Morbus Strangularis Post Living-Donor Kidney Transplantation: Case Report & Review of Literature

Abstract

A 56-year-old man kidney transplant since 1996 with decompensated hepatitis C liver disease. Running on triple immunosuppression in the form of low dose steroids, Tacrolimus and mycophenolate mofetil. He was admitted with history of dental abscess 3 days ago. Examination shows an acutely ill-appearing man with evident of facial edema in the area of the right jaw, no difficulty of swallowing nor acute respiratory distress. Vital signs include an oral temperature of 38.7°C (101.7°F), a heart rate of 90 bpm, and a respiratory rate of 18 breaths/min. Examination of the oral cavity reveals numerous carious teeth, dry oral mucosae, and tinge of jaundice due to decompensated liver disease. Intravenous fluids and intravenous ceftriaxone were started immediately. Three days later, the patient suffered face swelling from the neck to the lower part of the ears, bilaterally. The patient sits upright with his mouth open and his tongue protruding slightly. The floor of the mouth and anterior neck showed woody, tender edema with stiffness of the maxilla-mandibular joint and trismus.

Ultrasound of the neck showed evidence of supraglottic edema, a finding that is confirmed by a computed tomography (CT) scan that confirmed supraglottitis and soft-tissue gas. Leukocyte count was 36 x 10³/mm³, and anion gap measurement indicates metabolic acidosis. After stabilization by administration of intravenous fluids and antibiotic agents, the patient was transferred to the operating room, where drainage and cleaning of the anterior neck and floor of the mouth space was done through 3 incisions: two submandibular & one submental. Blood clots and necrotic tissues were optimally removed, tracheostomy was not indicated. The wounds were closed with drains. Four days later, an additional drainage procedure was performed because of an infected fluid collection, and complete dental extraction was done. Patient was discharged in a good condition and at basal graft function.

Keywords: Ludwig's angina; Post transplantation infection; Odontogenic infection; Angina Ludovici; Morbusstrangularis; Hidden neck infection; Angina maligna

Introduction

Ludwig's angina, otherwise known as angina Ludovici, is a serious, potentially life-threatening infection, of the floor of the mouth, usually occurring in adults with concomitant dental infections and if left untreated, may lead to obstruction of the airways, necessitating tracheotomy, it is named after the German physician, Wilhelm Friedrich von Ludwig who first described this condition in 1836. Other names include “angina Maligna”. Ludwig’s angina should not be confused with angina pectoris, which is also otherwise commonly known as “angina”. In general, the word “angina” comes from the Greek word ankhon, that mean “strangling”, Ludwig's angina refers to the feeling of strangling, not chest pain, though there may be chest pain in Ludwig's angina if the infection spreads into the retrosternal space. The life-threatening nature of this condition necessitates surgical management with critical care in an intensive care unit [1]. The microbiology of Ludwig’s angina is polymicrobial and includes many gram positive and negative aerobic/anaerobic organisms, but the commonly isolated are streptococcal spp., staphylococcus aureus, prevotella spp. And porphyromona spp [2]. Up to our knowledge this is the first case reports Ludwig’s angina in a living-donor kidney transplanted patient.

Case Presentation

A 56-year-old man kidney transplant since 1996 with decompensated hepatitis C liver disease. Running on triple immune suppression in the form of low dose steroids, Tacrolimus and mycophenolate mofetil. He was admitted with history of dental abscess 3 days ago. Examination shows an acutely ill-appearing man with evident facial edema in the area of the right jaw, no difficulty of swallowing nor acute respiratory distress. Vital signs include an oral temperature of 38.7°C (101.7°F), a heart rate of 90 bpm, and a respiratory rate of 18 breaths/min. Examination of the oral cavity reveals numerous carious teeth, dry oral mucosae, and tinge of jaundice due to decompensated liver disease. Intravenous fluids and intravenous ceftriaxone were started immediately. Three days later patient face has become swollen from his neck to the lower part of her ears, bilaterally. The patient sits upright with...
his mouth open and his tongue protruding slightly. The floor of the mouth and anterior neck showed woody, tender edema with stiffness of the maxilla-mandibular joint and trismus.

Ultrasound of the neck showed evidence of supraglottic edema, a finding that is confirmed by a computed tomography (CT) scan confirms supraglottitis and soft-tissue gas (Figure 1A & 1B). Leukocyte count was $36 \times 10^3$/mm$^3$, and anion gap measurement indicates metabolic acidosis. After stabilization by administration of intravenous fluids and antibiotic agents, the patient was transferred to the operating room, where a drainage and cleaning of the anterior neck and floor of the mouth space was done through 3 incisions: two submandibular & one submental. Blood clots and necrotic tissues were optimally removed, tracheostomy was not indicated. The wounds were closed with drains (Figure 2A). Four days later, an additional drainage procedure was performed because of an infected fluid collection, and complete dental extraction was done. Patient was discharged in a good condition and at basal graft function (Figure 2B).

Figure 1: CT Skull, Neck & Upper Chest.

Figure 2: dramatic improvement of the patient months after emergent neck drainage & broad spectrum antibiotics management.
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Discussion

Wilhelm Frederick von Ludwig was the first described Ludwig’s angina in 1836. Ludwig angina defined as a rapid evolution of cellulitis involving the region of the submandibular gland which disseminate through the anatomic contiguity without tendency towards abscess formation [3]. The main etiologic factors of the angina include: the dental infection, trauma, areculation, endo& periodontal condition [4]. Other factors may be involved such as submandibular sialadenitis, or peritonsillar abscesses [5]. Individuals with systemic compromised immunity showed higher susceptibility to Ludwig’s angina, as in HIV, glomerulonephritis, diabetes mellitus, aplastic anemia [6,7] and patients on maintenance immunosuppression medications.

The diagnosis is mainly clinical, patient usually shows a volume increasing hard to palpation in the sublingual, submandibular & submental region bilaterally, which can extend in many times to the suprahypoid region, leading to elevation of the oral floor & tongue [8,9]. Elevation of the tongue is associated with risk of airway obstruction, dysphagia, odynophagia, dysphonia and cyanosis. Signs & symptoms characteristic of infectious processes are observed: high fever, malaise, anorexia, tachycardia and chills [10,11]. Computed tomography is the most complete resource available because through both the axial/coronal cuts and differentiation of the density of soft tissues, it can provide more accurately the dimensions and localization of the infection areas [12]. According to Fogaca, the clinical examination is decisive for the diagnosis of Ludwig’s angina; however, it must be added by a complete anamnesis, image examinations and laboratorial tests. The laboratory tests, such as complete blood picture, renal function, culture and antibiotic sensitivity test, are also of vital importance to monitor the general state of the patient and to determine the microorganisms involved to define the antimicrobrial therapy.

Ludwig’s angina is a severe condition that has a fast evolution leading to life threatening condition either because of the obstruction of the airways secondary to the sublingual and submandibular swelling or due to the dissemination of the infection that could lead to mediastinitis, necrotizing fasciitis or sepsis [13]. Thus, the treatment concentrates around four approaches: maintenance of the airways, incision and drainage, broad-spectrum antimicrobial therapy and elimination of the infectious site [14]. The maintenance of the airways must be a priority in the treatment of the patient, the endotracheal intubation is not recommended because of the possibility of leading the infection to other sites through the rupture of pustules during intubation. Consequently, tracheostomy has been indicated for the most severe cases [15]. The stage of incision and drainage is highly indicated for the decompression of the fascial spaces involved and evacuation of suppuration. The execution of multiple incisions might be necessary [16]. It is recommended that surgical drainage to be associated with broad-spectrum antimicrobial therapy to avoid dissemination of infection to the internal anatomic spaces [1,17]. In conclusion, morbus strangularis is a dramatic, life-threatening condition. Up to our knowledge this is the first case report in a living-donor kidney transplant patient. Transplant nephrologists should intervene early in order to prevent its most dire consequences. It is important to identify the correct diagnosis based on careful and complementary clinical examination, together with an effective drug coverage and early surgical intervention to provide greater control of the transplanted patient health.

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