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Subcutaneous drainage of chronic refractory edema in cancer patients: case presentation

Abstract

Edema is a frequent manifestation in patients with advanced diseases. There is little research on it. The symptomatic impact it generates, the functional limitation, and the decreased quality of life are often underestimated in those affected patients. Many strategies for their management are employed, but the therapeutic response and tolerance are limited in patients with advanced diseases. The following study presents the case of a patient with progressive retroperitoneal sarcoma who develops severe lower limb lymphedema of multifactorial etiology, refractory to treatment, in whom subcutaneous lymphatic drainage was performed with clinical improvement and impact on quality of life. Finally, there is a discussion of the topic and a review of the available literature on the presence of lymphedema in patients with cancer and the different available therapeutic options.

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Keywords: cancer pain, lymphedema, palliative care, quality of life, subcutaneous drainage

Introduction

Lymphedema is an accumulation of proteinaceous material in soft tissues affected by alteration of the lymphatic system for different causes, which favors skin lesions and infections, and affects wound healing [1, 2]. It frequently affects the limbs of patients with chronic oncological and advanced non-oncological diseases, mainly lower limbs, generating marked functional limitations and impact on their quality of life and that of their caregivers, which the medical team

frequently underestimates [3]. Different studies have shown that patients with cancer and lymphedema have a greater functional impairment and psychological and social involvement than patients without lymphedema. Additionally, they have many limitations for exhaustive evaluations and conventional treatments [1, 4]. The World Health Organization and the different lymphedema consensus recommend palliative care for lymphedema management in cancer patients [1].

The diagnostic approach should include different etiologies, such as lymphatic obstruction, hypoalbuminemia

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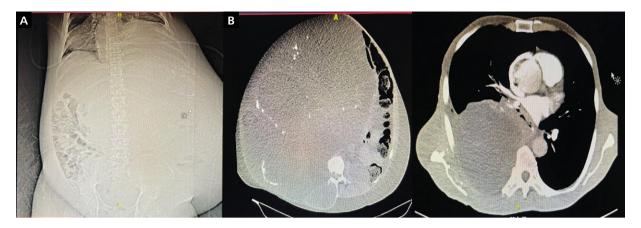


Figure 1. Radiograph (Rx) simple abdomen shows extensive abdominal occupation by tumor mass and displacement of other structures (A); computed tomography (CT) angiography of the chest and abdomen shows pleural and pericardial tumor involvement and associated right pleural effusion (B)

or diseases with protein loss, heart, kidney, or liver disease, infections, trauma, immobility, and medications. Etiologies such as previous pelvic surgery, radiation therapy, and metastatic lymphadenopathy stand out in cancer patients [5]. The therapeutic arsenal for lymphedema management includes options such as diuretic therapy, steroid use, and mechanical treatment with compressive drainage therapies. Although these types of strategies are often effective, in patients with multiple comorbidities and patients with advanced cancer, in terminal stages or palliative care, they may not be appropriate due to the increased risk of adverse effects and intolerance to them, which faces a more complex situation and requires an individualized and multidisciplinary management [5, 6].

A patient was reported with advanced retroperitoneal neoplasia in progression with severe lower limb lymphedema with marked functional limitation and impact on his quality of life, refractory to medical management, in whom was performed controlled subcutaneous drainage, achieving significant clinical improvement and overall quality of life. This type of intervention in patients with cancer and lymphedema is discussed.

Clinical case

A 48-year-old patient was presented, welder, separated, three children, and lived with a daughter. With a diagnosis of retroperitoneal liposarcoma of 3 years of evolution, he received oncological management with chemotherapy schema MAI (mesna, doxorubicine, ifosfamide). He presented tumor mass progression in 2021. He received a second line of chemotherapy with docetaxel and gemcitabine and later progressed again, and he started immunotherapy with pazopanib. It presented with somatic and neuropathic

mixed pain in the abdomen, in place of a large tumor mass, referred to the lower extremities, sarcopenia, and sensation of progressive dyspnea of tumor origin that increased with effort. Chest computed tomography angiography and abdominal computed tomography (Figure 1) showed the absence of pulmonary embolism, $14 \times 10 \times 13$ cm mass that involved right pleura and lung and caused right lower lobe atelectasis, left mediastinal displacement and compression of both atria, extrinsic compression of the source bronchi, intermediate and for the right lower lobe, compression of the inferior vena cava with decreased caliber and obliteration of the right inferior pulmonary vein. In the abdomen, he presented a giant tumor mass of $48 \times 36 \times 35.8$ cm that moved all organs to the left with compression of the urinary tract with dilation of the renal pelvis and collecting groups.

Clinically, the patient with progressive functional decline and dependence associated with abdominal tumor mass growth and severe and progressive lymphedema in lower limbs, limiting, with impact on quality of life, difficulty with urination, and pain of oncological origin. Use of oral morphine at home without improvement. A multimodal hospital analgesic treatment was started with acetaminophen 1 g q8h, pregabalin 75 mg day, hydromorphone 0.4 mg IV 6 h, and rescues of 0.2 mg, supplemental oxygen for dyspnea management and desaturation. A bladder catheter was placed and furosemide 40 mg IV q8h was ordered. Achieved was an improvement of diuresis, better control of pain, and the sensation of dyspnea.

He persisted, however, with severe limiting edema in his lower limbs with a perception of disability. Was a burden to his family and had death wishes. Due to frailty, pain, and disability, no compressive measures were performed, and it was decided, with informed consent, to perform subcutaneous lymphocentesis



Figure 2. Subcutaneous needles inserted into the patient's thigh, and pediatric ostomy pouches for each needle

Table 1. Lymphoedema Quality of Life Tool (LYMPQOL LEG)

Dominions	Inicio	72 h
Functionality	4	2
Appearance/body image	4	2
Symptoms	4	1
Behavior/emotional	4	2
Overall quality of life	2	6

of lower limbs using a Yelco #18 through sterile technique, one on each thigh connected to pediatric ostomy bags (Figure 2), with drainage of abundant clear fluid and progressive decrease of edema 2 cm in the perimeter of the extremities, allowing to get up and move to the bathroom with support, decreased pain and mood improvement. There were no systemic or infectious adverse effects secondary to subcutaneous drainage. The patient was discharged from the hospital to continue home management with support from a home care plan, subcutaneous lymphocentesis at home, subcutaneous opioid medication with low-dose hydromorphone for pain and dyspnea control, and supplemental oxygen by nasal cannula. Lymphoedema Quality of Life Tool (LYMPQOL LEG) scale is performed before and 72 hours after the intervention, with an assessment of 1 to 4, where a lower score is worse to a higher, and overall quality of life from 0 to 10, where the higher value is the better quality of life. The results showed improvement in the different parameters. Table 1 summarizes the main domains of the scale and the results reported by the patient.

Outpatient follow-up was performed by phone. The patient reported adequate pain control and more comfort for him and his caregivers. He needed a change of the needles due to accidental loss. The patient died two months later.

Discussion

The presence of edema in patients with advanced diseases is generally multifactorial, lymphatic, vascular, hypoproteinemic, or permeability edema, and has been shown to negatively impact the physical and psychosocial well-being and quality of life of patients and caregivers. The prevalence of lymphedema in cancer patients is 10-19%, but with even higher rates in gynecological and breast cancer at 47% and 60%, respectively [7, 8]. In the study patient, a giant retroperitoneal liposarcoma generating compression of lymphatic and vascular structures led to refractory lymphedema in lower limbs. Clinical evidence supports that there is an increase in health costs and a great impact on the quality of life of patients with lymphedema, and it is even worse in the presence of cancer due to the high symptomatic burden, fragility, and deterioration of the general condition, and is also directly related to palliative treatment intent and survival [4, 5, 9, 10].

The diagnosis is essentially clinical. Clinical manifestations of lymphedema are associated with edema, increased perimeter, immobility, and distension of soft tissues. Patients report heaviness, soft tissue tension, somatic and neuropathic pain, paresthesia, and exudation. To these are added psychosocial symptoms such as hopelessness, depression, anxiety, disgust, and isolation, clearly described in the study patient, severely impacting his quality of life [1, 7, 8].

Multiple strategies exist for the management of patients with chronic lymphedema with acceptable response, including limb elevation, compressive bandages, physiotherapy, acualinphatic therapy, medicines such as diuretics and steroids, and surgical treatments, as well as the combination of these strategies [1, 4, 10, 11]. Cancer patients have a greater risk of adverse effects with these measures like renal failure and hypotension secondary to diuretics, infections, steroid immunosuppression, and deterioration of heart failure and dyspnea associated with manual decongestant therapy, and they have less satisfactory responses [4, 5]. In addition, physical, manual, and decongestant therapies require trained personnel to ensure the expected goals [1]. All this generates stress in the medical staff and impotence when the desired results are not achieved. It is essential to have clear information, with education, risk prevention strategies, clear goals to achieve, an adequate risk/benefit balance, early diagnosis and intervention, and an emphasis on quality of life over the complete resolution

of lymphedema. All this has been shown to improve adhesion and outcomes [10, 12]. The study patient received pharmacological strategies with steroids and diuretics with poor response, foreseen by the anatomical involvement generated by the large tumor volume that involved intraabdominal vascular structures such as compression of the vena cava.

Several case reports of the use of subcutaneous drainage for the management of chronic lower limb lymphedema have shown benefits in fragile patients with advanced diseases, especially those refractories to conventional measures [2, 5, 8, 9]. Liao et al. [12] reported the therapy of controlled subcutaneous drainage in upper limb lymphedema refractory to decongestant therapy with adequate response in control of the symptoms and functionality.

Controlled subcutaneous lymphedema drainage was first reported by Clein and Purgachev [13] in 8 patients with advanced cancer and lower limb lymphedema, finding improvement in comfort and mobility without associated adverse effects. It is a measure reserved for patients who do not respond to other therapeutic strategies, achieving high success rates, and may also be an option in very fragile patients, prostrate, with poor accompaniment by the health team and absence of caregivers. Some factors that could be considered predictors of response are spontaneous fluid exudation and nonfibrotic lymphedema with foyea [5, 9].

Beck et al. [1] in a systematic review compared controlled subcutaneous drainage, manual lymphatic drainage and multiband decompression therapy, kinesio taping, and the mixture of these manual decompressive therapies in the management of lymphedema in patients with cancer, concluded that all strategies are safe and effective considered individually, notwithstanding the effectiveness cannot be established by the low quality of evidence-based only on case reports and retrospective studies. Grądalski [4] also reported the results of several cases of refractory edema, including cancer patients, managed with venous diuretic and decompressive therapy by trained physiotherapists, with remarkable improvement in the overall discomfort evaluation, improved edema, weight loss, and high adhesion.

Landers et al. [7] in a multicenter prospective observational study evaluated outcomes and impact on quality of life with the treatment of subcutaneous drainage in patients in hospice with refractory lymphedema, and they employed the LYMQOL scale, which is a validated scale to evaluate physical, emotional, and functional aspects in patients with lymphedema. They found improvement in edema, weight loss and perception of appearance, emotional symptoms, func-

tionality, and pain control. The rate of adverse events was 20%, mainly erythema, 2 cases of erysipelas, and 2 cases of acute renal failure. The study patient reached favorable results of subcutaneous drainage in lymphedema management in the overall assessment of quality of life and domains such as self-care, pain, and psychosocial aspects, reported using the LYMPQOL-scale LEG is a validated scale for reporting outcomes and impact on the quality of life of patients with lymphedema [14]. The degree of dependence remains very important in the context of advanced disease without an option for oncological management. No systemic or infectious adverse effects occurred.

There are no standardized recommendations or guidelines about controlled subcutaneous lymphedema drainage in patients with advanced diseases, especially cancer patients, and multiple protocols are employed in case reports of patients with lymphedema with different types of cancer that do not respond to other measures [5]. A different number of needles, the size of needles, butterflies, and yelco of different calibers also varies, and from 1 to 10 subcutaneous catheters. Improved edema, mood and mobility, weight loss, pain control, and comfort were reported outcomes. Instead, the different studies described minimal adverse effects, acute renal failure secondary to abundant fluid drainage, transient subcutaneous fistula, and recurrence of edema after catheter removal. The infection rate varies between 6 and 30%, but is considered low, and in many cases, the presence of erythema as a diagnostic criterion is explained by lymphedema [5, 7]. In the study, the patient used two yelco connected to pediatric urostomy bags. The patient tolerated the procedure well, and no complications occurred.

More evidence is needed based on better methodological quality studies, especially in cancer patients and palliative care since patients with lymphedema in palliative care tend to have advanced and sometimes end-of-life diseases, high fragility index, high symptomatic load, cognitive alterations, rapid deterioration, which makes them have poor tolerance to conventional treatments and difficult to recruit, randomize and follow for clinical studies. From an ethical point of view, based on the limited evidence available, subcutaneous drainage should be considered an off-label strategy, individualized in well-selected patients, and should have informed consent and clarity in risk/benefit uncertainty [2, 10].

Palliative care can support the management of these patients based on their philosophy of comprehensive care for patients and caregivers, communication process, education, transition from care to home, multidisciplinary management, symptom control, and emphasis on quality of life.

Conclusions

Chronic lymphedema of lower limbs is a very prevalent symptom in patients with oncological diseases, and health personnel should be aware because this affects their quality of life negatively. They are also fragile patients and usually do not tolerate multiple treatments. For this reason, it is necessary to highlight the importance of subcutaneous lymphocentesis of the lower limbs, which impacts the patient's symptomatology with minimal adverse effects. Although there is not enough published information, it creates the need for medical personnel to know this procedure as a therapeutic option in individualized patients with refractory chronic lymphedema in the lower limbs.

Article information and declarations

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Author contributions

All authors contributed to the development of the article.

Conflict of interest

All authors declare that they don't have a conflict of interest.

Ethics statement

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Supplementary material

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