

## Case Report

## Open Access

Paolo Laperuta, Filomena Napolitano, Rosa Maria Di Crescenzo, Pio Zeppa, Antonio Galderisi, Carmine Selleri, Alessandro Vatrella, Mario Capunzo, Vincenzo Giuseppe Di Crescenzo\*

# Idiopathic pleural panniculitis with recurrent pleural effusion not associated with Weber-Christian disease

DOI 10.1515/med-2016-0071

received September 16, 2016; accepted September 19, 2016

**Keywords:** Panniculitis; Thoracoscopy; Exploratory excision; Weber-Christian disease

**Abstract:** A 82-year-old patient with dyspnea and a recurrent history of pleural effusion was admitted into our unit. He performed a Chest computed tomography showing right pleural effusion. Video-assisted thoracoscopy (VATS) exploratory showed parietal pleural thickening of adipose tissue. The surgical procedure consisted, therefore, in the execution of multiple biopsies of the parietal pleura which appeared covered, on the whole surface, by islands of adipose tissue, without macroscopic pathological aspects. After the procedure was performed pleurodesis with talc. The definitive histological examination consisted of normal mesothelial cells surrounded by fatty tissue infiltrated by small lymphocytes in a patient without skin lesions or visceral or systemic signs of inflammatory involvement of the adipose tissue. We reported a rare case of idiopathic pleural panniculitis with recurrent pleural effusion not associated with Weber-Christian disease.

## 1 Introduction

Idiopathic panniculitis is a systemic inflammatory non suppurative disease of adipose tissue of unknown aetiology [1-4]. This clinical evidence is often observed in Weber-Christian disease (WCD) associated with cutaneous lesions (recurrent subcutaneous inflammatory painful nodules) and systemic manifestations (fever, malaise and, sometimes, flogistic involvement of the lungs, heart, gastrointestinal tract, spleen, kidneys, and adrenal glands). The diagnosis of lobular panniculitis, in our case, was obtained on pleural biopsy with histological examination consisting of normal mesothelial cells surrounded by fatty tissue infiltrated by small lymphocytes without skin lesions or visceral or systemic signs [6-15]. In our case, we reported a pleural panniculitis with recurrent pleural effusion not associated with Weber-Christian disease.

## 2 Case Report

A 82 year old man was hospitalized because of fever, dyspnea and right pleural effusion. He had an history of repeated thoracentesis, diabetes (NIDDM) and COPD. On hospital admission, clinical general conditions showed a pulse rate 80 and respiratory rate 34 per minute without hepato-splenomegaly neither painful erythematous subcutaneous nodules. A chest roentgenogram showed a right-sided pleural effusion. A subsequent total body computed tomography (CT) confirmed the pleural effusion with no evidence of pancreatic abnormality. Laboratory data showed an erythrocyte sedimentation (VES)

\*Corresponding author: Vincenzo Giuseppe Di Crescenzo, Department of Medicine and Surgery, University of Salerno, Baronissi Campus, Via S. Allende, 84081 Baronissi, Salerno, Italy, E-mail: vdicrescenzo@unisa.it

Paolo Laperuta, Filomena Napolitano, Department of Medicine and Surgery, Thoracic Surgery Unit, University of Salerno, Italy

Rosa Maria Di Crescenzo, Department of Medicine and Surgery, Pathology Unit, Federico II University of Naples, Italy

Pio Zeppa, Department of Medicine and Surgery, Pathology Unit, University of Salerno, Italy

Antonio Galderisi, Respiratory Disease Unit, A.O.U. "S. Giovanni di Dio & Ruggi D'Aragona", Salerno, Italy

Carmine Selleri, Hematology and Transplant Center, Department of Medicine and Surgery, University of Salerno, Salerno, Italy

Alessandro Vatrella, Department of Medicine and Surgery, Section of Respiratory Diseases, University of Salerno, Salerno, Italy

Mario Capunzo, Department of Medicine and Surgery, University of Salerno, Baronissi, 84081 Salerno

rate of 15 mm·h<sup>-1</sup> and C-reactive protein (CRP) of 3.0 mg·dL<sup>-1</sup>. White blood cell count was 7,500 cells·mm<sup>-3</sup> (7.5 x 10<sup>9</sup>·L<sup>-1</sup>) with Haemoglobin concentration of 130 g·L<sup>-1</sup>, and 240,000 platelets·mm<sup>-3</sup> (240 x 10<sup>9</sup>·L<sup>-1</sup>). Glutamic oxaloacetic transaminase (GOT) was 34 IU·L<sup>-1</sup>, and glutamic pyruvic transaminase (GPT) 40 IU·dL<sup>-1</sup>. Lactate dehydrogenase (LDH) was 650 IU·L<sup>-1</sup>, and creatine phosphokinase (CPK) was 350 IU·L<sup>-1</sup>. Serum amylase was 85 IU·L<sup>-1</sup>, and lipase was 30 IU·L<sup>-1</sup>. Electrophoretic protein pattern was 6.0g·dL<sup>-1</sup>, and albumin 3.0g·dL<sup>-1</sup>. No antibodies against itself were produced (tests for antinuclear factor, rheumatoid factor and immunocomplex) and urinalysis was normal. Alpha1-antitrypsin (A1AT) was 184 mg·dL<sup>-1</sup>. QuantiFeron test was negative. A cardiology evaluation showed no heart failure manifestations. Right-sided thoracentesis showed a sterile pleural exudate with pleural adenosine deaminase (ADA) level not elevated. Respiratory function showed a moderate obstructive deficit. Thoracoscopic examination revealed parietal pleural thickening of adipose tissue. The surgical procedure consisted, therefore, in the execution of multiple biopsies of the parietal pleura which appeared covered, on the whole surface, by islands of adipose tissue, without macroscopic pathological aspects (Fig. 1). After the procedure we performed pleurodesis with talc. The definitive histological examination consisted of normal mesothelial cells surrounded by fatty tissue infiltrated by small lymphocytes. The patient had no skin lesions or visceral or systemic signs of flogistic involvement of the adipose tissue. Treatment with prednisolone (50 mg·day<sup>-1</sup> for the first week, then 25 mg for the second week) was initiated. A one month-follow up showed no pleural effusion and a complete resolution of symptoms.

**Ethical approval:** The research related to human use has been complied with all the relevant national regulations, institutional policies and in accordance the tenets of

the Helsinki Declaration, and has been approved by the authors' institutional review board or equivalent committee.

**Informed consent:** Informed consent has been obtained from all individuals included in this study.

### 3 Discussion

Weber-Christian disease (WCD) [16-24] characters (recurrent non-suppurative nodular panniculitis [25-30], fever and painful cutaneous nodules, malaise, arthralgia, hepatosplenomegaly, weight loss and anorexia [31-38]) were described by Weber first, and Christian subsequently. While lobular panniculitis associated with extensive visceral involvement may lead to a severe prognosis and eventually to death, due to sepsis, hepatic failure, hemorrhage, and thrombosis, only cutaneous involvement was a positive prognostic indicator. Pleuritis is a rare complication of this systemic panniculitis [39-44]. Deficient levels of pleural ADA were not found. The thoracic complications, in our case, were not associated with extensive visceral involvement. A diagnostic thoracoscopy, performed for recurrent pleural effusion, showed pleural thickening on the whole surface with



**Figures 1-3:** Images of the parietal pleura which appeared covered, on the whole surface, by islands of adipose tissue, without macroscopic pathological aspects.

islands of adipose tissue, without macroscopic pathological aspects.

Pleural thickening biopsy allowed us to diagnose of Idiopathic pleural panniculitis.

In our case, thoroscopic biopsies allowed us to diagnose a rare case of idiopathic pleural panniculitis with recurrent pleural effusion and fever not associated with Weber-Christian disease.

## 4 Conclusions

Idiopathic pleural panniculitis is a very rare occurrence because in the literature are reported only few cases of pleural involvement secondary to idiopathic pulmonary panniculitis, as in the case of Weber-Christian disease or cases of benign pleural lipoma, in which is frequent the involvement of the mediastinum, lung, bronchus, but, rarely, however, the only involvement of the parietal pleura. The presence of pleural panniculitis form with parietal pleural thickening of adipose tissue, without pathological aspects of mesothelial cells, and the presence of lymphocytic infiltrates within the adipose tissue, in a patient with no cutaneous or visceral lesions or signs of systemic inflammatory disease of the adipose tissue, is an occurrence not reported so far in the literature.

**Conflict of interest:** The authors declare that they have no conflict of interest or any financial support.

## References

- [1] Lazarus GS. Panniculitis and disorders of the subcutaneous fat. In: Wyngaarden JB, Smith LH, eds. Cecil Textbook of Medicine, 18th edn. Philadelphia, W.B. Saunders Co., 1988; 2050–2051
- [2] Panush RS, Yonker RA, Dlesk AD, Longley S, Caldwell JR. Weber-Christian disease: analysis of 15 cases and review of the literature. *Medicine* 1985; 64: 181–191
- [3] Weber FP. A case of relapsing nonsuppurative panniculitis, showing phagocytosis of fat cells by macrophages. *Br J Dermatol* 1925; 37: 301–311
- [4] Christian HA. Relapsing febrile nodule nonsuppurative panniculitis. *Arch Intern Med* 1928; 42: 338–351
- [5] Smith KC, Pittelkow MR, Su WP. Panniculitis associated with severe alpha1-antitrypsin deficiency: treatment and review of the literature. *Arch Dermatol* 1987; 123: 1655–1661
- [6] Kumagai-Kurata N, Kunitoh H, Nagamine-Nishizawa M, Watanabe K, Nakamura N. Idiopathic lobular panniculitis with specific pleural involvement. *Eur Respir J* 1995; 1613-1615
- [7] Pfeifer V. Über einen Fall von herdweiser Atrophie des subkutanen Fettgewebes. *Deutsches Archiv für klinische Medizin, Leipzig* 1892: 438-449
- [8] Weber FP. A case of relapsing non-suppurative nodular panniculitis, showing phagocytosis of subcutaneous fat-cells by macrophages. *British Journal of Dermatology and Syphilis, Oxford* 1925: 301-331
- [9] Christian HA. Relapsing febrile nodular nonsuppurative panniculitis. *Arch Intern Med, Chicago* 1928: 338-351
- [10] Hyun SH, Kang YM, Kim CD, Lee JM, Kim IT, Kim NS. Weber-Christian disease presenting with proptosis: a case report. *J Korean Med Sci* 2000: 247-250
- [11] Di Crescenzo V, Laperuta P, Garzi A, Napolitano F, Cascone A, Vatrella A. Small cell lung cancer associated with solitary fibrous tumors of the pleura: a case study and literature review. *Int J Surg.* 2014; 12 Suppl 1:S19-21. doi:10.1016/j.ijso.2014.05.032. Epub 2014 May 22. Review. PubMed PMID: 24859397
- [12] Baganha MF, Pego A, Lima MA, Gaspar EV, Pharm B, Cordeiro AR. Serum and pleural adenosine deaminase: correlation with lymphocytic populations. *Chest* 1990; 97: 605–610
- [13] Laperuta P, Napolitano F, Garzi A, Amato B, Vatrella A, Di Crescenzo V. Extrathoracic recurrence of type A thymoma. *Int J Surg.* 2014;12 Suppl 1:S16-18
- [14] Di Crescenzo V, Laperuta P, Napolitano F, Carlomagno C, Danzi M, Amato B, Garzi A, Vitale M. Unusual case of exacerbation of sub-acute descending necrotizing mediastinitis. *BMC Surg.* 2013;13 Suppl 2:S31. Doi: 10.1186/1471-2482-13-S2-S31. Epub 2013 Oct 8
- [15] Caleo A, Vigliar E, Vitale M, Di Crescenzo V, Cinelli M, Carlomagno C, Garzi A, Zeppa P. Cytological diagnosis of thyroid nodules in Hashimoto thyroiditis in elderly patients. *BMC Surg.* 2013;13 Suppl 2:S41
- [16] Guerra A, Carrano M, Angrisani E, Puzziello A, Izzo G, Di Crescenzo V, Vatrella A, Vitale M. Detection of RAS mutation by pyrosequencing in thyroid cytology samples. *Int J Surg.* 2014; 12 Suppl 1:91-94
- [17] Peluso AL, Cascone AM, Lucchese L, Cozzolino I, Ieni A, Mignogna C, Pepe S, Zeppa P. Use of FTA cards for the storage of breast carcinoma nucleic acid on fine-needle aspiration samples. *Cancer Cytopathol.* 2015; 123(10):582-92. doi: 10.1002/cncy.21577. Epub 2015 Jun 29
- [18] Guerra A, Di Crescenzo V, Garzi A, Cinelli M, Carlomagno C, Pepe S, Zeppa P, Tonacchera M, Vitale M. Diagnostic utility of BRAFV600E mutation testing in thyroid nodules in elderly patients. *BMC Surg.* 2013;13 Suppl 2:S37. doi: 10.1186/1471-2482-13-S2-S37. Epub 2013 Oct 8
- [19] Olivieri A, Cimminiello M, Corradini P, Mordini N, Fedele R, Sella C, Onida F, Patriarca F, Pavone E, Svegliati S, Gabrielli A, Bresciani P, Nuccorini R, Pascale S, Coluzzi S, Pane F, Poloni A, Olivieri J, Leoni P, Bacigalupo A. Long-term outcome and prospective validation of NIH response criteria in 39 patients receiving imatinib for steroid-refractory chronic GVHD. *Blood.* 2013 Dec 12;122(25):4111-4118
- [20] Guerra A, Di Crescenzo V, Garzi A, Cinelli M, Carlomagno C, Tonacchera M, Zeppa P, Vitale M. Genetic mutations in the treatment of anaplastic thyroid cancer: a systematic review. *BMC Surg.* 2013;13 Suppl 2:S44. doi: 10.1186/1471-2482-13-S2-S44. Epub 2013 Oct 8. Review

- [21] Cozzolino I, Varone V, Picardi M, Baldi C, Memoli D, Ciancia G, Selleri C, De Rosa G, Vetrani A, Zeppa P. CD10, BCL6, and MUM1 expression in diffuse large B-cell lymphoma on FNA samples. *Cancer Cytopathol.* 2016 Feb;124(2):135–143. doi:10.1002/cncy.21626. Epub 2015 Sep 28
- [22] Laperuta P, Napolitano F, Vatrella A, Di Crescenzo RM, Cortese A, Di Crescenzo V. Post-pneumonectomy broncho-pleural fistula successfully closed by open-window thoracostomy associated with V.A.C. therapy. *Int J Surg.* 2014;12 Suppl 2:S17-9. doi: 10.1016/j.ijso.2014.08.390. Epub 2014 Aug 23. PubMed PMID: 25159544
- [23] Selleri C, Maciejewski JP, Montuori N, Ricci P, Visconte V, Serio B, Luciano L, Rotoli B. Involvement of nitric oxide in farnesyltransferase inhibitor-mediated apoptosis in chronic myeloid leukemia cells. *Blood.* 2003 Aug 15;102(4):1490-1498
- [24] Cozzolino I, Vigliar E, Todaro P, Peluso AL, Picardi M, Sosa Fernandez LV, Mignogna MD, Tuccari G, Selleri C, Zeppa P. Fine needle aspiration cytology of lymphoproliferative lesions of the oral cavity. *Cytopathology.* 2014 Aug; 25(4):241–249. doi: 10.1111/cyt.12132. Epub 2014 Feb 7
- [25] Baldi A, Mottolese M, Vincenzi B, Campioni M, Mellone P, Di Marino M, di Crescenzo VG, Visca P, Menegozzo S, Spugnini EP, Citro G, Ceribelli A, Mirri A, Chien J, Shridhar V, Ehrmann M, Santini M, Facciolo F. The serine protease HtrA1 is a novel prognostic factor for human mesothelioma. *Pharmacogenomics.* 2008 Aug;9(8):1069–1077
- [26] Cozzolino I, Vigliar E, Sosa Fernandez LV, Selleri C, Pepe S, Vitale M, Triggiani M, Zeppa P. Non lymphomatous clonal B-Cell populations in enlarged lymph nodes in acquired immunodeficiency syndrome. *Infez Med.* 2012;20 Suppl 2:35–42
- [27] Santini M, Fiorelli A, Messina G, Laperuta P, Mazzella A, Accardo M. Use of the LigaSure device and the Stapler for closure of the small bowel: a comparative ex vivo study. *Surg Today.* 2013 Jul;43(7):787–793. doi: 10.1007/s00595-012-0336-0. Epub 2012 Sep 15.
- [28] Zeppa P, Barra E, Napolitano V, Cozzolino I, Troncone G, Picardi M, De Renzo A, Mainenti PP, Vetrani A, Palombini L. Impact of endoscopic ultrasound-guided fine needle aspiration (EUS-FNA) in lymph nodal and mediastinal lesions: a multicenter experience. *Diagn Cytopathol.* 2011 Oct;39(10):723–729. Doi: 10.1002/dc.21450. Epub 2010 Oct 19
- [29] Tauchmanová L, Selleri C, De Rosa G, Esposito M, Di Somma C, Orio F, Palomba S, Lombardi G, Rotoli B, Colao A. Endocrine disorders during the first year after autologous stem-cell transplant *Am J Med.* 2005 Jun;118(6):664–670
- [30] Santini M, Fiorello A, Vicidomini G, Di Crescenzo VG, Laperuta P. Role of diffusing capacity in predicting complications after lung resection for cancer. *Thorac Cardiovasc Surg.* 2007 Sep;55(6):391–394
- [31] Vigliar E, Cozzolino I, Picardi M, Peluso AL, Fernandez LV, Vetrani A, Botti G, Pane F, Selleri C, Zeppa P. Lymph node fine needle cytology in the staging and follow-up of cutaneous lymphomas. *BMC Cancer.* 2014 Jan 6;14:8. doi: 10.1186/1471-2407-14-8
- [32] Tauchmanová L, Selleri C, De Rosa G, Esposito M, Orio F Jr, Palomba S, Bifulco G, Nappi C, Lombardi G, Rotoli B, Colao A. Gonadal status in reproductive age women after haematopoietic stem cell transplantation for haematological malignancies. *Hum Reprod.* 2003 Jul;18(7):1410–1416
- [33] Di Crescenzo V., Laperuta P., Napolitano F., Carlomagno C., Garzi A., Vitale M. Pulmonary sequestration presented as massive left hemothorax and associated with primary lung sarcoma. *BMC SURGERY* (2013). Pag.1-3 ISSN:1471–2482
- [34] Caleo A, Vigliar E, Vitale M, Di Crescenzo V, Cinelli M, Carlomagno C, Garzi A, Zeppa P. Cytological diagnosis of thyroid nodules in Hashimoto thyroiditis in elderly patients. *BMC Surg.* 2013;13 Suppl 2:S41. doi: 10.1186/1471-2482-13-S2-S41. Epub 2013 Oct 8
- [35] Fiorelli A, Morgillo F, Fasano M, Vicidomini G, Di Crescenzo V.G, Di Domenico M, Accardo M, Santini M. The value of matrix metalloproteinase 9 and vascular endothelial growth factor receptor 1 pathway in diagnosing indeterminate pleural. *INTERACTIVE CARDIOVASCULAR AND THORACIC SURGERY* (2012). Pag.1-7 ISSN:1569–9293
- [36] Zeppa P, Sosa Fernandez LV, Cozzolino I, Ronga V, Gesio R, Salatiello M, Picardi M, Malapelle U, Troncone G, Vigliar E. Immunoglobulin heavy-chain fluorescence in situ hybridization-chromogenic in situ hybridization DNA probe split signal in the clonality assessment of lymphoproliferative processes on cytological samples. *Cancer Cytopathol.* 2012 Dec 25;120(6):390-400. doi: 10.1002/cncy.21203. Epub 2012 Apr 19
- [37] Di Crescenzo V., Laperuta P., Napolitano F., Carlomagno C., Garzi A., Vitale M. An unusual case of primary choriocarcinoma of the lung. *BMC SURGERY* (2013). Pag.1-3 ISSN:1471–2482
- [38] Santini M, Fiorello A, Di Crescenzo VG, Vicidomini G, Busiello L, Laperuta P. Use of unidirectional endobronchial valves for the treatment of giant emphysematous bulla. *J Thorac Cardiovasc Surg.* 2010 Jan;139(1):224–226. doi: 10.1016/j.jtcvs.2008.05.069. Epub 2009 Feb 4
- [39] Fiorelli A., S. Mazzone, V.G. Di Crescenzo, G. Vicidomini, S. Costanza, A. Del Prete, A. Mazzone, M. Santini. A simple technique to facilitate dumon silicone stent placement in subglottic tracheal stenosis. *Interact Cardiovasc Thorac Surg.* 2014 Mar;18(3):390–392. doi: 10.1093/icvts/ivt504. Epub 2013 Dec 9
- [40] Santini M, Fiorelli A, Vicidomini G, Laperuta P, Di Crescenzo VG. Iatrogenic air leak successfully treated by bronchoscopic placement of unidirectional endobronchial valves. *Ann Thorac Surg.* 2010 Jun;89(6):2007–2010. doi: 10.1016/j.athoracsur.2009.10.015
- [41] Di Crescenzo V, Vitale M, Valvano L, Napolitano F, Vatrella A, Zeppa P, De Rosa G, Amato B, Laperuta P. Surgical management of cervico-mediastinal goiters: Our experience and review of the literature. *Int J Surg.* 2016 Apr;28 Suppl 1:S47–53. doi: 10.1016/j.ijso.2015.12.048. Epub 2015 Dec 23
- [42] Di Crescenzo V., Laperuta P., Napolitano F., Carlomagno, Danzi M., Amato B., Garzi A., Vitale M. Migration of surgical clips through a right lobectomy stump mimicking an asthmatic syndrome. *BMC SURGERY* (2013) Pag.1-3 ISSN:1471–2482
- [43] Santini M, Fiorello A, Di Lieto E, Di Crescenzo VG, D’aniello G, Vicidomini G, Perrone A, Pastore V. Surgical strategies in cervico-mediastinal goiters. *MINERVA CHIRURGICA*, vol. 61, 2006 p. 221-229, ISSN: 0026–4733

- [44] Rigacci L, Puccini B, Doderò A, Iacopino P, Castagna L, Bramanti S, Ciceri F, Fanin R, Rambaldi A, Falda M, Milone G, Guidi S, Martelli MF, Mazza P, Oneto R, Bosi A, Foà R, Leoni P, Liso V, Pioltelli P, Cascavilla N, Scimé R, Rizzoli V, Ballestrero A, Raimondi R, Arcese W, Musso M, Benedetti F, Guerrasio A, Majolino I, Lambertenghi DG, Baccarani M, Bacigalupo A, Petrini M, Carella AM, Levis A, La Nasa G, Merli F, Narni F, Lauria F, Cortelazzo S, Longinotti M, Olivieri A, Favre C, Cantore N, Da Prada GA, Selleri C, Bruno B, Giovanni M, Leone G, Di Bartolomeo P, Morandi S, Vallisa D. Allogeneic hematopoietic stem cell transplantation in patients with diffuse large B cell lymphoma relapsed after autologous stem cell transplantation: a GITMO study. *Ann Hematol.* 2012 Jun;91(6):931–939