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Lymphedema associated to sirolimus therapy after pancreas and kidney transplantation: Case report and review of literature

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Abstract

Recently, the development of lymphedema has been reported in patients taking sirolimus after kidney transplantation. The objective of the current study is to report on the association of sirolimus use with the appearance of lymphedema and to suggest a palliative therapy.

The case of a 25-year-old simultaneous pancreas and kidney transplant patient, with complaints of edema of the face and lower limbs after initiating treatment with sirolimus, is reported. Lymphoscintigraphy demonstrated delay in the drainage of radiopharmaceuticals with lymph drainage improving the clinical lymphedema but not the lymphoscintigraphic pattern. It was suggested that sirolimus therapy can trigger functional lymphedema which is reversible with cessation of the drug and that lymph drainage produces clinical improvement.

Key words: Rapamycin, lymphedema, lymph drainage, transplantion, treatment.

Introduction

Lymphedema is an abnormal accumulation of fluids and other substances in the tissue, resulting from impairment of the lymph drainage system [(1)].

Recently, the development of lymphedema has been reported in patients taking sirolimus after kidney transplantation [(2,5)]. Delay in drainage was demonstrated by lymphoscintigraphy suggesting mechanical obstruction, with lymph drainage improving the edema in these patients. Cessation of drug use also improves the lymphedema [(2,3)].

However, the physiopathologic mechanism of this phenomenon is unknown, suggesting further studies are necessary. The objective of the current study is to report the association of sirolimus use with the appearance of lymphedema.

Case Report

The case of a 25-year-old female patient, who had been submitted to simultaneous pancreas and kidney transplant, with subsequent complaints of edema of the face and lower limbs is reported. Transplantation had been performed three years previously as the patient had suffered from diabetes mellitus type I from the age of eight that evolved to renal failure. At around one year post-transplantation, the patient simultaneously presented with edema of the face and legs. She took corticosteroid daily and two years ago she initiated treatment with sirolimus. Soon after, the patient observed edema of the face and lower limbs. She was referred to this service where a lymphoscintigraphy was requested which demonstrated delay in the drainage of radiopharmaceuticals. An intensive treatment program using the Godoy & Godoy technique, was chosen. This technique involves 6- to 8-hour therapy sessions daily. Manual and mechanical lymph drainage was combined with a contention mechanism providing reduction in the edema as assessed by volumetry using the water displacement method. Subsequently the number of treatment sessions was reduced to three times weekly

for two weeks, then twice weekly for two weeks and currently once per week. Another lymphoscintigraphy was performed demonstrating no improvement in the drainage of the radiopharmaceuticals.

Discussion

This study highlights the occurrence of lymphedema in patients after kidney and pancreas transplant associated with the use of sirolimus. There are publications that describe case reports involving this association however no possible physiopathologic mechanism has been established. In the current study, incomplete drainage was observed by lymphoscintigraphy three hours after the administration of radiopharmaceuticals. This illustrates the difficulty of radiopharmaceuticals to enter collectors and to be transported by the lymphatic system. It has been reported that withdrawal of medication leads to an improvement in the lymphedema, thereby suggesting that there is no obstructive state as was conjectured in other studies [(2,3)]. Our hypothesis is that this drug interferes in the contractile mechanism of lymphangions, and hence causes a functional and not obstructive dysfunction. Improvement in the clinical lymphedema by lymph drainage provides a treatment option to reduce the signs and symptoms of this complication. In this patient, an association of edema of the face and legs was observed with clinical improvement seen with lymph drainage. The technique of lymph drainage used can influence the results and so reproduction of the method is important.

In conclusion, sirolimus drug therapy can trigger functional edema which is reversible with the withdrawal of the drug. Additionally, lymph drainage can provide a clinical improvement in the lymphedema.

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