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"Successful Treatment of Kimura's Disease with Radiotherapy: A Case Report"

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Abstract **Case Report**

Kimura's disease is a rare chronic inflammatory disorder with angiolymphaticproliferation 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 are characteristic features and the microscopic picture reveals lymphoid proliferation with eosinophilic infiltration. We report a case of Kimura's disease in a 30year-old male who presented with a Rightsubcutaneous mass and cervical lymphadenopathy. Radiation therapy was performed for recurrence after surgical excision three times. The prescribed radiation dose was 45 Gy, the patient was free of the disease.

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

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 lymphfolliculoid granuloma is 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 common. 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 venues [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. Hereinwe describe this case of Kimura's disease in a 30year-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

15.0 gm/dl, Total count of WBC 15,120/cumm, Differential count (Neutophil 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 Totalparotidectomy 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 lysed the follicles at places with formation of eosinophilic microabscess in some of germinal center. Some germinal center contains a proteinaceous precipitate. A few polykaryocyte 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 only. 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 4year 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 Totalparotidectomy 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 token 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 KDis still unknown [6]. Kimura's disease is a rare benign eosinophilic folliculoid 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,eotoxin trigger the production of lymphoid follicle and high IgE. Clonal T-cell population attributes to the disease development and recurrence. The immune reaction that'sbelieved 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 exci-sion 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 ther-apy 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 studyof 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 stoping, 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

1. Kim HT and Szeto C. Eosinophilic hyperplastic lymphogranuloma, comparison with Muku-licz's disease. J Chin Med. 1937; 23: 699-700.
2. Kimura T, Yoshimura S and Ishikawa E. On the unusual granulation combined with hyperplas-tic changes of lymphatic tissue. Trans Soc Pathol Jpn. 1948; 37: 179-180.
3. Kuo TT, Shih LY and Chan HL. Kimura's dis-ease: involvement of regional lymph nodes and distinction from angiolymphoid hyperpla-sia with eosinophilia. Am J Surg Pathol. 1988; 12: 843-854.
4. Chen H, Thompson LD, Aguilera NS and Ab-bondanzo SL. Kimura disease: a clinicopatho-logic study of 21 cases. Am J Surg Pathol 2004; 28: 505-513?
5. Rajpoot DK, Pahl M and Clark J. Nephrotic syn-drome associated with Kimura disease. Pedi-atr Nephrol. 2000; 14: 486-488.
6. Abuel-Haija M and Hurford MT. Kimura dis-ease. Arch Pathol Lab Med. 2007; 131: 650-1.
7. Kar IB and Sethi AK. Kimura's Disease: Report of a Case & Review of Literature. J Maxillofac Oral Surg. 2013; 12: 109-112.
8. Buder K, Ruppert S, Trautmann A, Bröcker EB, Goebeler M and Kerstan A. Angiolymphoid hy-perplasia with eosinophilia and Kimura's dis-ease - a clinical and histopathological compar-ison. J Dtsch Dermatol Ges. 2014; 12: 224-228.
9. Meningaud JP, Pitak-Arnnop P, Fouret P and Bertrand JC. Kimura's disease of the parotid region: report of 2 cases and review of the lit-erature. J Oral Maxillofac Surg. 2007; 65: 134-140.
10. Alcántara González J, Boixeda P, Truchuelo Díez MT, Pérez García B and Jaén Olasolo P. Angiolymphoid hyperplasia with eosinophilia treated with vascular laser. Lasers Med Sci. 2011; 26: 285-290.
11. Hareyama M, Oouchi A, Nagakura H, Asakura K, Saito A, Satoh M, Tamakawa M, Akiba H, Sakata K, Yoshida S, Koito K, Imai K, Kataura A and Morita K. Radiotherapy for Kimura's dis-ease: the optimum dosage. Int J Radiat Oncol Biol Phys. 1998; 40: 647-651.
12. Monzen Y, Kiya K and Nishisaka T. Kimura's Disease of the Orbit Successfully Treated with Radiotherapy Alone: A Case Report. Case Rep Ophthalmol. 2014; 5: 87-91.
13. Nonaka M, Sakitani E and Yoshihara T. Anti-IgE therapy to Kimura's disease: A pilot study. Au-ris Nasus Larynx. 2014; 41: 384-388.
14. Castro C and Winkelmann RK. Angiolymphoid hyperplasia with eosinophilia in the skin. Can-cer. 1974; 34: 1696-1705.
15. Chang AR, Kim K, Kim HJ, Kim IH, Park CI and Jun YK. Outcomes of Kimura's disease after radiotherapy or non-radiotherapeutic treat-ment modalities. Int J Radiat Oncol Biol Phys. 2006; 65: 1233-1239.