CASE REPORT

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“Burned out” testicular seminoma presenting as a primary gastric malignancy

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Abstract In contrast to primary gastric adenocarcinomas, germ cell tumors are potentially curable even when metastatic. It is therefore essential for clinicians and pathologists to be aware of the spectrum of unusual manifestations of germ cell malignancies. Here we report on a 55-year-old man who presented with clinical and endoscopic features indicative of a primary gastric carcinoma. Surprisingly, the ulcerative mucosal lesion was found to be due to a metastasis from an occult, “burned-out” testicular seminoma. This case describes the radiological and pathological features that helped differentiate this rare situation from the much more common gastric adenocarcinoma, and extends the diagnostic possibilities that must be considered in patients presenting with gastric ulcers.

Key words Neoplasm metastasis · Neoplasms, unknown primary · Seminoma · Stomach · Stomach ulcer · Testicular neoplasms

Introduction

Gastric metastasis as the initial presentation of a seminoma has been only very rarely reported.1–4 All these cases were associated with significant retroperitoneal disease or occurred late in the course of the disease. The “burned-out phenomenon” in germ cell tumors is described as the presence of a metastatic extragonadal tumor with evidence of a regressed testicular primary. This event is also very rare, but well documented, and thought to be secondary to immunologic events.5–9 We present here a unique case of seminoma, combining all of these very unusual features.

Case report

A 55-year-old man presented with a month-long history of epigastric pain, melena, and hematemesis. His past history included gastroesophageal reflux disease, hepatitis C, colonic villous adenoma, smoking, and alcohol abuse. Upper gastrointestinal endoscopy identified a deep ulcer at the greater gastric curvature with an exudative base and sharp, erythematous edges (Fig. 1A). Biopsies were taken from the ulcer base and periphery, and initially read as poorly differentiated adenocarcinoma. Computed tomography (CT) scans of the chest, abdomen, and pelvis did not demonstrate extragastric extension or evidence of abdominal lymphadenopathy. He underwent exploratory laparotomy and subtotal gastrectomy for presumed gastric adenocarcinoma. Intraoperatively, a 3-cm mass at the greater curvature was noted, without evidence of extragastric spread.

Histologically, the tumor consisted of sheets of infiltrating large epithelioid cells with clear cytoplasm and large round nucleus with prominent nucleolus, admixed with variable numbers of inflammatory cells with a preponderance of small mature lymphocytes (Fig. 1B). Tumor cells were diffusely and strongly positive for cytoplasmic glycogen (PAS+/PAS with diastase negative). Immunohistochemical studies showed strong and diffuse reactivity for c-KIT, membranous positivity for placental alkaline phosphatase (PLAP) and podoplanin (D2-40), and minimal focal positivity for cytokeratin cocktail (Fig. 1 C-F). The tumor was negative for CD15, CD30, CD34, CD43, CD45, CD68, cytokeratins 7 and 20, epithelial membrane antigen, synaptophysin, S-100, smooth muscle actin, desmin, vimentin, HepPar-1, alpha-fetoprotein (α-FP), and polyclonal carcinoembryonic antigen. Ultrastructural examination showed tumor cells with round nucleus containing a single, prominent, complex, nucleolus, copious cytoplasm with few organelles and abundant particulate glycogen, and poorly
Fig. 1. A *Gastroscopic appearance*: examination showed a $2 \times 3$ cm ulcer on the greater curvature with sharp erythematous edges (arrows). B *Histopathological features*: the gastric tumor consisted of confluent large cells with abundant cytoplasm and large rounded nuclei with prominent nucleoli. Mitotic figures were easily identified. An associated, predominantly mononuclear, inflammatory infiltrate was conspicuous throughout the sections. C–F *Immunohistochemical studies* showed expression of C c-kit, D placental alkaline phosphatase (PLAP), and E podoplanin (D2–40) with a membranous pattern, and F focal positivity for pankeratin. G *Electron microscopy* showed primitive tumor cells with abundant cytoplasm with few organelles and abundant particulate glycogen, round nucleus with prominent complex nucleolus, and poorly developed cell junctions. H *The right testicle* showed a hyalinized scar (“burned-out germ cell tumor”; lower left of the photomicrograph) and atrophic seminiferous tubules and hyperplastic Leydig cell nests (upper right). I *Ultrasound examination of the right testicle* demonstrating an ill-defined lesion (arrowheads). J and K *Positron emission tomography* (PET) scan (J) and PET computed tomography (CT) fusion image (K) showing metastases in the pelvic lymph nodes (arrowheads in J). B H&E, ×40; C–F ×40; G ×4000; H H&E, ×5

developed intercellular junctions (Fig. 1G). The morphologic, immunophenotypic, and ultrastructural findings were characteristic of seminoma. The tumor was predominantly mucosal with only focal superficial invasion into the submucosa. Five regional nodes were negative for metastasis and the resection margins were free of tumor by a wide margin.

A diagnostic workup for metastatic seminoma was next undertaken. Testicular ultrasound demonstrated an ill-defined hypodense lesion associated with calcifications in