ISSN- O: 2458 - 868X, ISSN–P: 2458 – 8687
National Library of Medicine - ID: 101731606

International Journal of Medical Science and Innovative Research (IJMSIR)
IJMSIR : A Medical Publication Hub
Available Online at: www.ijmsir.com
Volume – 8, Issue – 5, October – 2023 , Page No. : 08 – 11

Kimura Disease

[1]Prajakta Keluskar, Senior Resident, Department of ENT, K.J. Somaiya Medical College and Research Centre, Mumbai.

Corresponding Author: Prajakta Keluskar, Senior Resident, Department of ENT, K.J. Somaiya Medical College and Research Centre, Mumbai.

Citation this Article: Prajakta Keluskar, "Kimura Disease", IJMSIR- October - 2023, Vol – 8, Issue - 5, P. No. 08 – 11.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Aim and objectives: To study the case of Kimura Disease (KD) and arouse the awareness about the disease.

Materials And Methods: 17 years old male patient had left post auricular swelling since 2 months with no palpable lymph nodes. Laboratory assessment showed that his normal renal function, serum electrolytes, low serum albumin (0.8mg/dL), high cholesterol (248 mg/dL), proteinuria (8.0grams/day). His hemoglobin was normal and also the total white cell count but his differential count showed eosinophilia (20%). The virology profile was negative. However, erythrocyte sedimentation rate (ESR)was markedly elevated and IgE level was elevated. Fine needle aspiration cytology (FNAC) was suggestive of follicular lymphoid hyperplasia.

Results: HPE Reported the section was having focal effaced architecture of lymph node and showed follicular lymphoid hyperplasia with variable interfollicular eosinophilia. Proliferation of thin-walled vessels seen with variable hyalinization. There are presence of polykaryocytes and proteinaceous material in the germinal centre showing Kimura's disease.

Discussion: It was unilateral postaural swelling. Peripheral blood eosinophilia and elevated IgE levels observed. It was diagnosed by excisional biopsy. KD is often associated with autoimmune diseases such as ulcerative colitis and more frequently, unlike this case, bronchial asthma [4], typically responding to steroids [8]. In this case, I failed to diagnose the etiology after excluding autoimmune disease.

Conclusion: This case demonstrates the importance of excisional biopsy in recognizing KD. Awareness of this disorder not only by clinicians but also by pathologists might help prevent misdiagnosis of KD. It may be mistaken for serious pathology.

Keywords: Kimura Disease (KD), IGE level, Eosinophilia, Lymph nodes

Introduction

Kimura's disease (KD) A rare, benign inflammatory condition more common in Asians and often affecting the head and neck.

The condition was first described by T. Kimura, a Japanese physician, in 1948.

It is an uncommon lymphoproliferative fibroinflammatory disorder.

Patients present with head and neck subcutaneous nodules with or without lymphadenopathy.

Peripheral blood eosinophilia and elevated serum immunoglobulin E (IgE) levels are typical.

Etiology of KD is still unknown but it may be due to

impairment or interference with immune regulation, atopic reaction to a persistent antigenic stimulus by arthropod bites, virus, and neoplasm. The most interesting hypothesis suggests Candida acting as a source of persistent antigenaemia,[1] although neither hyphae nor spores have been isolated. The disease is manifested by hyperplasia of lymphoid follicles and vascular endothelium. Peripheral eosinophilia and the presence of eosinophils in the inflammatory infiltrate suggest that KD might be a kind of hypersensitivity reaction. With lymphocyte, T-helper 2 (Th2) might play a role.[1]

KD may present with a single enlarged painless lymph node or generalized lymphadenopathy (67% to 100%)[2]in head and neck region, predominantly in preauricular and submandibular area. It may be associated with lymphadenopathy (both local and distal), marked peripheral eosinophilia, and an raised IgE level.[3]

Renal disease is common, with an incidence ranging from 10% to 60% [4], while 10% to 12% of patients may suffer from nephrotic syndrome characterized by clinically relevant proteinuria in 12% to 16% of cases[5]. Renal impairment is probably due to immunocomplex-mediated damage or to Th2-dominant immune response disorders.

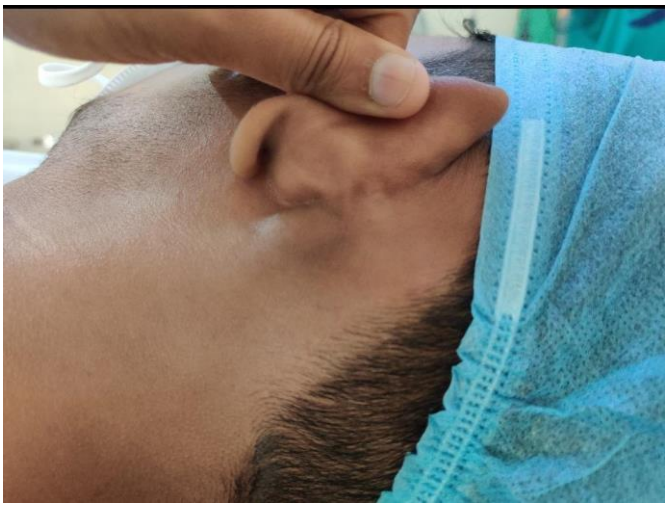


Figure 1

Case Report

17 years old male patient had left post auricular swelling since 2 months.

There was no palpable lymph nodes.

Laboratory assessment showed that his renal function was impaired (creatinine-1.0 mg/dL), normal serum electrolytes, low serum albumin (0.8mg/dL), high cholesterol (248 mg/dL), proteinuria (8.0grams/day). His hemoglobin was normal and also the total white cell count but his differential count showed eosinophilia (20%).

The virology profile was negative regarding hepatitis B and C, cytomegalovirus, and HIV. The immunological status revealed negative anti double strands DNA and both anticytoplasmic neutrophilic antibodies (ANCA-P and C).

However, erythrocyte sedimentation rate was markedly elevated and IgE level was elevated.

Investigations

Normal renal function, serum electrolytes

Low serum albumin (0.8mg/dL), high cholesterol (248 mg/dL), proteinuria (8.0grams/day). His hemoglobin was normal and also the total white cell count but his differential count showed eosinophilia (20%).

The virology profile was negative.

Erythrocyte sedimentation rate (ESR)was elevated.

Serum IgE level was elevated.

CXR PA view normal

FNAC

It was showing atypical lymphoid hyperplasia and few histiocytes.

USG neck

Left postaural lymph node shows fatty hilum.

Diagnosis

Patient underwent a left sided excisional biopsy.

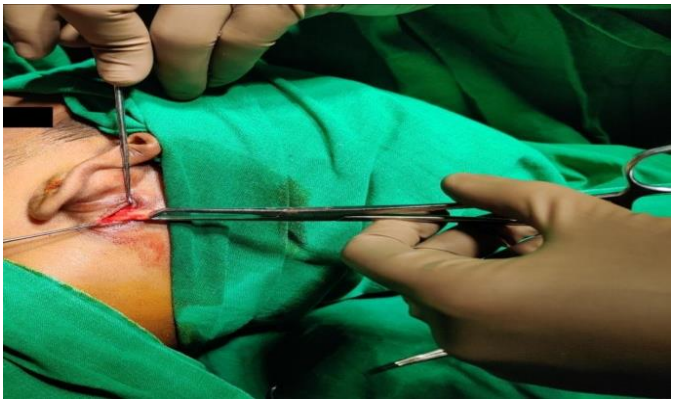


Figure 2

Figure 3

Figure 4

Histopathology

Macroscopy: Received soft tissue bit measuring 1.5x0.5 cm, irregular, Cut section firm and grey white. No cyst identified.

Microscopy: Section shows focal effaced architecture of lymph node and shows follicular lymphoid hyperplasia with variable interfollicular eosinophilia. Proliferation of thin walled vessels seen with variable hyalinization. There are presence of polykaryocytes and proteinaceous material in the germinal centre. It showed Kimura's disease.

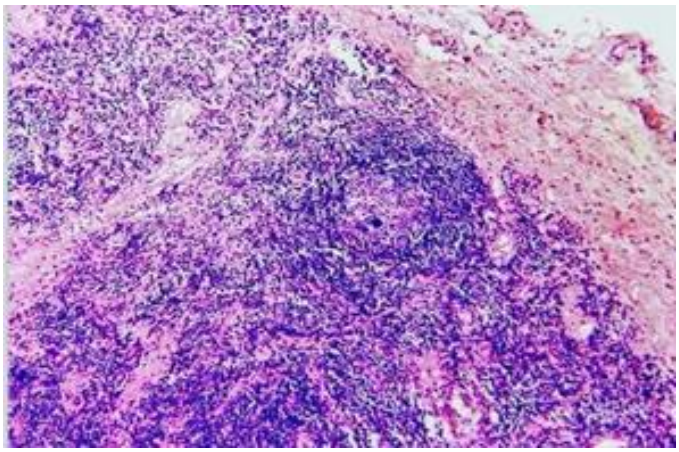
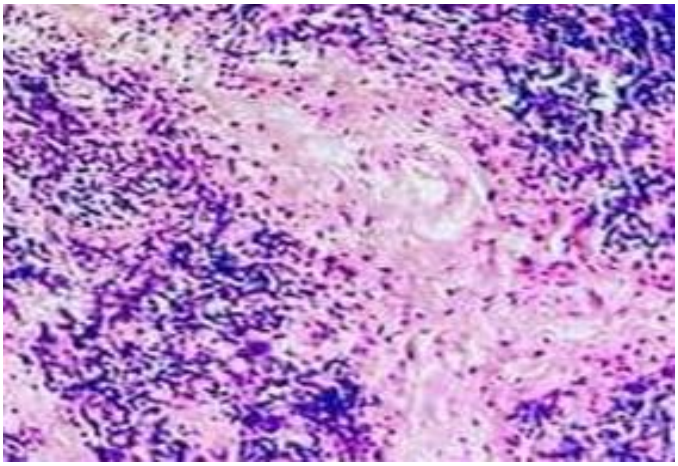


Figure 5



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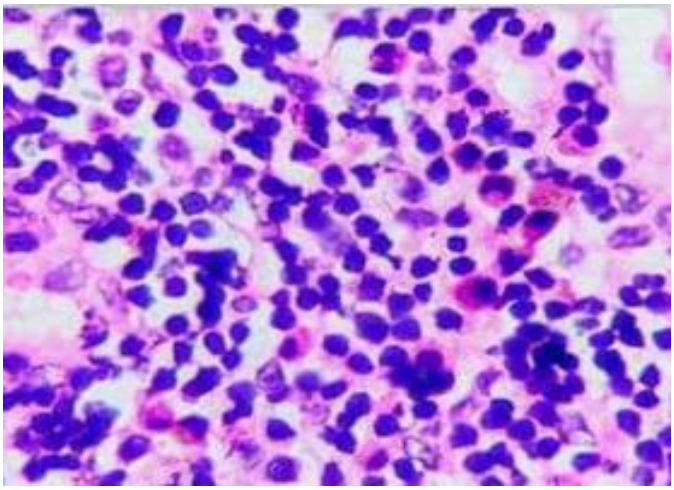

Figure 7

Treatment

Glucocorticoids were given.

- **Discussion**
- It was unilateral postaural swelling.
- Peripheral blood eosinophilia and elevated IgE levels observed.
- It was diagnosed by excisional biopsy.
- Fine needle aspiration cytology was without a diagnostic yield.

Lesion on histopathology showed tissue eosinophilia (100%) and follicular hyperplasia. It is speculated that eosinophilia may play a key role in the occurrence of embolism, but the specific mechanism is unclear [6].

8 weeks follow-up showed no lymph node enlargement.

Angiolymphatic proliferation, usually affecting young men of Asian race and coexisting renal disease is common, with an incidence ranging from 10% to 60% [4,7]. KD is often associated with autoimmune diseases such as ulcerative colitis and more frequently, unlike this case, bronchial asthma [4], typically responding to steroids [8]. In this case, I failed to diagnose the etiology after excluding autoimmune disease.

Conclusion

This case demonstrates the importance of excisional biopsy in recognizing KD.

Awareness of this disorder not only by clinicians but also by pathologists might help prevent misdiagnosis of KD. It may be mistaken for serious pathology.

Glucocorticoids can reduce blood eosinophil count and total IgE levels and play an important role in the treatment of KD.

KD should be considered in patients with subcutaneous masses, eosinophilia, and elevated IgE levels. Biopsy remains the gold standard of diagnosis.

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