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CASE REPORT

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Kikuchi Disease of Parotid Gland

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ABSTRACT

Aim: Kikuchi disease, often referred to as Kikuchi–Fujimoto disease (KFD), is an uncommon benign cervical lymphadenitis etiology. Cervical lymphadenopathy is Kikuchi disease's most prevalent clinical sign. The incidence of Kikuchi disease is reported more in young adult women. **Case Presentation:** We report a rare instance of parotid gland Kikuchi-Fujimoto disease (KFD). A 36-year-old woman presented to our department with a left preauricular tumor that had been present for three weeks. His Ultrasound revealed a well-defined anechoic cystic lesion with solid components in the preauricular area of the left jugular upper. FNAB was done and the results was an adenoma pleiomorfik with supurative inflammation. The patient underwent parotidectomy surgery and specimens sent to pathologic anatomic department. Patient confirmed with Kikuchi disease of parotid gland based on pathologic examination. **Conclusion:** Although rare, Kikuchi disease can be manifest in parotid gland and mimic parotid gland neoplasm. Complete assessment must be done to make right diagnosis to give appropriate treatment to the patient.

Keywords: Kikuchi disease; parotid gland neoplasm; pathologic examination.
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INTRODUCTION

An uncommon and benign cause of cervical lymphadenitis is Kikuchi disease, commonly referred to as Kikuchi–Fujimoto illness. Most cases have been recorded from East Asia and Japan, with fewer from North America and Europe. According to certain research, the female-to-male ratio for kikuchi sickness can reach 4:1, meaning that it is more common among women. Kikuchi illness usually affects young adults, with a mean age of 20 to 30. The most typical sign of Kikuchi sickness is a very sudden development of cervical adenopathy with fever and a flu-like prodrome.

Kikuchi-Fujimoto disease (KFD) usually lasts one to four months and is self-limited. Kikuchi-Fujimoto illness cannot be definitively diagnosed by laboratory testing. This condition does not show up in a typical way on computed tomography (CT) or ultrasonography exams. Only after a pathologic study can Kikuchi be definitively diagnosed. The purpose of this research is to describe a 36-year-old man who was diagnosed with Kikuchi disease of the parotid gland after developing a lump on his left preauricular region.[1,2,3]

CASE REPORT

A 36-year-old male patient arrived at our surgical department complaining of a lump on his left preauricular region that had been there for three weeks. Clinical examination results showed a 4x3 cm lump on his left preauricular region that was firm, somewhat movable, and nontender with no indications of inflammation. There was no evidence of facial nerve paralysis. The systemic examination and the remainder of the head and neck were within normal bounds.

There were no abnormalities in the blood chemistry, erythrocyte sedimentation rate, or complete blood count. A preoperative ultrasound revealed a well-defined anechoic cystic lesions with solid components inside, oval shape, firm borders, size 2.9 x 3.6 cm in the preauricular area of the left jugular upper with non-specific lymph node of left upper jugular.

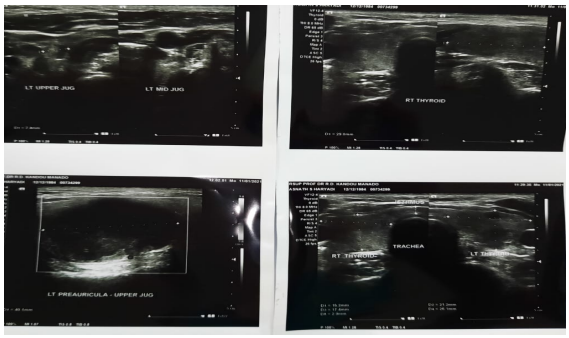

Figure 1 ultrasound well-defined anechoic cystic lesions with solid components inside

His FNAB result several groups of cells with a spindle nucleus and a round nucleus immersed in the mixoid matrix, the distribution of inflammatory lymphocytes, neutrophils, and necrotic debris. FNAB conclusion was an pleiomorphic adenoma with suppurative inflammation.

The patient underwent surgery under general anesthesia, identification of parotid tumor appeared to firmly bordered at superficial parotid gland. The facial nerve was preserved. The tumor was excised with adequate margin of normal gland tissue. Further identification did not appear to be abnormal. A superficial parotidectomy was performed. Bleeding controlled. Installed redon drain. The surgical wound was closed layer by layer.

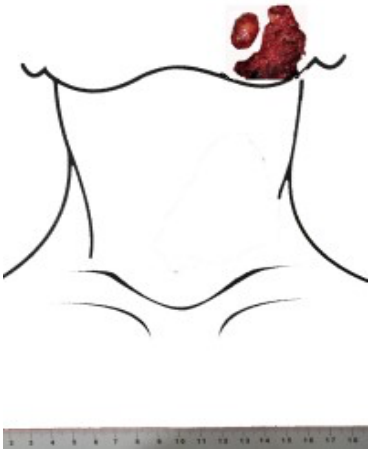

Figure 2. Specimen of parotid tumor and superficial parotid gland sent to pathologic department

On follow up the patient postoperative condition was good with no facial nerve paralysis. Postoperative wound is well, without signs of infection. Would dressing once daily with normal saline moist sterile gauze.

Histopathological analysis of the tumor revealed granulomatous inflammation, foamy histiocytes, karyorrhexis debris (nuclear fraction), and plasma cells with large necrotic areas and eosinophilic mass deposits. No epithelioid cells or Langhans giant cells were found. Pathologic examination final decision was granulomatous inflammation with necrosis tends to be a Kikuchi disease.

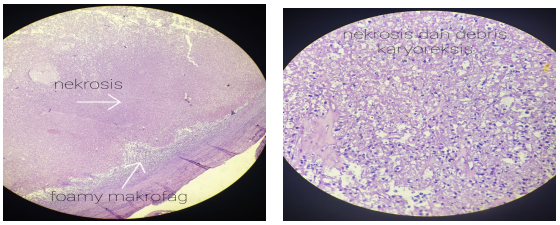

Figure 3. preparation illustrate foamy histiocytes, necrosis and karyorrhexis debris

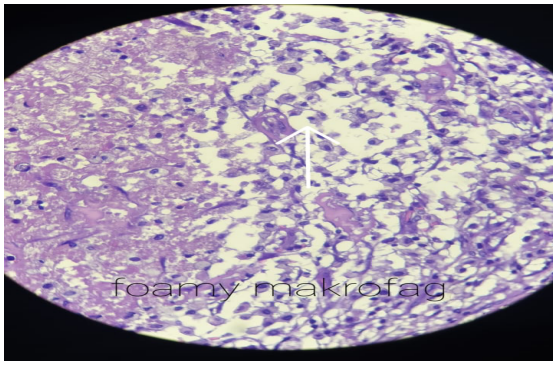

Figure 4. foamy macrophage.

DISCUSSION

Kikuchi disease, also known as Kikuchi-Fujimoto disease or histiocytic necrotizing lymphadenitis, is a rare, idiopathic, and often self-limiting cause of lymphadenitis. In Japan, Kikuchi initially reported the illness in 1972. In the same year, Fujimoto and associates separately described Kikuchi illness.[1,2]

Kikuchi sickness is rare, however cases have been documented worldwide and in all racial groups. Fewer instances have been recorded from North America and Europe, with the majority coming from East Asia and Japan. According to certain research, the female-to-male ratio for kikuchi sickness can reach 4:1, meaning that it is more common among women. Kikuchi illness usually affects young adults, with a mean age of 20 to 30. The etiology of KFD is a subject of significant conjecture; theories include autoimmune or viral causes.[1,2]

The most common presentation of Kikuchi illness is a very sudden start of cervical adenopathy accompanied by fever and a flu-like prodrome. KFD usually lasts one to four months and is self-limited. There have been reports of a modest recurrence rate of 3% to 4%.[1]

The following are characteristics of lymphadenopathy: [1]

- Although numerous nodal chains may be implicated, 83% of patients have lymphadenopathy that is localized to a single site.
- Eighty percent of patients have impacted cervical nodes, with the posterior triangle cervical nodes accounting for 65–70% of these cases.

Microorganism	Supportive Data	Contrary Data
Yersinia enterocolitica	Positive IIF assay applied to lymphatic tissue in 1 case[24]; positive serologic results reported[23]	Histologic features of mesenteric lymphadenitis differ from those of KFD
Toxoplasma gondii	Positive serologic results reported[25]	Histologic features of toxoplasmic lymphadenitis differ from those of KFD
Epstein-Barr virus	Detected by ISH[19,26] and PCR[19,27]	Not detected by ISH,[27-29] SB,[30] or PCR[31]; 50% positive detection in KFD but also 50% positive detection in control samples by PCR[30]
HHV-6	Detected by PCR[32]	Not detected by PCR[27] or SB[32]; 100% positive detection in KFD but also 50% positive detection in control samples by ISH[32]
HHV-8	23% incidence rate by PCR[33]	Not detected in KFD, but 100% detection in control samples by PCR[33]
HTLV-1	Positive serologic results reported[34,35]	Not detected by ISH or PCR[26]
Hepatitis B virus	None	Not detected by ISH[28]
Parvovirus B19	Detected by immunohistochemical analysis in 1 case[36]	Not detected by immunohistochemical analysis[26]
Herpes simplex, CMV, varicella zoster	None	Not detected by PCR[30,31]

CMV, cytomegalovirus; HHV, human herpesvirus; HTLV-1, human T-lymphotropic virus type 1; IIF, indirect immunofluorescence; ISH, in situ hybridization; KFD, Kikuchi-Fujimoto disease; PCR, polymerase chain reaction; SB, Southern blotting.

Figure 5. Microorganisms more frequently reported to have a causative role in KFD.

Extranodal findings:

The thyroid, parotid, myocardium, uvea, and bone marrow are among the infrequently affected extranodal sites. Kikuchi-Fujimoto illness cannot be definitively diagnosed by laboratory testing. This condition does not show up characteristically on computed tomography (CT) or ultrasonography exams.[2]

It is only by histological analysis that Kikuchi-Fujimoto illness may be definitively diagnosed. Typically, Kikuchi Fujimoto illness manifested as a necrotizing condition, with neutrophils either absent or insufficient and patchy or confluent regions of necrosis linked to karyorrhexis. Aspiration with a fine needle cannot definitively diagnose Kikuchi-Fujimoto illness.[1,2]

Histologic finding

Kikuchi disease has three histological stages, which are:[3]

- The proliferative phase includes karyorrhexis, histiocytes, and crescent-shaped nuclei, or crescescentic nuclei.
- Necrotizing phase: widespread necrosis that might disintegrate the lymph node's natural architecture.
- Xanthomatous (foamy cell) phase: the period of recovery when necrosis has been resolved.

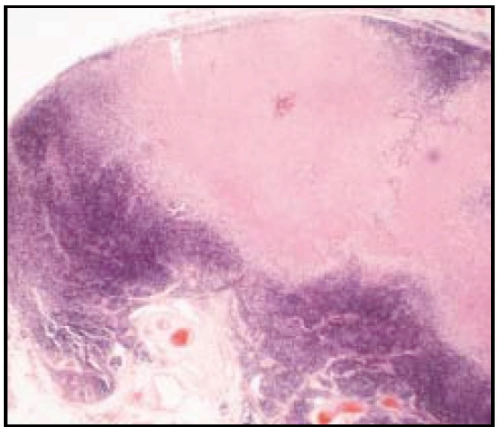

Figure 6. Kikuchi-Fujimoto Disease (KFD) with extensive paracortical area of coagulative necrosis.

Characteristic plasmacytoid monocytes were identified with the use of immunohistochemistry. The latter are natural type 1 interferon-producing cells that are not phagocytic and are most likely engaged in cytotoxic immune responses. Recent research on paraffin-embedded sections utilizing CD68 and HECA-452 antibodies showed that these antibodies, along with CD4, indicated the plasmacytoid monocytes. There has been prior documentation of the co-expression of HECA-452 and CD68 (PG-M1) in plasmacytoid monocytes in Kikuchi's disease.[4]

KFD sufferers do not have a specific therapy. However, as the illness is self-limiting, only symptomatic therapies (such as rest, analgesics, and antipyretics) should be utilized to alleviate distressing local and systemic problems. [5]

In this case report we described a rare presentation of Kikuchi-Fujimoto disease (KFD) in parotid gland. We report a 36 years old man came to our department with swelling on left preauricular area. The onset is subacute evolving during a period of three-weeks. The clinical examination result was a mass measured 4x3 cm without sign of inflammation, firm, moderately mobile and nontender mass on his left preauricular area. No facial nerve paralysis identified.

Ultrasound revealed a well-defined anechoic cystic lesion with solid components inside, oval shape, firm borders, size 2.9 x 3.6 cm in the preauricular area of the left jugular upper with non-specific lymph node of left upper jugular. Patient underwent FNAB examination, the results is an adenoma pleiomorphic with suppurative inflammation. Then he underwent parotid gland tumor removal with superficial parotidectomy. The specimens sent to pathologic anatomic department to confirm diagnosis.

On follow up, pathologic examination results confirmed patient's diagnosis was Kikuchi disease of parotid gland. An enlarged lymph node with paracortical necrotic foci—which are encircled by plasmacytoid monocytes, immunoblasts, and crescentic histiocytes and lack neutrophils—is the pathologic hallmark of Kikuchi's illness. One of the hallmarks of KFD is the presence of nuclear debris, which may be a sign of apoptosis-induced cell death.

CONCLUSION

This case report presented of the 36-year-old man with initial presentation mass on left preauricular for 3 weeks with ultrasound and FNAB of mass suggested parotid gland neoplasm. Pathologic examination of mass revealed Kikuchi disease intra parotid. Diagnosis of Kikuchi disease intra parotid gland was challenging because its presentation mimics parotid gland neoplasm. The definitive diagnosis of Kikuchi only can be made on

pathologic examination. Thus, complete assessment must be done to make right diagnosis to give appropriate treatment to the patient. Awareness of this disorder is important for clinician to prevent misdiagnosis and inappropriate treatment

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DISCLOSURE

The authors affirm no conflict of interest

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