic liver disease,[3–5] including cases associated with liver cirrhosis secondary to chronic hepatitis B[3] or unknown etiology[4] or with noncirrhotic chronic hepatitis C.[5] However, to the best of our knowledge, we describe the first case in association with liver transplantation. Even though this association could be coincidental, it is possible that post-transplant immunosuppression may have played a role in the development of malignancy. Further studies are needed to determine whether immunosuppression is effectively a risk factor for this uncommon malignancy.

In conclusion, this case demonstrates that alpha-fetoprotein producing gastric cancer must be considered in the differential diagnosis of elevated alpha-fetoprotein, particularly in the presence of associated dyspeptic symptoms. Although the incidence of this malignancy does not appear to be increased in patients with liver disease, hepatologists should be aware of its existence when following these patients due to frequent use of alpha-fetoprotein in this setting.

Conflict of interest

The authors declare that they have no conflict of interest.

References

1. Wong RJ, Ahmed A, Gish RG. Elevated alpha-fetoprotein: differential diagnosis – hepatocellular carcinoma and other disorders. Clin Liver Dis. 2015;19:309–23.
2. Baek SK, Han SW, Oh DY, Im SA, Kim TY, Bang YJ. Clinicopathologic characteristics and treatment outcomes of hepatoid adenocarcinoma of the stomach, a rare but unique subtype of gastric cancer. BMC Gastroenterol. 2011;11:56.
3. Gallo P, Gentilucci UV, Taffon C, Galati G, De Vicentis A, Muda AO, et al. A challenging alfa-fetoprotein in a cirrhotic patient. Acta Gastroenterol Belg. 2014;77:66–7.
4. Umekawa Y, Watanabe M, Ikeda S, Fukumoto S, Hirakawa H, Shimada Y. Alpha-fetoprotein-producing early gastric cancer accompanying liver cirrhosis: a case report. J Gastroenterol. 1994;29:66–70.
5. Sohda T, Tomioka Y, Inomata S, Morita I, Eguchi K, Aoyagi K, et al. Alpha-fetoprotein (AFP)- and des-gamma-carboxy prothrombin (DCP)-producing adenocarcinoma of the stomach with liver metastasis in a patient with chronic hepatitis C. Intern Med. 2005;44:294–8.

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Tofacitinib for the treatment of inflammatory condition of the ileoanal pouch refractory to infliximab

Tofacitinib para el tratamiento de la afección inflamatoria del reservorio refractaria a infliximab

Up to 5%–15% of patients with ulcerative colitis (UC) require surgical treatment, with proctocolectomy with ileoanal pouch being the technique of choice. Pouchitis is its most common complication. Pouchitis includes a range of situations that remain ill-defined, ranging from classical pouchitis to Crohn's disease of the pouch.[1] The increasing incidence of this complication and the risks and loss of quality of life associated with the extraction of the pouch have increased interest in finding therapeutic alternatives in this situation.[2] We present the case of a patient with a pouch and chronic iron deficiency anaemia secondary to patchy inflammatory involvement of the pouch who received rescue treatment with tofacitinib after failure of treatment with TNF inhibitors.

This was a 48-year-old male patient with left-sided UC of 18 years of evolution requiring proctocolectomy with pouch due to refractory disease. Five years after surgery, there was onset of rectorrhagia and severe iron deficiency anaemia. Hence, a pouchoscopy revealed deep, friable ulcers in the pouch, causing stenosis of the access to afferent and efferent loops. Other causes of anaemia were ruled out (negative coeliac disease study, gastroscopy and capsule endoscopy without findings) and, given the characteristics of the lesions, it was decided to start treatment with oral budesonide with no clinical response, so combined treatment with infliximab (5 mg/kg every eight weeks) and azathioprine was started. Despite this, the patient required high-dose intravenous iron repletion and even periodic blood transfusion, with no endoscopic improvement, so infliximab was intensified to every six weeks. However, in successive endoscopies, ulcerated stenosis of the access orifice to afferent and efferent loops persisted, so local injection of infliximab and antibiotic therapy with metronidazole 500 mg/every eight hours for four weeks was administered without success. Given the patient's reluctance to have the pouch removed and the lack of therapeutic alternatives in this situation, treatment with infliximab was maintained for 10 years, with a progressive increase in the requirement for