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Splenic lymphoma with complex gastro-spleno-diaphragmatic fistula: 3D laparoscopic multivisceral resection. The first literature case report

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ABSTRACT

INTRODUCTION: Gastrosplenic fistula is a rare disease involving stomach and spleen that can lead to dangerous complications like massive gastrointestinal bleeding. Diffuse large B-cell lymphoma (DLBC) is the principal pathological cause of gastrosplenic fistula.

CASE REPORT: We report a case of A 76-year-old caucasian woman came to the emergency room with fever for two week and gravative pain in left upper quadrant of the abdomen. CT scan of thorax and abdomen demonstrated an inhomogeneous hypodense large lymphomatous mass (10 × 6 cm) of upper pole of the spleen deforming medial profile and infiltrating gastric fundus and left diaphragm. with the diagnosis of complex gastro-spleno-diaphragmatic fistula we performed an en-block resection using a 3D laparoscopic vision system.

DISCUSSION: Gastrosplenic fistula is a rare complication of several clinical conditions. Among the different causes diffuse large B-cell lymphoma is the most frequent although diffuse histiocytic lymphoma, Hodgkin's lymphoma and extranodal NK/T-cell lymphoma are also described. After a literature review we found less then 30 cases of gastrosplenic fistula secondary to lymphoma. In our case report we do the first description of three-organs fistula involvement, stomach, spleen and diaphragmatic dome, managed with 3D laparoscopic approach.

CONCLUSION: Gastrosplenic fistula can represent a fatal evolution of splenic or gastric lymphoma independently from chemotherapy treatment. The diagnosis of this condition is very difficult and related to its rarity. To our opinion, laparoscopy represents a valid and safe alternative to open surgery in management of these patients.

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1. Introduction

Gastrosplenic fistula is a rare disease involving stomach and spleen that can lead to dangerous complications like massive gastrointestinal bleeding. Diffuse large B-cell lymphoma (DLBC) is the principal pathological cause of gastrosplenic fistula, but other several diseases can be responsible such as advanced gastric cancer, splenic abscess, crohn's disease and trauma [1]. Transverse/splenic flexure colon cancer locally advanced can induce formation of splenocolic fistula. To our knowledge this is the first description of a complex fistula involving three different organs, spleen, stomach and diaphragm, caused by DLBC lymphoma and managed by

3D laparoscopic multivisceral resection. It is reported in line with the SCARE criteria [2,3].

2. Case report

A 76-year-old caucasian woman came to the emergency room with fever for two week and gravative pain in left upper quadrant of the abdomen associated to weight loss (about 5 kg in 3 months) and asthenia. She referred also recurrent episodes of melena. On physical examination her abdomen was soft with a palpable, not painful large mass in left hypochondrium suggestive for splenomegaly. Laboratory exams showed a chronic microcitic anemia (Hb 6,8 g/dl, MCV 68 fl). We performed a contrast enhanced CT scan of thorax and abdomen that demonstrated an inhomogeneous hypodense large lymphomatous mass (10 × 6 cm) of upper pole of the spleen deforming medial profile and infiltrating gastric fundus and left diaphragm (Fig. 1). In consideration of Hb levels and referred melena we performed preoperative endoscopic exploration that confirmed gastric infiltration. We found also a raised area with central ulceration and signs of recent bleeding

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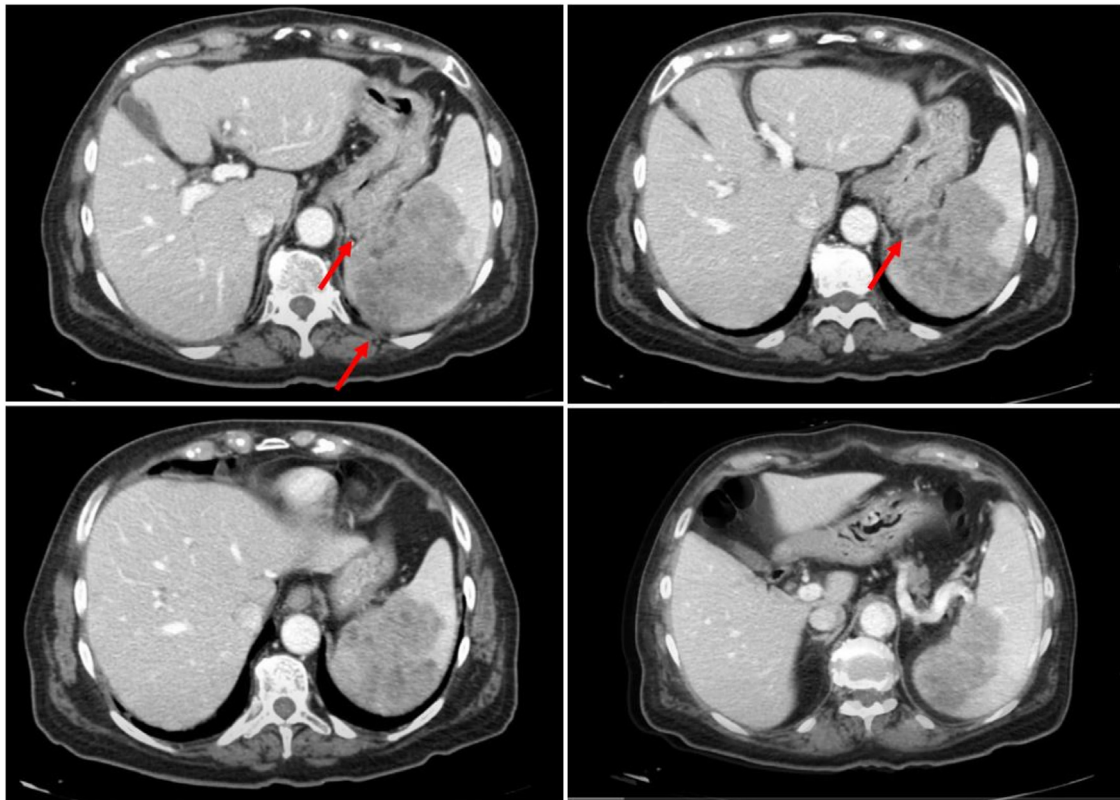


Fig. 1. CT scan of the thorax and abdomen showed an inhomogeneous hypodense large lymphomatous mass (10 × 6 cm) of upper pole of the spleen deforming medial profile and infiltrating gastric fundus and left diaphragm (arrows).

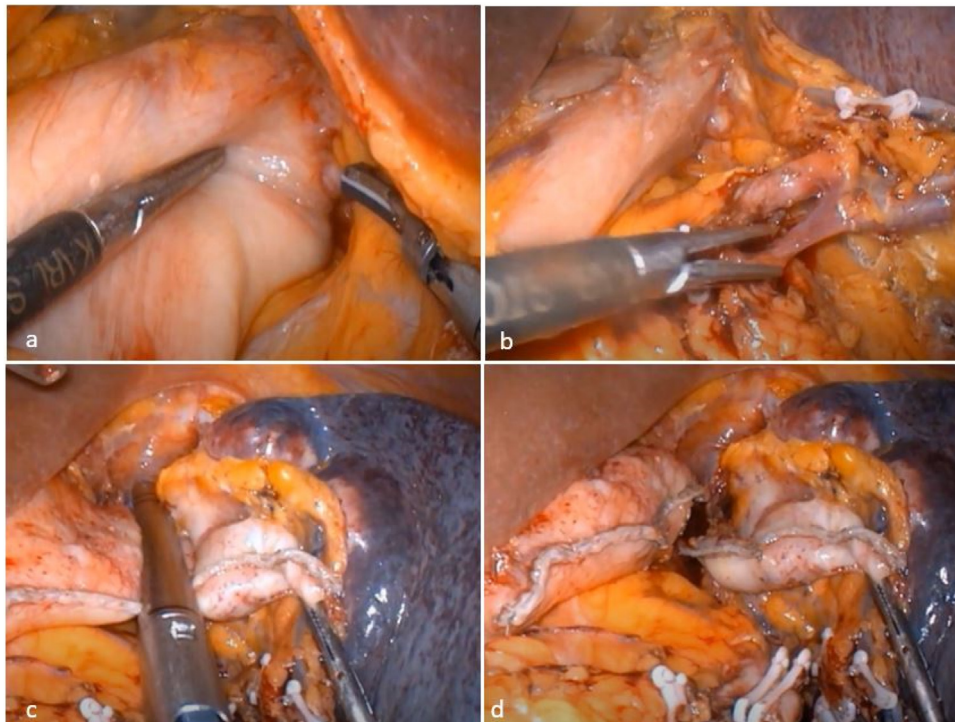


Fig. 2. a) laparoscopic exploration with evidence of gastrosplenic fistula; b) dissection of hilum of the spleen with identification of splenic artery and vein; c, d) partial sleeve gastrectomy with Echelon Flex™ 45 mm blue reload.

strongly suggestive for gastrosplenic fistula. On the basis of site and radiological features of the lesion and considering the potential fatal evolution of a gastro-splenic-diaphragmatic fistula and the absence of others nodal and extranodal localizations our multidisci-

plinary tumor board opted for surgical multivisceral resection with laparoscopic approach [4]. The patient was placed in right lateral decubitus on operating table in order to obtain the best exposition of hilum of the spleen and to prevent significative bleeding

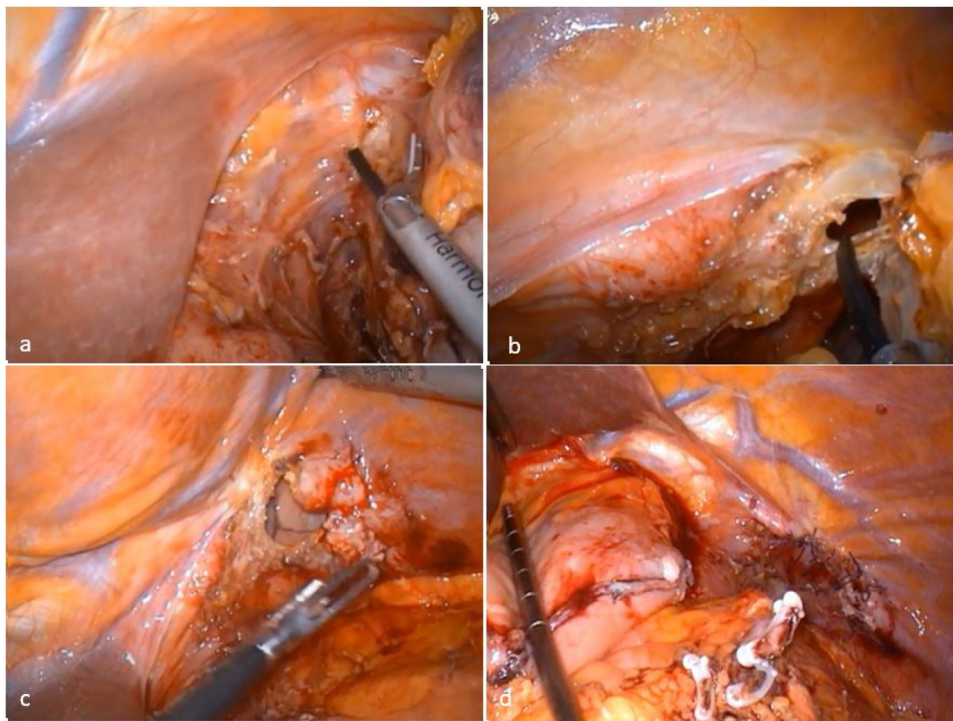


Fig. 3. a) diaphragmatic invasion originating from upper pole of the spleen; b) in-block resection of part of diaphragmatic dome; c) evidence of no iatrogenic lesion of pulmonary parenchyma; d) surgical field at the end of laparoscopic procedure with resected gastric fundus and sutured diaphragmatic dome.

[5,6]. We induced pneumoperitoneum with Veress needle and put four trocars in left subcostal region [7]. We used a 3D laparoscopic camera with the aim to improve dissection and facilitate surgical maneuvers [8,9]. The procedure began with detachment of splenocolic ligament and medial access to the hilum of the spleen (Fig. 2). We identified, clipped and divided splenic artery and vein. During dissection of the upper pole of the spleen we detected the gastrosplenic fistula and carried out a partial sleeve gastrectomy with Echelon Flex™ 45 mm (blue reload, Ethicon Endo Surgery INC - Johnson & Johnson, NJ, USA) With surgical dissection from down-to-up we reached diaphragmatic dome that was infiltrated from splenic mass. We performed an in block multivisceral resection of left diaphragmatic dome with no iatrogenic lesion of pulmonary parenchyma (Fig. 3). At the end of procedure we proceeded with intracorporeal suturing of diaphragm and we left only a drain in splenic loggia [10]. We used a suprapubic caesarean section for extraction of surgical specimen positioned in endo-bag. The post-operative course was uneventful with no pleural nor abdominal collection. Food intake resumed on 2nd POD and patient was discharged on POD 5. On the last follow-up control, 5 years later, the patient was in good general clinical condition and with no clinical and radiological evidence of recurrence. Histopathological and immunophenotypic examination (ki67 = 85%, CD20+, CD10+, CD3-, bcl6-, CD30-) revealed a germinal centre type diffuse large B-cell lymphoma.

3. Discussion

Gastrosplenic fistula is a rare complication of several clinical conditions. Among the different causes diffuse large B-cell lymphoma is the most frequent although diffuse histiocytic lymphoma, Hodgkin's lymphoma and extranodal NK/T-cell lymphoma are also described. After a literature review we found less than 30 cases of gastrosplenic fistula secondary to lymphoma. In our case report we do the first description of three-organs fistula

involvement, stomach, spleen and diaphragmatic dome, managed with 3D laparoscopic approach. Only Al-Ashgar HI et al. [11] in 2007 described a conservative laparoscopic treatment with closure of gastrosplenic fistula and repair of gastric wall. The authors stressed the opportunity of miniinvasive conservative approach as the bridge to definitive treatment with chemotherapy and radiotherapy. In 2017 Kang DH et al. [12] described the first gastrosplenic fistula occurred in patient with NK/T-cell lymphoma and did an extensive literature search with 26 patients. In 88,9% of cases the patients underwent to visceral resection because gastrosplenic fistula can occur also after chemotherapy and in all patients represents a potential fatal evolution of disease. The pathophysiology of gastrosplenic fistula take into consideration tissue necrosis due to lymphoma with involvement of splenic capsule and gastric wall. The chronic process lead to fistula formation [13]. In our clinical case the patient came to our observation with acute presentation (recurrent melena and anemia) and CT scan showed splenic lymphoma with infiltration of gastric wall and diaphragmatic dome. This presentation has potential fatal evolution with massive gastric bleeding and pleural effusion. CT scan of thorax and abdomen is fundamental for diagnosis of splenic lymphoma and to detect gastrosplenic fistula and, in generally, for study of parenchymatous organs of this anatomical region [14,15]. On the contrary the identification of diaphragmatic lesions represents a challenge for radiologist. Multifasic spiral CT increased the accuracy (sensitivity: 80%; specificity: 90%) [16]. Magnetic resonance imaging (MRI) when feasible is the best technique to identify lesions of diaphragmatic dome [17]. We decided to carry out a laparoscopic multivisceral resection for good outcomes of these patients and to prevent hemorrhagic complications. The use of a laparoscopic 3D vision system allowed us advanced performances in a complex surgical field with small and deep spaces (splenic loggia occupied by a large spleen) and promoted achieving of arduous surgical laparoscopic procedures as sutures and intracorporeal knotting for diaphragmatic repair [18–20].

4. Conclusion

Gastrosplenic fistula can represent a fatal evolution of splenic or gastric lymphoma independently from chemotherapy treatment. The diagnosis of this condition is very difficult and related to its rarity. This is the first literature report of three-organ fistula due to a splenic lymphoma with radical 3D laparoscopic treatment. To our opinion, laparoscopy performed by experienced team in oncological and emergency laparoscopic surgery [21–23] represents a valid and safe alternative to open surgery in management of these patients with the main advantages of miniinvasive technique and excellent surgical and functional outcomes. The use of the 3D visual system also facilitates surgical maneuvers and reduces operative time, further improving the results obtained with traditional laparoscopy.

Declaration of Competing Interest

The authors report no declarations of interest.

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Ethical approval

Ethical Approval was not necessary for this study. We obtained written patient consent to publication.

Consent

We obtained written patient consent to publication.

Author's contribution

Di Buono Giuseppe: study design, data collections, data analysis and writing.

Buscemi Salvatore: study design.

Maienza Elisa: data collections.

Bonventre Giulia: data collections.

Romano Giorgio: study design, data collections, data analysis and writing.

Agrusa Antonino: study design, data collections, data analysis and writing.

Registration of research studies

Not applicable.

Guarantor

Di Buono Giuseppe.

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References

- [1] J.E. Choi, H.J. Chung, H.G. Lee, Spontaneous gastrosplenic fistula: a rare complication of splenic diffuse large cell lymphoma, *Abdom. Imaging* 27 (2002) 728–730.
- [2] R.A. Agha, A.J. Fowler, A. Saetta, I. Barai, S. Rajmohan, D.P. Orgill, for the SCARE Group, The SCARE statement: consensus-based surgical case report guidelines, *Int. J. Surg.* 34 (2016) 180–186.
- [3] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, The SCARE 2018 statement: updating consensus Surgical Case Report (SCARE) guidelines, *Int. J. Surg.* 60 (2018) 132–136.
- [4] A. Agrusa, G. Di Buono, S. Buscemi, B. Randisi, L. Gulotta, V. Sorce, G. Badalamenti, D. Albano, M. Galia, G. Romano, G. Gulotta, Dedifferentiated retroperitoneal large liposarcoma and laparoscopic treatment: is it possible and safe? The first literature case report, *Int. J. Surg. Case Rep.* 57 (2019) 113–117, <http://dx.doi.org/10.1016/j.ijscr.2019.03.023>, Epub 2019 Mar 26.
- [5] G. Di Buono, S. Buscemi, A.I. Lo Monte, G. Geraci, V. Sorce, R. Citarrella, E. Gulotta, V.D. Palumbo, S. Fazzotta, L. Gulotta, D. Albano, M. Galia, G. Romano, A. Agrusa, Laparoscopic adrenalectomy: preoperative data, surgical technique and clinical outcomes, *BMC Surg.* 18 (Suppl. 1) (2019) 128, <http://dx.doi.org/10.1186/s12893-018-0456-6>, Apr 24.
- [6] G. Romano, A. Agrusa, D. Chianetta, G. Frazzetta, V. Sorce, G. Di Buono, G. Gulotta, Laparoscopic management of adrenal tumors: a four-years experience in a single center, *Minerva Chir.* 69 (2014) 125–129.
- [7] A. Agrusa, G. Romano, G. De Vita, G. Frazzetta, D. Chianetta, G. Di Buono, G. Gulotta, Adrenal gunshot wound: laparoscopic approach. Report of a case, *Int. J. Surg. Case Rep.* 5 (2014) 70–72.
- [8] A. Agrusa, G. Romano, L.J. Dominguez, G. Amato, R. Citarrella, L. Vernuccio, G. Di Buono, V. Sorce, L. Gulotta, M. Galia, P. Mansueto, G. Gulotta, Adrenal cavernous hemangioma: which correct decision making process? *Acta Medica Mediterranea* 32 (2016) 385–389.
- [9] A. Agrusa, G. Di Buono, D. Chianetta, V. Sorce, R. Citarrella, M. Galia, L. Vernuccio, G. Romano, G. Gulotta, Three-dimensional (3D) versus two-dimensional (2D) laparoscopic adrenalectomy: a case-control study, *Int. J. Surg.* 28 (Suppl. 1) (2016) S114–7, <http://dx.doi.org/10.1016/j.ijso.2015.12.055>.
- [10] A. Agrusa, G. Romano, D. Chianetta, G. De Vita, G. Frazzetta, G. Di Buono, V. Sorce, G. Gulotta, Right diaphragmatic injury and lacerated liver during a penetrating abdominal trauma: case report and brief literature review, *World J. Emerg. Surg.* 9 (2014) 33, <http://dx.doi.org/10.1186/1749-7922-9-33>, Apr 28, eCollection 2014. Review.
- [11] H.I. Al-Ashgar, M.Q. Khan, A.M. Ghamdi, F.Y. Bamehriz, I. Maghfoor, Gastrosplenic fistula in Hodgkin's lymphoma treated successfully by laparoscopic surgery and chemotherapy, *Saudi Med. J.* 28 (2007) 1898–1900.
- [12] D.H. Kang, J. Huh, J.H. Lee, Y.K. Jeong, H.J. Cha, Gastrosplenic fistula occurring in lymphoma patients: systematic review with a new case of extranodal NK/T-cell lymphoma, *World J. Gastroenterol.* 23 (35) (2017) 6491–6499, <http://dx.doi.org/10.3748/wjg.v23.i35.6491>.
- [13] J. Senapati, Aj Devasia, S. Sudhakar, A. Viswabandya, Asymptomatic gastrosplenic fistula in a patient with marginal zonal lymphoma transformed to diffuse large B cell lymphoma—a case report and review of literature, *Ann. Hematol.* 93 (9) (2014) 1599–1602, <http://dx.doi.org/10.1007/s00277-013-1986-8>.
- [14] M. Galia, D. Albano, A. Bruno, A. Agrusa, G. Romano, G. Di Buono, F. Agnello, G. Salvaggio, L. La Grutta, M. Midiri, R. Lagalla, Imaging features of solid renal masses, *Br. J. Radiol.* 90 (1077) (2017), 20170077, <http://dx.doi.org/10.1259/bjr.20170077>, Aug, Epub 2017 Jul 13. Review.
- [15] D. Albano, F. Agnello, F. Midiri, G. Pecoraro, A. Bruno, P. Alongi, P. Toia, G. Di Buono, A. Agrusa, L.M. Sconfienza, S. Pardo, L. La Grutta, M. Midiri, M. Galia, Imaging features of adrenal masses, *Insights Imaging* 10 (1) (2019) 1, <http://dx.doi.org/10.1186/s13244-019-0688-8>, Jan 25.
- [16] D.M. Stein, G.B. York, S. Boswell, K. Shanmuganathan, M. Haan, T.M. Scalea, Accuracy of computed tomography scan in the detection of penetrating diaphragm injury, *J. Trauma* 63 (3) (2007) 538–543.
- [17] K. Sanmuganathan, S.E. Mirvis, C.S. White, S.M. Pomerantz, MR imagining evaluation of hemidiaphragms in acute blunt trauma: experience with 16 patients, *AJR* 167 (1996) 397–402.
- [18] A. Agrusa, G. Di Buono, S. Buscemi, G. Cucinella, G. Romano, G. Gulotta, 3D laparoscopic surgery: a prospective clinical trial, *Oncotarget* 9 (25) (2018) 17325–17333, <http://dx.doi.org/10.18632/oncotarget.24669>, Apr 3, eCollection 2018 Apr 3.
- [19] A. Agrusa, G. Romano, G. Navarra, G. Conzo, G. Pantuso, G.D. Buono, R. Citarrella, M. Galia, A.L. Monte, G. Cucinella, G. Gulotta, Innovation in endocrine surgery: robotic versus laparoscopic adrenalectomy. Meta-analysis and systematic literature review, *Oncotarget* 8 (60) (2017) 102392–102400,

- <http://dx.doi.org/10.18632/oncotarget.22059>, Oct 19, eCollection 2017 Nov 24.
- [20] G. Cucinella, G. Calagna, G. Romano, G. Di Buono, G. Gugliotta, S. Saitta, G. Adile, M. Manzone, G. Accardi, A. Perino, A. Agrusa, Robotic versus laparoscopic sacrocolpopexy for apical prolapse: a case-control study, *G Chir.* 37 (3) (2016) 113–117, May-Jun.
- [21] A. Agrusa, G. Frazzetta, D. Chianetta, S. Di Giovanni, L. Gulotta, G. Di Buno, V. Sorce, G. Romano, G. Gulotta, Relaparoscopic management of surgical complications: the experience of an Emergency Center, *Surg. Endosc.* 30 (2016) 2804–2810.
- [22] A. Agrusa, G. Romano, G. Di Buono, G. Frazzetta, D. Chianetta, V. Sorce, V. Billone, G. Cucinella, G. Gulotta, Acute appendicitis and endometriosis: retrospective analysis in emergency setting, *GLOG* 35 (2013) 728–732.
- [23] G. Romano, A. Agrusa, M. Galia, G. Di Buono, D. Chianetta, V. Sorce, L. Gulotta, G. Brancatelli, G. Gulotta, Whipple's pancreaticoduodenectomy: surgical technique and perioperative clinical outcomes in a single center, *Int. J. Surg.* 21 (Suppl. 1) (2015) S68–71, <http://dx.doi.org/10.1016/j.ijssu.2015.06.062>, Sep, Epub 2015 Jun 26.

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