“Successful Treatment of Kimura’s Disease with Radiotherapy: A Case Report”

H. N. Ashikur Rahaman¹, Shravana Kumar Chinnikatti², A.K. Al Miraj³, Md. Magfur Rahman⁴, Md. Saif Ullah Khan⁵

¹Registrar, Dept. of Clinical Oncology, Enam Medical College & Hospital, Savar, Dhaka, Bangladesh
²Research Assistant, Department of Vascular Surgery, Bangabandhu Sheikh Mujib Medical University (BSMMU), Dhaka, Bangladesh
³Cardiac Surgeon & Consultant Cardiologist & Diabetologist, Department of Cardiac Surgery, Bangabandhu Sheikh Mujib Medical University (BSMMU), Dhaka, Bangladesh
⁴Associate Professor & Chairman, Department of Vascular Surgery, Bangabandhu Sheikh Mujib Medical University (BSMMU), Dhaka, Bangladesh

DOI: 10.36347/sjmcr.2020.v08i01.020 | Received: 30.12.2019 | Accepted: 07.01.2020 | Published: 22.01.2020

*Corresponding author: H. N. Ashikur Rahaman

Abstract

Kimura’s disease is a rare chronic inflammatory disorder with angiolymphatic proliferation of unknown cause. It primarily involves the head and neck region, presenting as deep subcutaneous masses and is often accompanied by regional lymphadenopathy, peripheral blood eosinophilia and elevated serum immunoglobulin E (IgE) levels. The disease is endemic in Asians but occurs sporadically in other racial groups and shows a distinct male predilection. Kimura’s disease (KD) or eosinophilic lymph folliculocid granuloma is a rare benign disorder and was first described by Kim and Zeto in China in 1937 [1] and characterized by Kimura et al. in 1948 [2]. Kimura disease (KD) is uncommon benign chronic inflammatory disease first reported by Kimm and Szeto in 1937 in China, but the disease was formally coined as “KD” when in 1948, a Japanese doctor named Kimura et al. published a systemic description of the disease. It is generally seen in young adults, men are affected more commonly. The commonly involved sites are periauricular, groin, orbit, and eyelids. Peripheral blood eosinophilia and elevated serum immunoglobulin E (IgE) levels are constant features of Kimura’s disease. Kimura’s disease is often associated with regional lymphadenopathy, solitary or multiple subcutaneous nodules, sometimes with salivary gland enlargement and elevated serum immunoglobulin E (IgE) levels and peripheral blood eosinophilia [3]. Histopathology of KD shows a marked follicular hyperplasia, eosinophilic infiltrates, and proliferation of post capillary venules [4]. Although KD is a benign condition the lesions can be very difficult to manage. Recurrence rate is very high (above 60%) after local excision [5]. The diagnosis of KD is often difficult and the biopsy or excision of the involved mass for a pathological study is necessary. It is very rare in Bangladesh. Previously not reported this case. We found this case in our OPD in oncology Department of Enam Medical college hospital. Herein we describe this case of Kimura's disease in a 30-year-old Bangladeshi male who presented with a Right subcutaneous mass and cervical lymphadenopathy and successfully treated with radiotherapy.

Keywords: Kimura’s disease (KD), surgery, recurrence, radiotherapy.

Copyright © 2020: This is an open-access article distributed under the terms of the Creative Commons Attribution license which permits unrestricted use, distribution, and reproduction in any medium for non-commercial use (NonCommercial, or CC-BY-NC) provided the original author and source are credited.

INTRODUCTION

The disease is endemic in Asians, but occurs sporadically in other racial groups and shows a distinct male predilection. Kimura’s disease (KD) or eosinophilic lymph folliculocid granuloma is a rare benign disorder and was first described by Kim and Zeto in China in 1937 [1] and characterized by Kimura et al. in 1948 [2]. Kimura disease (KD) is uncommon benign chronic inflammatory disease first reported by Kimm and Szeto in 1937 in China, but the disease was formally coined as “KD” when in 1948, a Japanese doctor named Kimura et al. published a systemic description of the disease. It is generally seen in young adults, men are affected more commonly. The commonly involved sites are periauricular, groin, orbit, and eyelids. Peripheral blood eosinophilia and elevated serum immunoglobulin E (IgE) levels are constant features of Kimura’s disease. Kimura’s disease is often associated with regional lymphadenopathy, solitary or multiple subcutaneous nodules, sometimes with salivary gland enlargement and elevated serum immunoglobulin E (IgE) levels and peripheral blood eosinophilia [3]. Histopathology of KD shows a marked follicular hyperplasia, eosinophilic infiltrates, and proliferation of post capillary venules [4]. Although KD is a benign condition the lesions can be very difficult to manage. Recurrence rate is very high (above 60%) after local excision [5]. The diagnosis of KD is often difficult, and the biopsy or excision of the involved mass for a pathological study is necessary. It is very rare in Bangladesh. Previously not reported this case. We found this case in our OPD in oncology Department of Enam Medical college hospital. Herein we describe this case of Kimura’s disease in a 30-year-old Bangladeshi male who presented with a Right subcutaneous mass and cervical lymphadenopathy and successfully treated with radiotherapy.

CASE REPORT

A 30-year-old male patient came with complaint of swelling in Right Check region for 1 month which was gradually increasing. On examination mass was seen which was firm in consistency, mobile, non-tender and non-adherent to the skin and mandible associated with cervical lymphadenopathy. Others examination was unremarkable. There was no axillary or inguinal lymphadenopathy, hepatosplenomegaly. There was no symptom to suggest pulmonary tuberculosis such as chronic cough, fever, anorexia, weight loss. Hematological examination revealed Hb

© 2020 Scholars Journal of Medical Case Reports | Published by SAS Publishers, India
15.0 gm/dl. Total count of WBC 15,120/cumm, Differential count (Neutrophil 42%, Lymphocyte 20%, Monocyte 03%, Eosinophil 35%, Basophil 00% and platelet was 2,45,000/cumm. Serum total IgE level was 3033.0 IU/mL. S. Creatinine was within normal range 0.7mg/dl. Fine needle aspirate was done from Right parotid region, report was smear show few large atypical cell, lymphocyte, tiny fragments of skeletal muscle and blood. Possibility of a malignant tumor cannot be excluding. Biopsy was advised to confirm diagnosis and histological typing. So Right Total parotidectomy was done in Enam medical college hospital May 26, 2019 and operative sample was sent for histopathology, report was suggestive Non-Hodgkin’s Lymphoma, Immunohistochemistry advised to confirmed diagnosis and rule out malignancy. Immunohistochemistry was done in Armed Forces Institute of Pathology, report was: microscopic examination: several reveal lymph node, perinodal tissue and parotid tissue. All three show florid follicular hyperplasia. The paracortical and interfollicular zone are occupied by numerous eosinophils with formation of eosinophilic microabscess at places. The eosinophils have infiltrated with lyed the follicles at places with formation of eosinophilic microabscess in some of germinal center. Some germinal center contains a proteinaceous precipitate. A few polikaryocyte are also seen in the germinal center. Stoma is fibrosed at place. No R-S cells or its variant is present. No malignancy seen. Immunohistochemistry (IH-628/19 Comments: kimuras’s Disease. The history of patient was he went to hospital with swelling in the Right and Left Parotid region for 3 month which was insidious in onset and gradually increasing in 2008. Left sided superficial parotidectomy was done in June 2008 and operative sample was sent for histopathology, diagnosis was Non-Hodgkin’s Lymphoma and that’s time Patient was taken Tab. Prednisolone 5 mg twice daily. After 2 year in 2010 Right Sided Superficial parotidectomy was done due to swelling was more increasing. No operative sample was sent for histopathology. That’s time again Patient was taken Tab. Prednisolone 5 mg twice daily only and swelling become in subside in Right side Parotid region. Patient was taken Tab. Prednisolone regularly and irregularly. In 2015 again swelling was more in Right sided parotid region, and Right Sided Superficial parotidectomy was done. No operative sample was sent for histopathology. After 4 year in May 2019 patient came our OPD with complaint of swelling in the Right Parotid region for 1 month which was gradually increasing. Then Right Total parotidectomy was done in May 26, 2019. Operative sample was sent for histopathology, report was suggestive Non-Hodgkin’s Lymphoma, Immunohistochemistry advised to confirmed diagnosis and rule out malignancy. Immunohistochemistry report was No malignancy seen, comments: Kimura’s Disease. Patient was taken Tab. Hydrocortisone 20mg twice daily, Tab. Citrizine Dihydrochloride10 mg once daily, while stop medicine swelling was increase. After 2 months patient came with our OPD complaint of swelling in right cheek region which was gradually increasing. Then we had started to evaluation the patient.

**DISCUSSION**

The exact cause and pathogenesis of Kimura’s Disease are unknown. Allergic reaction, infection and autoimmune reaction with an aberrant immune reaction have been suggested. So far, the etiology for KD is still unknown [6]. Kimura’s disease is a rare benign eosinophilic folliculocrypt granuloma. Clinical presentation is predominantly affecting deep subcutaneous tissue and lymph nodes of head and neck region [6, 7]. Its histological characteristics are the follicular hyperplasia, eosinophilic infiltrates and proliferation of postcapillary venules [8]. The disease classified as a benign reactive process. Studies have also shown that the proliferation of CD4+ T cells, especially the CD4 T helper2 (Th2) cells and resultant overproduction of their cytokines, such as granulocyte macrophage colony stimulating factor, tumor necrosis factor-α, IL-4, IL-5, etoxin trigger the production of lymphoid follicle and high IgE. Clonal T-cell population attributes to the disease development and recurrence. The immune reaction that’s believed to the root of Kimura’s disease. The medical therapies for Kimura disease including surgical resection, regional or systemic steroid therapy, radiotherapy, cytotox-ic and laser therapy have been utilized, but none has been proved to be the optimum modality [6, 9, 10]. Surgery with complete excision is difficult since the infiltrative nature of the lesion and swelling of regional lymph nodes [11]. Recurrent lesions may develop several times after initial presentation and surgical removal. It has been suggested that drug therapy is the first choice in middle-aged or elderly patients with KD [12]. Steroid therapy has been shown transiently effective, after weaning, the tumors often increase in size [4]. Recently, anti-IgE therapy has been introduced [13]. The diagnosis of KD is not easy and differential diagnosis include Hodgkin and Non Hodgkin lymphoma, tuberculosis, dermatofibrosarcoma protuberans, Kaposi’s sarcoma, pyogenic granuloma, and other infectious lymph node enlargement for example toxoplasmosis. The diagnostic challenge of Kimura’s disease is generally solved by histological study, fine needle aspiration cytology is helpful in some case, and definitive diagnosis can be obtained by histological study of excised lesion. Through kimura’s disease is a benign disease, radiotherapy is definitive treatment for kimura’s disease. Without radiotherapy the treatment options including surgical resection, regional or systemic steroid therapy, cytotoxic and laser therapy have been used but no one of these proven to be optimum modality. Complete excision of surgery is very difficult since the infiltrative nature of the lesion and swelling lymph node. Recurrent lesion may develop several times after initial presentation and surgical removal. Steroid therapy has been shown effective, after stopping, the swelling often increase in size. The
size of diseased regions and the peripheral blood eosinophil were all decreased after anti-IgE therapy, but complete remission of the tumor was not observed. Radiotherapy has been used in cases of refractory disease [14], and shown to be effective for local control of KD [11, 15]. We treated the patient with radiotherapy and shown to be effective for local control of Kimura’s disease. We prescribed radiation therapy were 45 Gy in 25 fractions, 1.8 Gy per fraction, 6 fractions per week with electron 12 Mev with alternate day bolus over the lesion. After the treatment mass significantly reduced in size and did not shown any side effects. The patient’s condition has remained free from recurrence at the time paper was written.

CONCLUSION
Herein we described a case of kimura’s disease in a 30 year old male patient successfully treated with radiotherapy after failure of surgical removal of three times and medical treatment. Radiotherapy is an effective treatment for kimura’s disease. The radiation field should be limited to the lesion. This strongly suggests that no surgical procedure other than a biopsy should carry out. Although a long time follow up is essential, radiotherapy appears to be a successful and effective treatment for kimuras’s disease of the Right Submandibular region.

REFERENCES