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Thoracic Outlet Syndrome Caused by Hodgkin's Lymphoma - Case Report

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Abstract

The neurogenic thoracic outlet syndrome (TOS) is a term used to describe symptoms of neurovascular compression in the cervical and superior thoracic regions. TOS presents a variety of etiologies, whether benign or malignant, idiopathic or secondary to a primary disease. Imaging exams play an important role in diagnosis and treatment consists of surgical intervention or conservative management. The authors show the case of a young 30 years old woman who presented a tumor in the left supraclavicular fossa region and upper limb irradiated pain ipsilateral to the lesion, compatible with TOS. Cervical region computed tomography scan has revealed numerous nodular lesions with a well-defined contour adjacent to the brachial plexus. The patient underwent surgery and the biopsy result revealed Hodgkin's Lymphoma (LH). Therefore, this work discuss the TOS caused by LH in the supraclavicular fossa region, its consequences, as well as the diagnostic investigation and the surgical treatment.

Keywords: Brachial plexus; Thoracic outlet syndrome; Cervicobrachialgia; Hodgkin's lymphoma

Introduction

Toracic outlet syndrome (TOS) is a broad term used to define various signs and symptoms resulting from abnormal compression of the brachial plexus in the thoracic region, limiting the habitual and labor activities of the affected individual [1]. Some known predisposing factors are age, female gender and longilineous biotype, as well as occupations that require the raising of the arms [2]. It is believed that for the syndrome to occur, a combination of two factors is needed: 1) anatomical narrowing; 2) some type of trauma that triggers the symptoms [3].

The forms of presentation of this syndrome can be classified into two major groups: vascular and neurogenic. The vascular variant accounts for about 5% of cases of TOS and can be subdivided into arterial and venous forms [4]. Neurological symptoms are more frequent and currently correspond to most of the cases, accounting for approximately 95% of clinical complaints. Arterial complications are rare, however, they are more severe than neurological manifestations, and may cause significant sequelae. Its diagnosis is essentially clinical and its treatment differs according to literature [5].

Hodgkin's lymphoma (HL) is a unique type of lymphoma characterized by the proliferation of morphologically variable cells, usually multinucleated B lymphocytes, called Reed-Sternberg cells [6,7]. In the developed countries, the age-incidence curve of the disease has a bimodal pattern characterized by a low incidence in childhood, rapid elevation with a first peak around 20 years, a plateau with a low incidence throughout middle age and a progressive increase of incidence from the age of 55 [8]. Due importance should be given to staging the disease to plan the treatment and determine its prognosis [9]. There are few cases in the literature relating TOS to HL. Therefore, the objective of this study is to describe a framework of radiculopathy triggered by tumor lesions in the region of the supraclavicular fossa and its consequences, as well as diagnostic investigation and surgical treatment.

Methodology

This is a descriptive study of the case report type. Initially, information obtained through a patient's chart review was used. Then, in the period from 05/24/2017 to 06/15/2017, an exploratory and retrospective bibliographic research of the

national and international literature was carried out, covering articles such as case report, literature review, original articles and books on surgery of peripheral nerves and brachial plexus. The articles were selected from the Scielo (Scientific Electronic Library Online), Lilacs and Pubmed published in the years 1975 to 2017 and written in the English and Portuguese languages through the descriptors: "Linfoma de Hodgkin, Hodgkin Lymphoma, Hodgkin Disease, síndrome do desfiladeiro torácico, thoracic outlet syndrome". Among the articles found, those that did not present a significant contribution to the present study were excluded.

Case Report

Patient NLG, 30 years old, female, complaining of moderate intensity cervicobrachialgia in the left upper limb started approximately 6 months ago. The reported pain did not affect a specific dermatome. She denied trauma in the supraclavicular region in the previous months, cyanosis or pallor in the left arm. During the same period of onset of pain, the appearance of nodulation of hardened consistency in the left supraclavicular fossa that caused bulging of the region was observed. He reported that manual compression of the lesion caused shock pain that radiated from the cervical region to the left arm. Patient was taking Losartan for the treatment of Systemic Arterial Hypertension, Sertraline for the treatment of Mood Disorder and Omeprazole. Patient reported being smoker, with consumption of 5-6 cigarettes / day. She stated that she underwent cesarean section in her first pregnancy. Family history was positive for lung tumor.

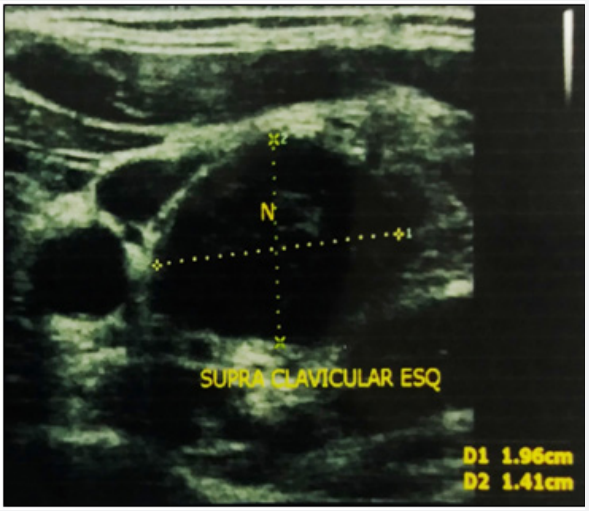

Figure 1: Ultrasonography of the cervical region presenting an oval image located in the left supraclavicular region that presents hypoechogenic, with well-defined contour and with discrete posterior acoustic reinforcement.

Ultrasonography of the cervical region showed the presence of two oval images. These lesions were hypoechogenic, with a well-defined contour that had a slight posterior acoustic reinforcement and were located in the left supraclavicular region, the first one, measuring 2.4 x 1.6 cm and the second, measuring 2.0 x 1.4 cm (Figure 1).

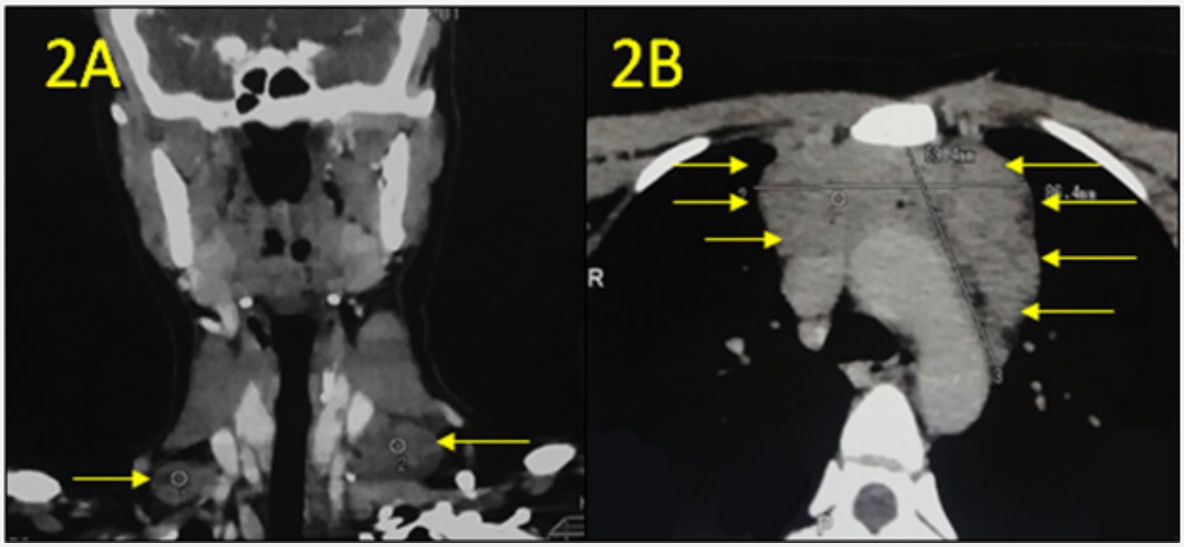

Figure 2A: Contrasted CT scan of the cervical region in a coronal section. Note the bilateral presence of oval-shaped tumor lesion, with no contrast enhancement with well-defined contour in the supraclavicular region. **Figure 2B:** Contrasted CT scans of the lower cervical region and mediastinum in axial section. Note the presence of a large volume tumor lesion in a mediastinal region that shows a well-defined contour and no contrast enhancement.

Contrast computed tomography of the left cervical region revealed the presence of multiple lymph nodes in the pre-aortic anterior mediastinum and in the cervical chain IV, VA and VB bilaterally. The lesions showed no contrast enhancement and presented well defined contour (Figure 2A & 2B).

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The patient underwent surgical treatment in which supraclavicular access was used. For best esthetic purpose, it was decided to perform an incision with one of the cervical folds. The procedure allowed the excision of the nodular lesions as a result of decompression of the left brachial plexus. In the immediate pertoperative period and in the control after 30 days, the presence of cervicobrachialgia relief (Figure 3A, 3B & 4).

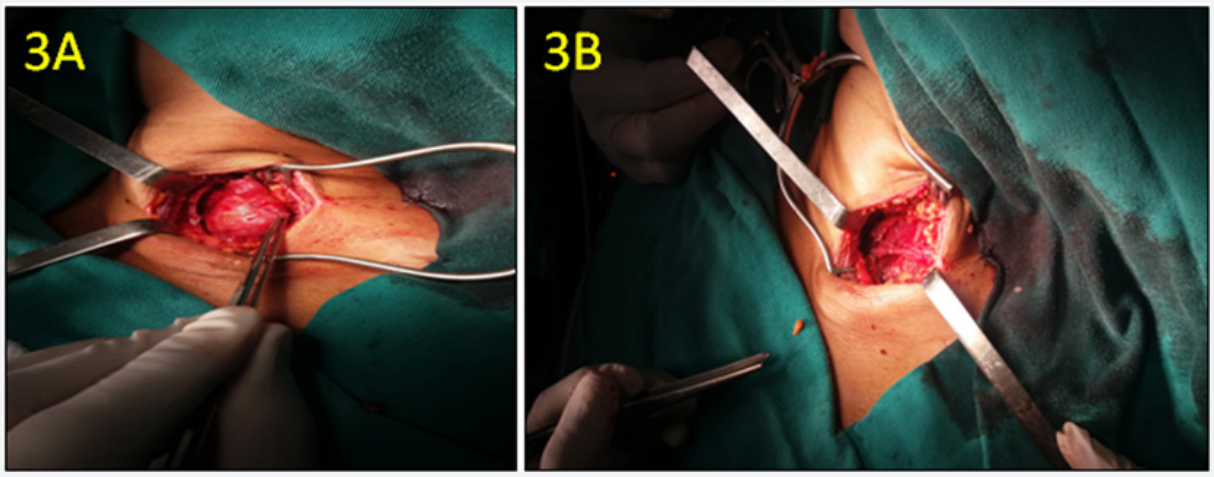

Figure 3A: Intraoperative photography of the left clavicular region. Note the presence of a large fibroelastic tumor with a well-defined contour located under the left brachial plexus. **Figure 3B:** Photograph of the left brachial plexus region after removal of the lesions.

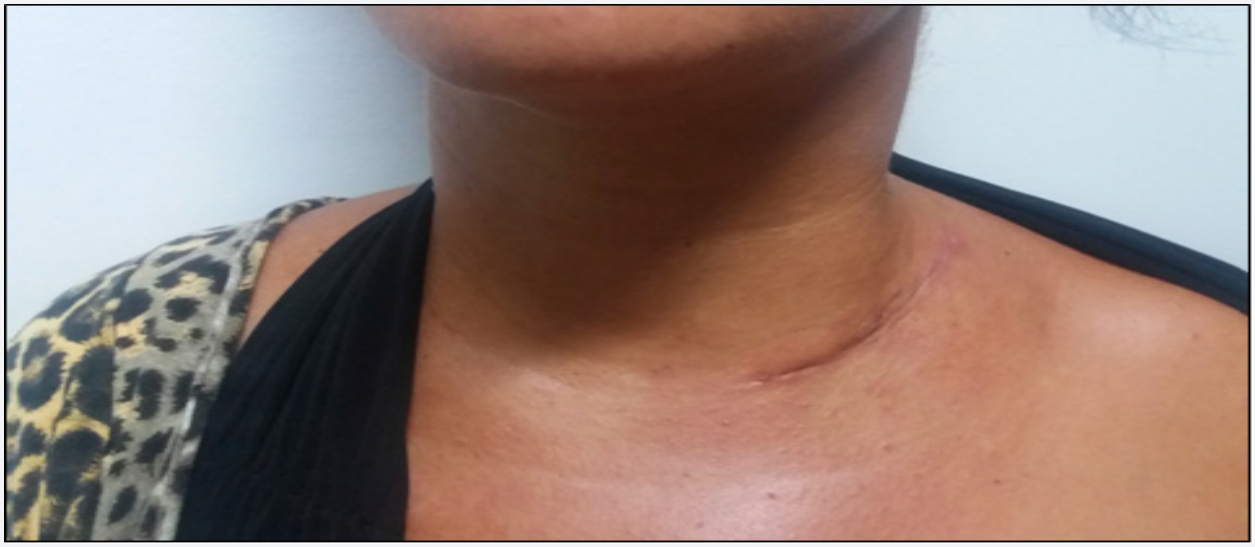

Figure 4: Photograph of the surgical scar with approximately 30 postoperative days. Note the aesthetic aspect of the incision that accompanied one of the cervical folds.

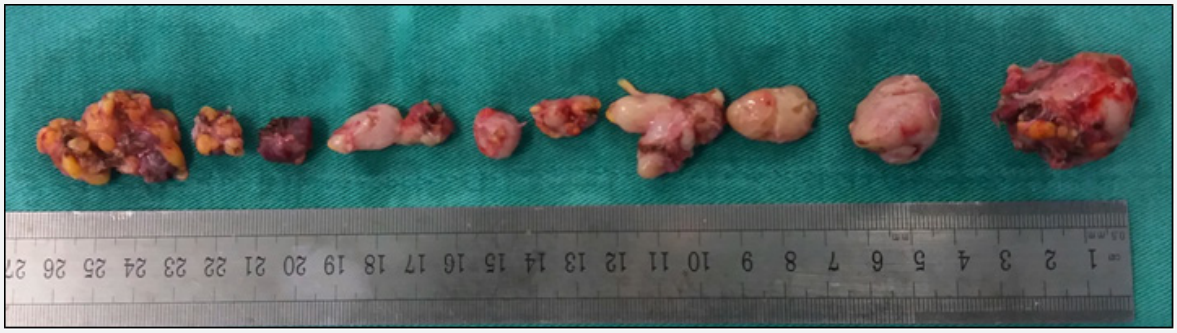

Figure 5: Resected tumor lesions presenting in general, nodular appearance, whitish color and fibroelastic consistency.

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The anatomopathological results revealed 14 nodules, capped, measuring 6.5 x 6.0 x 2.2cm, weighing 24 grams, soft to the cut. In the microscopy revealed lymph nodes presenting alteration in the architecture, with proliferation of bands of fibrous collagen tissue delimiting irregular cellular nodules. The nodules are formed by lymphocytes, some eosinophils, plasma cells, Hodgkin cells and Reed-Sternberg cells, mainly of the lacunar variant. Several mitoses. The findings made up the diagnosis of Hodgkin lymphoma (Figure 5).

The diagnosis was confirmed by the immunohistochemical study and the patient was referred to the hematology to initiate the specific treatment.

Discussion

"Thoracic Outlet Syndrome" was a term first used by Peet et al. in 1956. Epidemiologically, its worldwide incidence varies from 3 to 80 cases / 1,000 inhabitants, being more prevalent in women between 20 and 50 years [10]. Anatomically, thoracic outlet is a region located between the root of the neck, thoracic apex and the beginning of the upper limb, composed of structures such as brachial plexus, subclavian artery and vein, which in their path may suffer compression, designating specific clinical symptoms [11-13]. Several etiologies may lead to compressions, be they benign or malignant, idiopathic or secondary to a basic disease, these will characterize the TOS. There are three sites most prone to compression: triangle of scalene muscles, costoclavicular space and subcoracoid space.

A. Compression in the triangle region of the scalene, the presence of the cervical rib and the formation of fibrous bands can lead to TOS. In this place the compression occurs mainly in its inferior trunk, formed by the roots of C8 and T1 [1,11,14].

B. Compression in the costoclavicular space is a form of TOS usually triggered by repetitive movements that approach the clavicle of the first rib. Physical activities that need to carry heavy objects and movements of prolonged hyperabduction are more associated with this form of TOS [11].

C. Compression in the subcoracoid space is the form of TOS that shows strong connection with shoulder hyperabduction. The coracoid process is lowered by pressing the structures against the minor pectoral muscle [11,13].

Among secondary diseases that compromise any structure of the complex brachial plexus anatomy, which may lead to TOS, we have primary tumors of the brachial plexus, primary tumors of bones and soft parts of the neck (which may invade the brachial plexus directly), Pancoast tumor, amyloidosis, malignant lymphadenopathy [11]. Metastases may also produce symptoms of plexus compression. For example, in breast tumors, lymphadenopathy may compress vessels or nerves [11,15]. The patient in the case had Hodgkin's lymphoma in the supraclavicular fossa.

The clinical manifestations of TOS depend on the structure and location of compression. Neurogenic involvement leads to pain and paresthesias in the affected limb. Pain also in the supra and infraclavicular regions, back of the neck, medial aspect of the forearm and hand, besides the fourth and fifth digits. Muscle weakness can also occur primarily distally, on the hand. Impingement of the subclavian vein can lead to edema and venous distention; of subclavian artery absence of pulse and thrombosis [11]. Imaging tests play a very important role in the diagnosis. In the neurogenic form, the electrophysiological tests have, in a certain way, greater importance than the imaging tests, which is reversed when the vascular form is observed. Electroneuromyography and evoked potential, simple cervical spine radiography, cervical spine and brachial plexus resonance, subclavian and axillary artery angioresonance may be useful [10,11].

Conservative treatment with postural changes, targeted physical therapy, non-steroidal anti-inflammatory drugs and myeloelaxants may be employed. Surgical approach for decompression of the brachial plexus will depend on the clinical presentation and cause of the TOS. True neurogenic forms with motor impairment and neurogenic forms refractory to conservative treatment are indications for surgery, and the most commonly used approach is the supraclavicular approach. As in the vascular forms, which resection of the first rib by transaxillary access is the initial conduct. Such surgical approaches are employed when the cause of TOS is primary. In secondary cases, for example, tumor, this should be treated, logically, with the best access to the tumor for its resection [11].

After surgery, young patients with recent symptoms have a better prognosis. Smoking patients, older patients, chronic pain and opioid abuse have a more reserved prognosis. It should be borne in mind that surgical treatment, however often well tolerated and effective, is also associated with the risk of complications, such as injury of the brachial plexus elements and vascular lesions, and selection should be judicious. of the patients who will undergo the surgical treatment [11,16]. Hodgkin lymphomas (HL) represent a group of neoplasms derived from the clonal expansion of lymphoid cells and, depending on the stage of cell maturation in which malignant transformation occurs, will determine different types of lymphomas [17-19]. They are characterized by the presence of Reed-Sternberg cells involved in an inflammatory context [17]. It occurs mostly in ganglionic tissue and, more rarely, in extraganglionic tissue, such as lung, bone marrow and bones [19].

The onset of LH can occur at any age, although it is rare in children and has a peak incidence in young adults, being more frequent in males [17,19,20]. The cause of this neoplasm is still unknown, however, immune system disorders such as Epstein-Barr Virus (EBV) and Human Immunodeficiency Virus (HIV) infections, chemotherapy, radiation exposure or

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immunosuppressive drugs have been implicated as possible causes. Most patients have no history of these disorders or exposure to radiation therapy or chemotherapy [20,21].

The main associated clinical manifestation is the appearance of cervical adenomegaly without phlogistic signs, with characteristic localization in the cervical, supraclavicular and axillary ganglionic chains [17,18,22]. The classic symptoms of fever, night sweats and weight loss may be present in about 25% of patients. Bone pain and compressive phenomena can be found depending on the affected area and, in the case of very extensive thoracic disease, respiratory complaints [17,20].

The diagnosis is made by histopathological confirmation through a biopsy of the affected site. Histopathological interpretation under the microscope shows some different types of Hodgkin's disease: nodule sclerosis, mixed cellularity, lymphocyte predominance or lymphocyte depletion [20,21,23]. One of its essential characteristics is the presence of a small number of giant cells. Documentation of lymph node location, size and size, as well as the presence of hepatosplenomegaly, is important in determining disease staging [23]. HL is a rare neoplasm that is clinically important because it is curable in more than 75% of cases [19]. The choice of treatment and prognosis depend on the stage of the disease, the patient's age and general conditions. The classic treatment of LH in general consists of multidrug therapy, with or without radiotherapy [19,22].

Conclusion

The case reported raises an important discussion about the importance of the correct investigation of the etiology of the TOS. In this specific case, the imaging tests helped to identify a tumor-like etiology that after the surgical treatment turned out to be HL.

Referances

1. Poole GV, Thomae KR (1996) Thoracic outlet syndrome reconsidered. Am Surg 62(4): 287-291.
2. http://www.emedicine.com/med/topic2774.htm.
3. Sanders RJ Roos DB (1989) The surgical anatomic of the scalene triangle. Contemp Surg 35: 11-16.
4. Short DW (1975) The subclavian artery in 16 patients with complete cervical ribs. J Cardiovasc Surg (Torino) 16)(2): 135-141.
5. Adams JGJ, Silver D (1999) Síndrome do Desfiladeiro Torácico. In: Sabiston DCJ, Lyerly HK, (Eds.), Tratado de cirurgia. As bases biológicas da prática cirúrgica moderna. 15a. Edição, Editora Guanabara Koogan, Rio de janeiro, Brazil, p. 1752-1755.
6. Soares FA (2004) A classificação morfológica e os aspectos histológicos do linfoma de Hodgkin. In: Zago MA, Falcão RP e Pasquini R,(Eds.), Hematologia: fundamentos e prática. São Paulo: Atheneu, Brazil, p. 600.
7. Rocha CVM, Marca FF, Gripp DL, Alves AC, Guimarães MFS, et al. (2011) Síndrome de Sweet associada a linfoma de Hodgkin: relato de caso. An bras dermatol 86(5): 1016-1018.
8. Zago MA, Falcão RP, Pasquini R (2013) Tratado de hematologia. Atheneu, São Paulo, Brazil, p. 549.
9. Souza LNS, Junior PTM, Almeida MTA, Weltman E, Cornacchioni AL, et al. (2010) Hodgkin's lymphoma in children and adolescents: 15 years of experience with the DH-II-90 protocol. Rev Bras Hematol Hemoter 32(4): 295-302.
10. Francisco MC, Yang JH, Barella SM, Francisco FC, Natour J, et al. (2006) Estudo por imagem da síndrome do desfiladeiro torácico. Rev Bras Reumatol 46(5): 353-355.
11. Siqueira MG, Martins RS (2011) Lesões do plexo braquial. Di Livros, São Paulo, Brazil.
12. Moore KL (2011) Anatomia orientada para a prática clínica. 6ed. Rio de Janeiro: Guanabara Koogan, Brazil.
13. Klaassen Z, Sorenson E, Tubbs RS, Arya R, Meloy P, et al. (2014) Thoracic outlet syndrome: a neurological and vascular disorder. Clin Anat 27(5): 724-732.
14. Silvestri K, Wagner F, Moro AND (2005) Síndrome do desfiladeiro torácico: revisão teórica. ACM arq Catarin de med 34(4): 92-96.
15. Narahashi E, Caldana WCI, Zoner C, Honda E, Caporrino FA, et al. (2005) Diagnóstico por imagem do plexo braquial. Rev Bras Reumatol 45(4).
16. Kuwayama DP, Lund JR, Brantigan CO, Glebova NO (2017) Choosing Surgery for Neurogenic TOS: The role of physical exam, physical therapy, and imaging. Diagnostics (Basel) 7(2): pii-E37.
17. Hoffbrand AV, Moss PAH (2012) Fundamentos em Hematologia-6, Edição. Artmed.
18. Marinho DF, Paiva SG, Santos GJVG (2012) Linfoma de Hodgkin: relato de caso. Revista Científica do ITPAC, Araguaína, 5(2).
19. Lopes GCB, Moreira WB, Soares AN, Avaliação (2011) dos resultados do tratamento de pacientes portadores de linfoma de Hodgkin com esquema ABVD em primeira linha. Revista brasileira de oncologia clínica.
20. Barros MHM (2007) Linfoma de Hodgkin na infância e adolescência: um estudo das características histológicas, clínicas, epidemiológicas e de associação com o vírus Epstein-Barr. Rio de Janeiro: Programa de Pós-Graduação em Oncologia- Instituto Nacional de Câncer, p. 182.
21. Machado ACS (2013) Linfoma de Hodgkin biologia, diagnóstico e tratamento. Dissertação (mestrado).
22. Costa TI, Gomes JO, Junior GOD, Giordane E (2018) Linfoma de Hodgkin. Relato de Caso. Revista Brasileira de Clínica Médica 6: 276-278.
23. Spector N (2004) Abordagem atual dos pacientes com doença de Hodgkin. Rev Bras Hematol Hemoter 26(1): 35-42.

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