Metachronous splenic metastasis in an anal squamous cell carcinoma

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Although the spleen is a highly vascularised organ, metastasis remains a very rare situation, with an incidence ranged between 0.3 and 7.3% (1). They mainly occur in a context of multivisceral metastatic cancer at terminal stage and present as multiple and asymptomatic lesions diagnosed by incidental radiological assessment performed either in the regular follow-up of patients with cancer or in the workup at the time of the primary tumor diagnosis. Anal squamous cell carcinoma (ASCC), a relatively rare gastrointestinal malignancy, mainly associated with liver and lymph nodes metastases, has not been described yet as a potential primary tumor associated with spleen metastases (2).

We describe here the case of a 60-year- old woman admitted in our hospital for a locally advanced poorly differentiated squamous cell anal carcinoma clinically classified as an cT2cN3cM0 tumor. Multimodal therapies including chemotherapy (combination of cisplatin 100 mg/m^2 day 1 and vepesid 100 mg/m^2 days 1-2-3, every 3 weeks) and radiation therapy (28 fractions of 1.8 Gy, for a total of 50.4 Gy) have been sequentially administered, leading to complete local disease remission. Spleen recurrence appeared twelve weeks after completion of radiotherapy (Fig. 1). Since the spleen was the only metastatic organ, splenectomy was performed, with histopathological findings consistent with the anal tumor. Six months later, tumor recurred in an isolated lymph node located near the pericardium, which was also surgically removed, with a disease-free survival of 2 years.

Splenic metastases from solid tumors are considered exceptional, due to inhibitory effect of the splenic microenvironment on the growth of metastatic cells as well as secondary to the constant blood flow through the spleen (3). They are usually multiple and generally appear in a context of multivisceral disease, in contrast with our case where splenic involvement was the only observed metastatic site. Solitary spleen metastases are in fact rather rare with only a few cases described in the literature (4). Adenocarcinomas, skin melanomas and squamous cell carcinomas are observed in about 60, 30 and 10% of the cases, respectively (1,3,5). In case of splenic involvement only, some authors consider patients eligible for surgical treatment. Palliative surgery could be used in case of symptomatic splenomegaly to improve quality of life, whereas curative intent, a rare condition, could achieve long-term remission in patients with solitary metastases confined to the spleen (4). Our attitude was therefore consistent with the literature and allowed our patient to stay disease-free 2 years after last metastatic removal. To our knowledge, ASCC, a relatively rare gastrointestinal malignancy with an incidence of 0.3 to 1.0/100,000/year (2), has not been described yet as a potential tumor giving spleen lesions. Usually confined in the anal wall, regional lymph node involvement is observed in approximately 50% of the patients, whereas metastatic disease appears in 10 to 17% of patients previously treated for their regional disease (5). This rapid spread into regional lymphatic system contrasts with the usual haematogenous disseminative mechanism by which spleen metastases appear (6). Locally advanced tumors are usually managed by chemoradiation therapy (7,8), whereas surgery is reserved for patients with either residual disease or local recurrence. The choice of chemotherapy instead of chemoradiation as first-line treatment in our patient was based upon patient's tumor

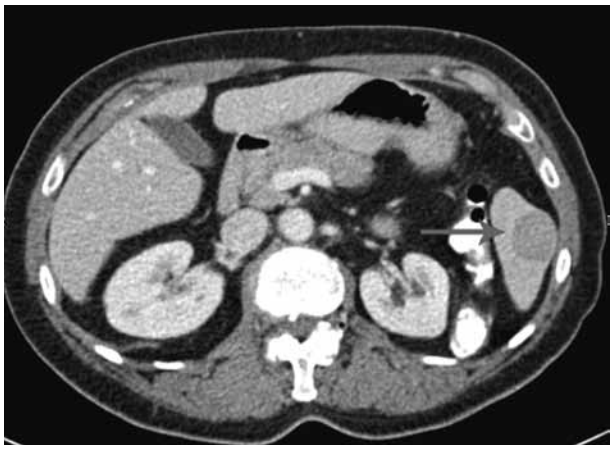

Fig. 1. — Computerized Tomography scan showing splenic metastasis (arrow).

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aggressiveness (bilateral inguinal and mesorectal lymph node involvement associated with a nearly 100% Ki67). Furthermore, Meropol *et al*. showed promising results with an induction cisplatin-based chemotherapy followed by radiation therapy (complete response : 82% ; 61% disease-free survival after a 4-years follow-up) (9).

In conclusion, splenic involvement is a rare metastatic site, rather exceptional in ASCC. Close radiological follow-up allowed physicians to quickly detect and manage recurrence by adequate radical splenectomy. This attitude achieved long-term survival despite aggressive tumor behavior. In case of ASCC, follow-up guidelines are mainly based upon physical examination and works-up performed when symptoms potentially related to the disease appear. This case therefore points out the potential need for close follow-up in ASCC.

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