



FATAL CASE OF HEMORRHAGIC VIRAL ENCEPHALITIS IN A PATIENT WITH KIKUCHI-FUJIMOTO DISEASE : RARE CNS MANIFESTATION- A CASE REPORT

Neurology

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ABSTRACT

Kikuchi-fujimoto disease is a rare cause of cervical lymphadenopathy which is seen mostly in children and young adults. Only few cases of neurological manifestations have been reported in literature in a patient with KFD. Here we report a case of hemorrhagic viral encephalitis with active Epstein-Barr virus infection in a known case of KFD. Patient presented with severe headache, vomiting and altered sensorium. In a due course the patient was intubated, after a few days the patient developed a ventilator-associated A. Baumannii pneumonia. Despite taking all medical measures, the patient couldn't be saved.

KEYWORDS

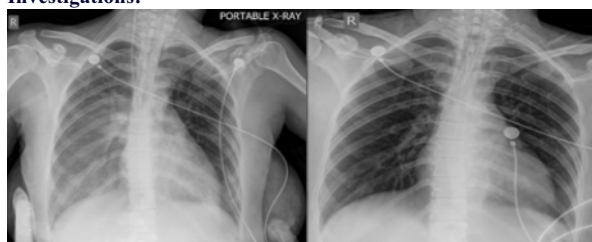
INTRODUCTION:

Kikuchi-Fujimoto disease (KFD) is a rare cause of lymphadenopathy commonly involving cervical lymph nodes. It is more commonly seen in younger females usually in the 2nd to 3rd decade of life. It is typically seen in patients from far eastern and certain south Asian countries. The typical presentation includes cervical lymphadenopathy, fever, arthralgias and rash. Other rare manifestations include axillary lymphadenopathy, splenomegaly, parotitis and interstitial lung disease. Autoimmune etiology has been proposed for the etiology. EBV, parvovirus, Hepatitis B and toxoplasma infections are often associated with KFD infection. Neurological complications are extremely rare and cases have been reported for encephalitis with aseptic meningitis and meningoencephalitis.

CASE PRESENTATION:

A 25 years old female was admitted to the emergency medicine department of SVP hospital, Ahmedabad with high grade fever (101 degrees) and developed altered sensorium after an episode of generalized tonic clonic seizure. Patient was a confirmed case of Kikuchi-Fujimoto disease on excision biopsy from the right-sided cervical lymph node. Patient was eventually intubated in view of low oxygen saturation and altered sensorium. Serum antibodies against Epstein-Barr virus (IgM positive) showed a pattern of recent infection. Series of laboratory investigations were done after confirmed diagnosis of Kikuchi-Fujimoto disease to rule out other associated autoimmune disease which turned out to be positive for SLE. Vitals of the patient were strictly monitored, respiratory and neurological parameters continued to deteriorate, hence higher antibiotics were started accordingly. Culture from the endotracheal tube turned out to be positive for A. Baumannii infection. Patient was managed with injection meropenem, injection acyclovir, injection colistin, injection rituximab and injection hydrocortisone and other supportive treatment was started according to the protocol. Serial laboratory investigation showed worsening of vitals, patient collapsed suddenly a few hours after intubation and CPR was started according to ACLS guidelines but patient could not be revived.

Investigations:



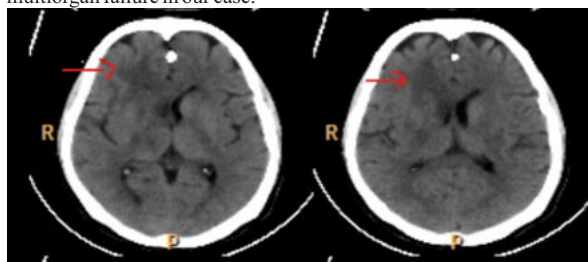
[Figure I]

[Figure II]

Figure II shows chest x-ray at the time of admission.

Figure I shows chest x-ray after 15 days of admission

Chest x-ray showed new development of patchy pneumonic patch in right lower zone, subsequent sputum examination showed growth of A. Baumannii organism which was responsible for septicemia and multiorgan failure in our case.

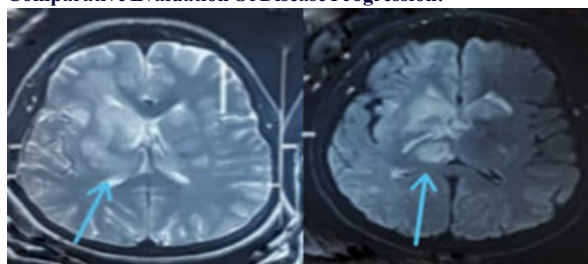


[Figure III]

CT Brain plain study shows abnormal areas of hypodensity involving right corona radiata, right gangliocapsular region and right thalamus. Findings are consistent with MRI study for features of Kikuchi-Fujimoto related encephalitis.

Patient was scanned through a 3T MRI machine (Siemens - SKYRA MODEL) for the MRI brain with venography study at our institute.

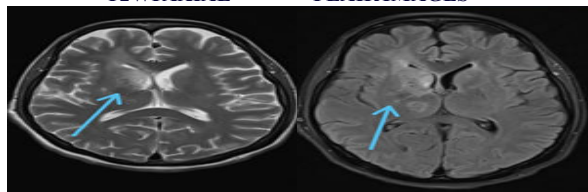
Comparative Evaluation Of Disease Progression:



(MRI done on 10/11/2021)

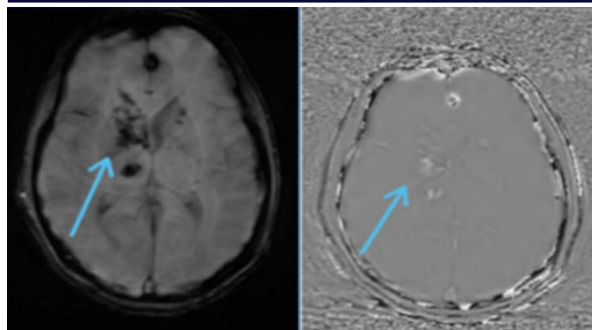
T2WIAXIAL

FLAIR IMAGES



T2WIAXIAL

FLAIR IMAGES



SWI IMAGES PHASIC IMAGES
(MRI done on 21/12/2021)
[FIGURE IV]

T2WI / FLAIR Image shows hyperintensity involving bilateral internal capsule, right thalamus and head of right caudate nucleus.

Susceptibility weighted imaging and phasic imaging showing hemorrhagic foci in bilateral gangliocapsular region and right caudate nucleus. Venography study turned out to be normal.

Scan done after 1 month shows reduction in areas of involvement in corona radiata and thalamus.

Laboratory Profile Of Patient:

Anti Nuclear Antibody (ANA) Profile, Immunoblot			
Parameters	Results	Index	Autoimmune disease association
dsDNA	Weak positive	2.06	SLE
SmD1	Strong positive	4.58	SLE
SS-A	Strong positive	6.10	SS or SLE
Ro-52	Strong positive	4.68	SS or SLE or idiopathic inflammatory myelopathy
nRNP/Sm	Weak positive	2.03	MCTD
Ku	Strong positive	4.45	SLE, Myositis, Systemic sclerosis
Signal visual evaluation	Index/Intensity	Result	
No signal	0- 0.79	Negative	
Very weak band	0.8-1.14	Borderline	
Medium band	1.15-2.49	Weak positive	
Strong band	2.5-3.99	Mid level positive	
Very strong band	>4.00	Strong positive	

DISCUSSION AND LITERATURE REVIEW:

KFD can be a tricky condition to diagnose, in our case diagnosis arrived after exclusion of other possible differentials including neoplastic and demyelinating etiologies after obtaining proper history, clinical examinations and laboratory diagnosis. Repeat MRI study showed reduction in involvement just with supportive therapy which excluded neoplastic etiology, absence of oligoclonal bands in CSF and normal clinical examination excluded demyelinating etiologies. Diagnosis is confirmed on the basis of histopathological findings of a lymph node biopsy. KFD can easily be misdiagnosed as a case of tuberculosis, hence confirmation with the biopsy is of paramount importance. Immunocompromised state in KFD with neurological manifestation carries grave prognosis, although involvement of the nervous system is rare,¹ aseptic meningitis, ² acute cerebellar ataxia and ³ brainstem encephalitis have been reported to complicate the disease. ⁴ A case of focal weakness and associated subcortical CNS lesions in a patient with Kikuchi-Fujimoto disease has been reported in literature.

Hemorrhagic viral encephalitis has not been reported in the literature till date to the knowledge of the author. Because of the rarity of conditions other lesser known neurological features are yet to be reported which may further improve the overall management.

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Ethical Approval And Consent To Participate

The requirement of ethical approval for this case report has been

waived. The patient described in this report has provided informed consent to publish this case. (Publication of imaging results included)

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