surgeries

MDPI

Case Report

Complex Vascular Reconstruction following Resection of a Large Retroperitoneal Teratoma

Björn-Ole Stüben [1], Asmus Heumann [1], Anastasios D. Giannou [1], Sabine Wipper [2], Peter Bannas [3] and Jun Li [1,*]

1 Department of General, Visceral and Thoracic Surgery, University Medical Center Hamburg-Eppendorf, 20246 Hamburg, Germany; b.stueben@uke.de (B.-O.S.); a.heumann@uke.de (A.H.); a.giannou@uke.de (A.D.G.)
2 Department of Vascular Medicine, University Heart & Vascular Center Hamburg-Eppendorf, 20246 Hamburg, Germany; s.wipper@uke.de
3 Department of Diagnostic and Interventional Radiology and Nuclear Medicine, University Medical Center Hamburg-Eppendorf, 20246 Hamburg, Germany; p.bannas@uke.de
* Correspondence: j.li@uke.de

Abstract: Germ cell tumors (GCTs) are a common malignancy in males, with variations in differentiation making different therapeutic strategies necessary. Generally, GCTs show good curation rates due to their good response to radiotherapy or chemotherapy. However, certain subtypes are resistant to these therapies and require surgery. We present a case of a 25-year-old patient with a large retroperitoneal GCT with somatic malignant transformation, where resection of large abdominal blood vessels with complex reconstruction was necessary to completely remove the tumor. The tumor was completely resected, and the patient has since been recurrence-free in the follow-up period. GCTs with somatic transformation show high resistance rates to chemo- and radiotherapy, and the patient in the presented case study did indeed show only a limited response to carboplatin-based chemotherapy. Patients suffering from these conditions should be resected whenever possible, as curation can be achieved by complete tumor resection. Infiltration of neighboring structures is no contraindication to surgery. The case presented here shows that interdisciplinary surgical planning including vascular and general surgeons as well as radiologists is vital to ensure successful tumor resection.

Keywords: germ cell tumor; malignant somatic differentiation; complex vascular reconstruction

Citation: Stüben, B.-O.; Heumann, A.; Giannou, A.D.; Wipper, S.; Bannas, P.; Li, J. Complex Vascular Reconstruction following Resection of a Large Retroperitoneal Teratoma. *Surgeries* **2021**, *2*, 139–143. https://doi.org/10.3390/surgeries2020013

Academic Editor: Paul Quax

Received: 7 March 2021
Accepted: 25 March 2021
Published: 29 March 2021

1. Introduction

Germ cell tumors (GCTs) are the most common solid malignancy in the third and fourth decade in males, with the gonads being the most common localization [1]. The incidence of these diseases has doubled over the last 25 years [2–4].

Extragonadal manifestations are rare, with the mediastinum being the most common site closely followed by the retroperitoneum [5].

GCTs show great variation in respect to differentiation, and these variations are important when considering response to therapy. Some tissues are radiosensitive, whilst others respond well to chemotherapy. In general, GCTs show good therapeutic response even when discovered in patients with existing metastases, and cure rates of up to 80% can be achieved [6].

Of these histological subtypes, somatic malignant transformation (SMT) is extremely rare, making up 2% of all male GCTs [7]. These tumors can histologically resemble any of the three germinal layers, sarcomas being the most common subtype [8,9]. These tumors have been shown to be resistant to traditional cisplatin-based chemotherapy regimens, and therefore present a therapeutic challenge. Aggressive surgical resection is currently considered the therapy of choice with curation rates of 50–60% [8–10].

2. Case Description

A 25-year-old male patient had been admitted to a regional hospital due to lower abdominal pain.

Contrast enhanced computed tomography (CT) scan of the abdomen showed a tumor mass of the left psoas major muscle. Subsequently, magnetic resonance imaging (MRI) demonstrated extensive tumor growth in the retroperitoneal space with a tumor size of >10 × 10 cm^2 in diameter with concomitant obstructive uropathy grade III on the left side (Figure 1).

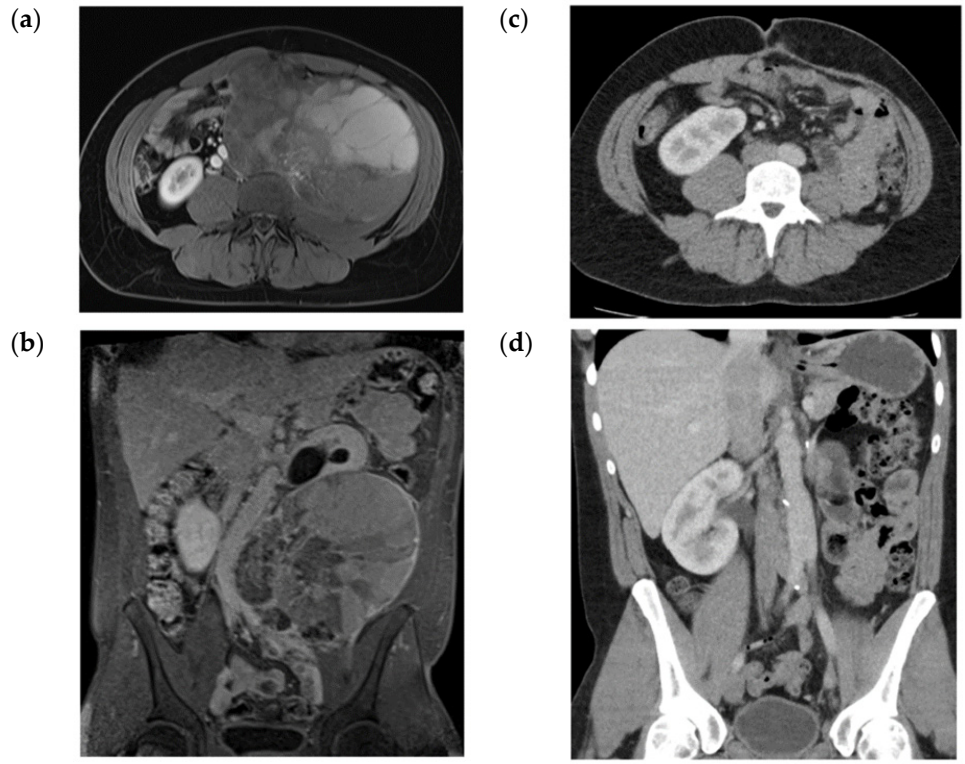


Figure 1. (**a**). Axial contrast-enhanced MRI, with tumor lesion in the left retroperitoneum; (**b**). Coronary MRI showing extent of the tumor with left sided uropathy as well as compression of the aorta and the iliac bifurcation (**c**,**d**). Postoperative axial and coronary contrast-enhanced CT.

The patient was referred to our university medical center for further diagnostics and treatment.

During admission, the patient was in good health with a normal BMI. Detailed anamnesis of the patient revealed a weight loss of 3 kg in the previous two months. He also complained of increased perspiration whilst sleeping and fatigue as well as lower abdominal pain.

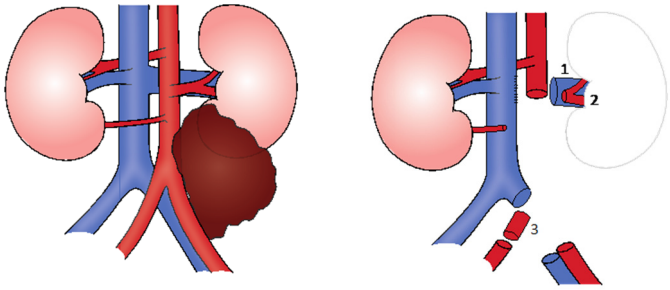
A CT guided biopsy of the tumor mass was performed, the histopathological examination showing a somatic type teratoma. The tumor markers CEA, CA 19-9, β-HCG and AFP were negative.

The multidisciplinary tumor board (MDT) decided on neoadjuvant therapy with carboplatin AUC2 and etoposid followed by three x ifosfamide over a period of three months.

The re-staging (FDG-PET/CT) showed only minimal response to therapy and revealed in addition thrombosis of the left common and internal iliac vein. It became apparent that the tumor had extensively infiltrated the retroperitoneum as well as the left ureter, the left common iliac artery, the left iliac vein and the infrarenal aorta.

Therefore, the MDT decided for surgery, leading to an interdisciplinary concept involving the departments of vascular surgery, transplant surgery and general surgery.

After dissection of the retroperitoneum, the infrarenal aorta as well as both iliacal vessels were crossclamped following intravenous administration of 5000 IU Heparin. The tumor was resected en bloc with the infrarenal aorta, both common iliac arteries, left iliac vein, left kidney and left ureter (Figure 2).



(**a**) (**b**)

Figure 2. (**a**) Tumor involving the left ureter, infrarenal aorta, the iliac bifurcation, the left iliac artery and left iliac vein; (**b**) The tumor was completely resected with vessels harvested for further reconstruction (1. left renal vein; 2. left renal artery including two segment arteries; 3. right common iliac artery).

In addition, the mesosigmoid, which also showed tumor infiltration, was resected along with the inferior mesenteric artery. An intraoperative florescence examination confirmed good perfusion of the left and sigmoid colon, thus avoiding a segmental colon resection.

Subsequently, the infrarenal aorta was replaced by a collagen-, silver-, and triclosan covered bifurcated polyester graft (Intergard Synergy 18/9 mm). The left common iliac vein was reconstructed using the previously harvested right common iliac artery and the left renal vein, which had been resected simultaneously during the nephrectomy. The left renal artery was used to connect the lower pole artery of the right kidney to the aortic graft (Figure 3).

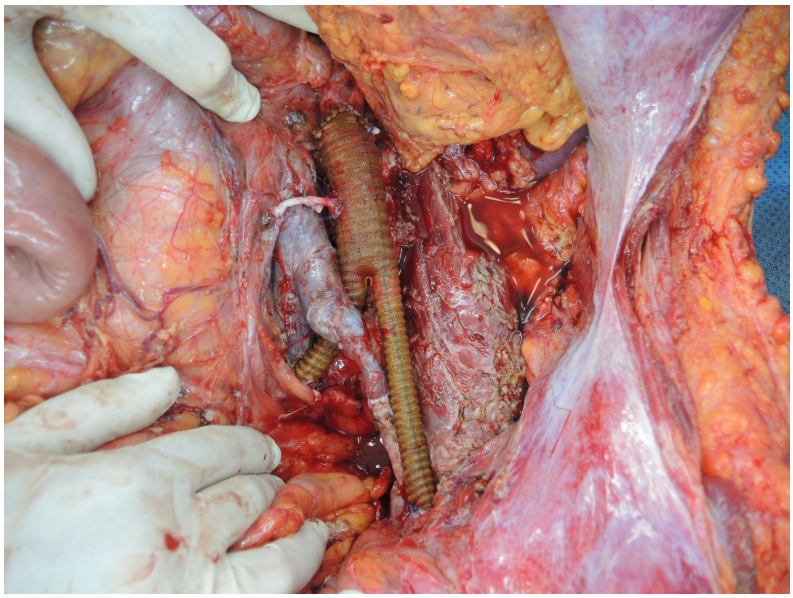
Macroscopic histopathological analyses showed a tumor 10.2 cm in diameter resected en bloc with the infrarenal aorta, the left ureter and Gerota fat, internal iliac artery and vein as well as the psoas muscle. Microscopic analysis revealed a sarcomatous and carcinomatous differentiation. The tumor was completely resected (R0), and was classified as a stage IIIB, intermediate risk tumor.

The postoperative course was uneventful with the patient being discharged after seven days. No edema of the left leg was observed. The renal function was well compensated with serum creatinine in the normal range. Ultrasound exams revealed normal arterial and venous blood flow under therapeutic anticoagulation with low-molecular heparin (enoxaparin). The enoxaparin was given for six months to prevent venous thrombosis after complex iliac vein reconstruction.

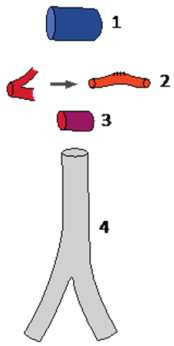
Follow-up after one year (thoracoabdominal CT) revealed a normal postoperative status without tumor recurrence. All blood vessels were circulated.

Whilst GCTs generally have a good prognosis with curation rates of 80% being achievable, somatic differentiation severely impacts prognosis, with significantly lower survival rates of 50–60% following resection and systemic therapy [6,8–10]. This is largely due to chemoresistance seen in somatically differentiated tumors. Rice et al. demonstrated that patients with somatic differentiation in GCT had a complete response following cisplatin-based chemotherapy of 13% compared to the expected 70% which would be seen in metastatic GCT without somatic differentiation [11]. The patient in the presented case received neoadjuvant therapy followed by surgery due to the extensive tumor growth, with the aim of allowing for a limited resection by tumor reduction under chemotherapy. Without obvious evidence of response to chemotherapy, surgery was performed in time to reach a R0 resection without significant delay. The limited response to the neoadjuvant

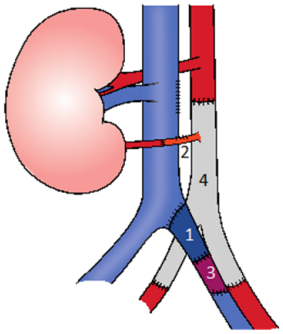
therapy in this case is in line with the available current literature which describes high rates of chemoresistance of these tumors. Similar to patients with soft tissue sarcomas, surgical radical resection is the main stay to reach an acceptable survival chance for patients with GCT and somatic differentiation. The current case involved meticulous vascular reconstruction including aorta, iliaca vein and renal artery reconstruction. Wortmann et al. described two-year patency rates of arterial bypasses following surgery of soft tissue sarcomas in the retroperitoneum of 88% [12]. Reconstruction of the iliaca vein by autologous graft reduced the risk of postoperative lower extremity edema that led to an early discharge in this case. Preoperative design to harvest left renal vein and right iliaca artery for the venous reconstruction played an important role. The transplant experience made the important intraoperative decision to reconstruct the right lower polar renal artery by the left renal artery conduit which avoided the ureter necrosis and impairment of the renal function.



(**a**)



(**b**)



(**c**)

Figure 3. (**a**). vascular reconstruction following tumor resection: 18/9 Synergy Y-Prosthesis, infrarenal aorta; 2: V. cava inferior; 3: right common iliac vein and reconstructed right common iliac artery; 4: reconstructed left common iliac artery; 5: reconstructed left common iliac vein; (**b**). Overview of the vascular grafts for the venous and arterial reconstruction (1: left renal vein; 2. left renal artery; 3. Right common iliac artery; 4. 18/9 Synergy Y-prosthesis); (**c**). Complete vascular reconstruction of the infrarenal aorta including iliac bifurcation, the right lower pole renal artery and the left common iliac vein.

3. Conclusions

GCTs with somatic transformation show high resistance rates to chemo- and radiotherapy, and the patient in the presented case study did indeed show only limited response to carboplatin-based chemotherapy.

Patients suffering from these conditions should be resected whenever possible, as curation can be achieved by complete tumor resection. Infiltration of neighboring structures is no contraindication to surgery. The case presented here shows that interdisciplinary surgical planning including vascular, transplant and general surgeons as well as radiologists is vital to ensure successful tumor resection.

Author Contributions: B.-O.S. and J.L. wrote the manuscript. J.L., A.H. and S.W. were the operating surgeons. P.B. was the radiologist responsible for the preoperative radiological evaluation. A.D.G. critically revised the manuscript. All authors have read and agreed to the published version of the manuscript.

Funding: This research received no external funding.

Institutional Review Board Statement: Ethical review and approval were waived for this case report, due to the nature of this article (case report).

Informed Consent Statement: Informed consent was obtained from the patient involved in the study.

Data Availability Statement: All data respecting the anonymity of patients are available on request.

Acknowledgments: A special thank you goes to Sabine Wuttke of the graphics department of the University Medical Center Hamburg-Eppendorf for the drawings shown in Figure 3b,c.

Conflicts of Interest: The authors declare no conflict of interest.

References

1. Hayes-Lattin, B.; Nichols, C.R. Testicular cancer: A prototypic tumor of young adults. *Semin. Oncol.* **2009**, *36*, 432–438. [CrossRef] [PubMed]
2. Carver, B.S.; Sheinfeld, J. Germ cell tumors of the testis. *Ann. Surg. Oncol.* **2005**, *12*, 871–880. [CrossRef] [PubMed]
3. McKiernan, J.M.; Goluboff, E.T.; Liberson, G.L.; Golden, R.; Fisch, H. Rising risk of testicular cancer by birth cohort in the United States from 1973 to 1995. *J. Urol.* **1999**, *162*, 361–363. [CrossRef]
4. Purdue, M.P.; Devesa, S.S.; Sigurdson, A.J.; McGlynn, K.A. International patterns and trends in testis cancer incidence. *Int. J. Cancer* **2005**, *115*, 822–827. [CrossRef] [PubMed]
5. Arora, R.S.; Alston, R.D.; Eden, T.O.; Geraci, M.; Birch, J.M. Comparative incidence patterns and trends of gonadal and extragonadal germ cell tumors in England, 1979 to 2003. *Cancer* **2012**, *118*, 4290–4297. [CrossRef] [PubMed]
6. Carver, B.S.; Serio, A.M.; Bajorin, D.; Motzer, R.J.; Stasi, J.; Bosl, G.J.; Vickers, A.J.; Sheinfeld, J. Improved clinical outcome in recent years for men with metastatic nonseminomatous germ cell tumors. *J. Clin. Oncol.* **2007**, *25*, 5603–5608. [CrossRef] [PubMed]
7. Ahmed, T.; Bosl, G.J.; Hajdu, S.I. Teratoma with malignant transformation in germ cell tumors in men. *Cancer* **1985**, *56*, 860–863. [CrossRef]
8. Comiter, C.V.; Kibel, A.S.; Richie, J.P.; Nucci, M.R.; Renshaw, A.A. Prognostic features of teratomas with malignant transformation: A clinicopathological study of 21 cases. *J. Urol.* **1998**, *159*, 859–863. [CrossRef]
9. Motzer, R.J.; Amsterdam, A.; Prieto, V.; Sheinfeld, J.; Murty, V.V.; Mazumdar, M.; Bosl, G.J.; Chaganti, R.S.K.; Reuter, V.E. Teratoma with malignant transformation: Diverse malignant histologies arising in men with germ cell tumors. *J. Urol.* **1998**, *159*, 133–138. [CrossRef]
10. Little, J.S., Jr.; Foster, R.S.; Ulbright, T.M.; Donohue, J.P. Unusual neoplasms detected in testis cancer patients undergoing post-chemotherapy retroperitoneal lymphadenectomy. *J. Urol.* **1994**, *152*, 1144–1149. [CrossRef]
11. Rice, K.R.; Magers, M.J.; Beck, S.D.; Cary, K.C.; Einhorn, L.H.; Ulbright, T.M.; Foster, R.S. Management of germ cell tumors with somatic type malignancy: Pathological features, prognostic factors and survival outcomes. *J. Urol.* **2014**, *192*, 1403–1409. [CrossRef] [PubMed]
12. Wortmann, M.; Alldinger, I.; Bockler, D.; Ulrich, A.; Hyhlik-Durr, A. Vascular reconstruction after retroperitoneal and lower extremity sarcoma resection. *Eur. J. Surg. Oncol.* **2017**, *43*, 407–415. [CrossRef] [PubMed]