Advanced testicular cancer associated with life-threatening tumour lysis syndrome and choriocarcinoma syndrome

Kohei Kobatake, MD; Masao Kato, MD; Koji Mita, MD

Department of Urology, Hiroshima City Asa Hospital, Hiroshima, Japan

Abstract

Tumour lysis syndrome (TLS) and choriocarcinoma syndrome (CS) are severe complications of chemotherapy for testicular cancer. They are rare, but can be life-threatening. A 37-year-old man complaining of persisting cough was referred to our hospital. A computed tomography (CT) scan revealed huge tumours that occupied the peritoneal cavity, with multiple lung, liver, and para-aortic metastases. Although there was no abnormal finding in the testes, serum testicular tumour markers showed marked elevation. A CT-guided biopsy for the peritoneal tumours revealed extragonadal germ cell tumour (GCT), including yolk sac tumour and choriocarcinoma. Chemotherapy with bleomycin, etoposide, and cisplatin (BEP) was started after admission. The morning after the beginning of BEP, the patient developed hemorrhagic shock, in addition to acute pulmonary and renal failure, because of TLS and massive hemorrhage at bilateral lung metastases. He was intubated and resuscitated. Despite appropriate therapy, his renal function did not recover and hemodialysis was started. The patient eventually died of severe respiratory distress syndrome and infection. To our knowledge, this is the first case report of TLS and CS as complications of the hemorrhage at the lung metastases in advanced testicular cancer leading to death.

Introduction

One of the standard chemotherapies for advanced testicular cancer is BEP (bleomycin, etoposide, and cisplatin). This chemotherapy has serious adverse effects, such as choriocarcinoma syndrome (CS) and tumor lysis syndrome (TLS). CS can manifest as hemorrhage at metastatic sites and can cause acute pulmonary disorder when those sites are in the lungs.1,2 TLS can be caused by antineoplastic-drug-sensitive tumours or bulky tumours.

We present the clinical course of an extragonadal germ cell tumour cancer in a patient who died after developing both CS and TLS. To our knowledge, this is the first report of TLS and CS as complications of the hemorrhage at the lung metastases in advanced testicular cancer leading to death.
ed. Operative techniques to stop the bleeding were thought
to be difficult because the respiratory specialist we consulted
thought that there might be multiple bleeding sites in both
lungs. We therefore managed the bleeding conservatively
using styptics. The hemorrhage was marginally controlled,
and the thoracostomy tube was removed on day 7 because
of the decrease of drainage.

The acute renal failure was suspected to be due to both
hypovolemia and TLS, so blood, sodium bicarbonate, and
allopurinol were infused intravenously. The renal failure,
however, kept exacerbating, so hemodialysis was started
on day 4.

Meanwhile, bone marrow suppression was evident on
day 14, and septic shock and disseminated intravascular
coagulation were evident on day 18. Despite administra-
tion of several antibiotics and γ globulin, on day 21 the
patient died of the exacerbation of respiratory failure due
to the severe respiratory infection and respiratory distress
syndrome.

Discussion

We present the clinical course of an advanced metastatic
extragonadal GCT case with life-threatening pulmonary
hemorrhage and TLS. Logothetis has described the hemor-
rhage at the site of metastasis in advanced GCTs with high
volume choriocarcinomatous elements and termed it “cho-
riocarcinoma syndrome” or CS.

Both TLS and CS are rare, but sometimes lethal. While
there are several case reports of either TLS or CS in which the
patient was able to be saved, there is only 1 in which both
the syndromes occurred at the same time (liver and small
intestine hemorrhages associated with hyperuricemia).3

CS is liable to occur in cases with significantly high hCG;
tumour invasion into pulmonary small vessels can cause the
syndrome.4 The mechanism and the frequency of occurrence
is still unclear. Acute pulmonary hemorrhage is the most
frequent manifestation of CS, but hemorrhages due to CS
can develop at any site of metastasis: liver, brain, and small
bowel.3 It may develop immediately after chemotherapy, or
in patients with rapidly progressing disease.2 There are some
reports describing cases in which pulmonary bleeding was
controlled by lobectomy.5

TLS is known as a syndrome developing commonly in
hemodyscrasia, such as leukemia or malignant lymphoma.
Physiolysis of the cancer cells or destruction of cancer tissue
by chemotherapy makes the cancer cells release nucleic
acid, potassium, phosphorus, cytokine, and uric acid as
metabolites of nucleic acid. Furthermore, the release of
cytokines can cause a systemic inflammatory response that
sometimes leads to multiple organ failure.6

After Blonke reported the first case of TLS in 2000,7 there
have been similar reports about TLS in testicular cancer.
These reports included a case associated with acute respira-
tory failure, but none with choriocarcinoma.

Known risk factors for TLS include an LDH level more
than the twice the normal upper limit, intravital total tumour
volume more than 10 cm, and solid cancer with a prolifer-
ating ability and higher sensitivity to treatment.8 Adequate
hydration and the administration of allopurinol and rasburi-
case are recommended, but these preventive measures might
be inadequate; one case describes acute renal failure occur-
ing despite their use.9 Currently, there is also no effective
prevention for CS.

Conclusion

These 2 syndromes are extremely rare and their onset is dif-
cult to predict accurately, but they may lead to treatment-
related death. The risk of these complications should be recognized; in addition, physicians should explain the risk to patients starting chemotherapy for a testicular cancer with significantly high hCG, a huge primary lesion, or metastatic lesions.

Competing interests: Authors declare no competing financial or personal interests.

This paper has been peer-reviewed.

References


Correspondence: Dr. Kohei Kobatake, Department of Urology, Hiroshima City Asa Hospital, 2-1-1, Kabe-minami, Asakita-ku, Hiroshima 731-0293, Japan; koukoba2710@gmail.com