

Squamous Cell Carcinoma Arising in a Testicular Teratoma and Presenting as Sister Mary Joseph Nodule

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INTRODUCTION

The most common somatic type malignancy arising in patients with testicular germ cell tumors (GCTs) is sarcoma.^[1] Development of carcinomas, especially squamous cell carcinoma is an extremely rare event.^[1,2] Most cases of metastatic umbilical nodules (Sister Mary Joseph nodule) develop from adenocarcinomas.^[3] Fifteen percent of such cases have unknown origin^[4] but development from a testicular squamous cell carcinoma has not yet been reported in the literature. We report a rare case of somatic type squamous cell carcinoma arising in a testicular teratoma. It is also possibly the first reported case of its kind which presented with a metastatic umbilical nodule.

CASE REPORT

A 22-year-old male patient presented with diffuse abdominal pain and anorexia for last two months and a painless umbilical nodule of one-month duration which had recently ulcerated. On examination a solid, firm, nontender, irregular right testicular swelling was palpable. Mild hepatomegaly was also detected. A superficial umbilical nodule measuring 3.9 × 2.3 cm [Figure 1] with surface ulceration was detected. Whole abdominal ultrasonography revealed an 8 × 3 cm heterogeneous space-occupying lesion (SOL) of the right testis with multiple small nodules in the right tunica vaginalis

ABSTRACT

The most common somatic type malignancy arising in patients with testicular germ cell tumors (GCTs) is sarcoma. Development of carcinomas, especially squamous cell carcinoma is an extremely rare event. Most cases of metastatic umbilical nodules (Sister Mary Joseph nodule) develop from adenocarcinomas. Fifteen percent of such cases have unknown origin; but development from a testicular squamous cell carcinoma has not yet been reported in the literature. We report a rare case of somatic type squamous cell carcinoma arising in a testicular teratoma. It is also possibly the first reported case of its kind which presented with a metastatic umbilical nodule. This possibility should be kept in mind while evaluating metastatic umbilical nodules in young male patients.

Key words: Sister Mary Joseph nodule, squamous cell carcinoma, teratoma, testicular germ cell tumors

[Figure 2] and bilateral vaginal hydrocele. Two hypochoic masses measuring 9.4 × 8 cm and 2.8 × 2.5 cm, the former with central cystic degeneration were seen in the right lobe of the liver. A superficial SOL with similar echogenecity was noted at the umbilicus. All the masses appeared neoplastic on imaging and multiple metastases with adhesions were detected in the periaortic region along with mild left hydronephrosis and hydroureter. Moderate elevations of serum beta human chorionic gonadotrophin (HCG) (11.02 mIU/ml) and lactate dehydrogenase (LDH) (683.7 IU/L) levels were found but serum alpha fetoprotein (AFP) level was within normal limits (4.91 ng/ml). Complete blood count showed microcytic hypochromic anemia and raised Erythrocyte Sedimentation Rate (ESR). Chest X-ray revealed no abnormality.

Fine needle aspiration cytology (FNAC) performed from both the umbilical nodule and the hepatic lesions revealed metastatic deposits of squamous cell carcinoma [Figure 3]. Ill-defined granulomas were additionally detected in the umbilical aspirates.

Right-sided high inguinal orchiectomy was done. No inguinal lymph nodes were detected and scrotum was found to be free peroperatively. On cut section two-thirds of the testicular tissue was found to be replaced by a whitish inhomogeneous fleshy mass with similar nodular deposits on the surface.

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Figure 1: Ulcerated umbilical nodule

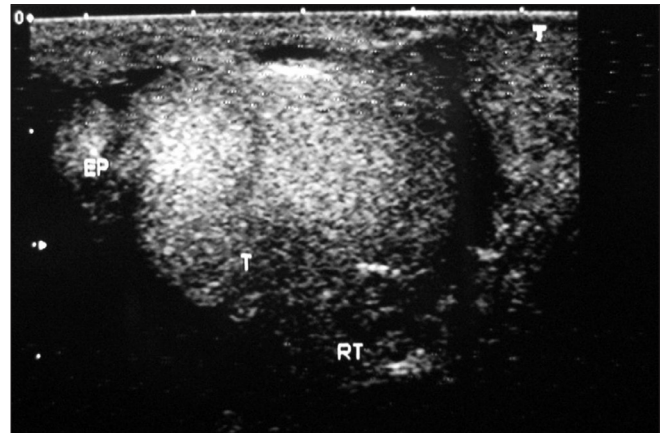


Figure 2: Ultrasonogram of heterogeneous SOL of the right testis with multiple small nodules in the tunica vaginalis and vaginal hydrocele

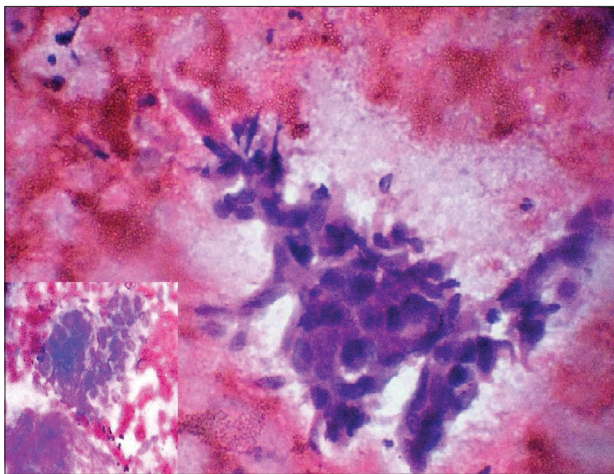


Figure 3: FNAC from hepatic SOL showing metastatic squamous cell carcinoma (Pap; $\times 400$). Inset - FNAC from umbilical nodule showing metastatic deposits of squamous cell carcinoma (Leishman; $\times 400$)

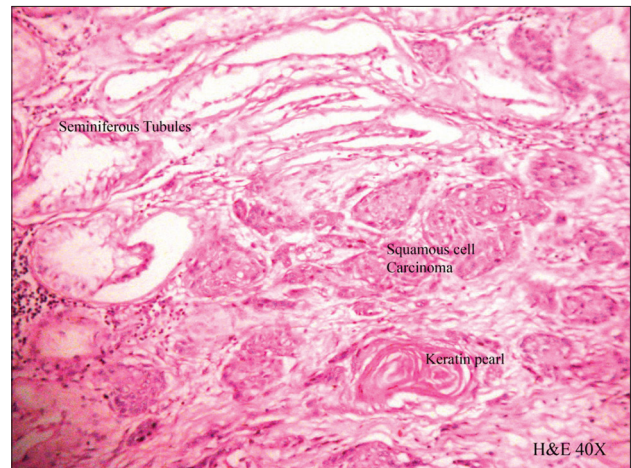


Figure 4: Well-differentiated squamous cell carcinoma invading the testicular tissue; the seminiferous tubules show gross suppression of spermatogenesis (H and E; $\times 100$)

Histopathology revealed well-differentiated (keratinizing) squamous cell carcinoma with plenty of keratin pearl formation invading the testicular tissue [Figure 4]. Elsewhere, foci of teratoma were detected and the existing seminiferous tubules showed gross suppression of spermatogenesis. No IGCNU (intratubular germ cell neoplasia unspecified) or other “*in situ*” lesions were found after careful histopathological examination.

The patient showed no response to three pulses of first-line postoperative chemotherapy with Cisplatin and 5 Fluorouracil (5FU). Institution of second-line chemotherapy was being planned but before that the patient died of disseminated carcinoma and multiorgan failure 25 weeks after initial presentation.

DISCUSSION

Testicular germ cell neoplasia, a disease predominantly

of young men, is, for unknown reasons, increasing in incidence.^[5] The World Health Organization (WHO) has defined teratoma with somatic type malignancy (ICDO code 9084/3) as a teratoma containing a malignant component of a type typically encountered in other organs and tissues, e.g. sarcomas and carcinomas.^[6] Carcinomas are less often associated with GCTs. When uncommon somatic type malignancies (usually squamous cell carcinoma) occur in mature cystic teratomas of the ovary, this is a *de novo* form of malignant transformation; similar tumors in the testis, a very rare event, represent overgrowth of teratomatous elements that originated from malignant, non-teratomatous germ cell tumors and, therefore, had previously undergone malignant transformation.^[2]

Tumors that metastasize to the testis are extremely rare. Among them, the lung is the most common primary site.^[7]

Lymphatic spread from nonseminomatous testicular

tumors is usually localized in retroperitoneal lymph nodes including aortic, common iliac and caval nodes.^[8]

In metastatic sites, the somatic-type malignancies have a poor prognosis. They do not respond to germ cell tumor chemotherapy; surgical resection is the treatment of choice.^[9]

An umbilical metastatic lesion is called 'Sister Mary Joseph's nodule'. It is an uncommon clinical or radiographic finding, and it is rare as the first sign of a malignant disease.^[10] A review of all 407 cases of umbilical metastatic lesion published during the period 1966 to 1997 showed the most common origin to be gastrointestinal (52%).^[4] Even after a diligent search of the published literature not a single reported case was found in which squamous cell carcinoma arising in the testis was detected as the origin of an umbilical metastasis.

How exactly the metastases reach the umbilicus remains largely unknown. Proposed mechanisms for the spread of cancer cells to the umbilicus include direct transperitoneal spread, via the lymphatics which run alongside the obliterated umbilical vein, hematogenous spread, or via remnant structures such as the falciform ligament, median umbilical ligament, or a remnant of the vitelline duct.^[11]

The reported case is extremely rare as somatic type squamous cell carcinoma arises sparingly in a testicular teratoma. Above that, it is also possibly the first reported case of its kind which presented with a metastatic umbilical nodule. This possibility should be kept in mind while evaluating metastatic umbilical nodules in young male patients.

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