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Case Report

Recurrent buccal Kimura's disease with contralateral parotid gland, lymph nodes, and subcutaneous involvement: A rare case

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Abstract

Background: Kimura's disease (KD) is a rare chronic inflammatory condition predilected in the head-neck region as a painless lump that may mimic malignancy. No previously recurrent KD has been reported in Indonesian males, especially with contralateral involvement.
Case Illustration: We report a 30-year-old male who had a progressive Rahmad Mulyadi[1], Noprianty E Pratiwi[1], Ening Krisnuhoni[2]
buccal lump with a history of biopsy-proven KD. Neck US and MRI imaging and histopathology confirmed a recurrence of buccal KD with involvement of the contralateral right parotid gland, bilateral lymph nodes, and subcutaneous buccal region. After superficial right parotidectomy, steroid, and further chemoradiotherapy, postoperative residuals were shown in follow-up CT. Combined neck US and MRI/CT can demonstrate mass extension. Follow-up imaging is important to evaluate mass extensions and residuals.
Discussion: Kimura's disease is rare in Indonesia. Diagnosis relies on chronic disease history, imaging findings, and histopathological confirmation. Imaging features are non-specific and may mimic neoplasms, making MRI the modality of choice, though biopsy is essential to exclude malignancy. Management includes surgery and prolonged low-dose steroids, with radiotherapy considered for recurrences or steroid-resistant cases.
Conclusion: Diagnosis of recurrent Kimura's disease requires clinical evaluation supported by imaging and histopathology, with ultrasound and MRI/CT useful in assessing lesion extent and residual involvement.

Keywords*: buccal swelling, Kimura's disease, lymphadenopathy, recurrent, parotid*

Abstrak

Latar Belakang: Penyakit Kimura (Kimura's disease/KD) merupakan kondisi inflamasi kronik yang langka, dengan predileksi di daerah kepala dan leher, ditandai oleh benjolan tidak nyeri yang dapat menyerupai keganasan. Belum pernah dilaporkan kasus KD rekuren sebelumnya pada pria Indonesia, khususnya dengan keterlibatan sisi kontralateral.
Ilustrasi Kasus: Kami melaporkan laki-laki berusia 30 tahun dengan benjolan bukal progresif dan riwayat KD yang telah dikonfirmasi melalui biopsi. Pemeriksaan ultrasonografi (US) leher, MRI, dan histopatologi mengonfirmasi rekurensi KD bukal dengan keterlibatan kelenjar parotis kanan kontralateral, kelenjar getah bening bilateral, dan jaringan subkutan bukal. Setelah menjalani parotidektomi superfisial kanan, terapi steroid, dan kemoradioterapi lanjutan, ditemukan adanya residu pascaoperasi pada CT scan tindak lanjut. Kombinasi US leher dengan pencitraan MRI/CT dapat menunjukkan perluasan massa. Pencitraan tindak lanjut penting dilakukan untuk mengevaluasi perluasan massa dan sisa lesi.
Diskusi: Penyakit Kimura jarang ditemukan di Indonesia. Diagnosis bergantung pada riwayat penyakit kronis, temuan pencitraan, dan konfirmasi histopatologi. Karakteristik pencitraan bersifat tidak spesifik dan dapat menyerupai neoplasma, sehingga MRI menjadi modalitas pilihan, meskipun biopsi tetap penting untuk menyingkirkan keganasan. Penatalaksanaan meliputi pembedahan dan pemberian steroid dosis rendah jangka panjang, dengan radioterapi sebagai pertimbangan pada kasus rekuren atau yang tidak responsif terhadap steroid.
Kesimpulan: Diagnosis penyakit Kimura rekuren memerlukan evaluasi klinis yang didukung oleh pencitraan dan histopatologi. Pemeriksaan USG dan MRI/CT bermanfaat untuk menilai luasnya lesi dan residu penyakit.

Kata kunci*: limfadenopati, parotis, pembengkakan bukal, penyakit Kimura, rekuren*

Introduction

Kimura's disease (KD) is a chronic inflammatory granulomatous disease predilected in Asian males.[1] The typical manifestation is a progressive, painless lump in the head-neck that may resemble malignancy, potentially resulting in excessive or delayed treatment.[1–4] Only 200 cases have been histopathologically reported worldwide. There are no reported cases of recurrent KD in Indonesian males, especially in the contralateral subcutaneous tissue.[5–7]

Case Illustration

A 30-year-old Indonesian male presented with a painless lump on the right cheek for about seven years. Laboratory showed increased eosinophils and total IgE. At age fourteen, he had a biopsy-proven left-sided KD managed with parotidectomy, steroids, and radiotherapy. On physical examination, there was a skin-coloured rubbery mass with palpable neck lymph nodes. Imaging showed involvement of the right parotid gland, buccal subcutaneous tissue, and bilateral lymph nodes **[Figure 1].** Histopathology confirmed a recurrent KD. He underwent a right parotidectomy, mass excision-regional flap, steroids, and further chemoradiotherapy for postoperative residuals after a follow-up CT.

Imaging Findings

Neck ultrasonography revealed an ill-defined heterogeneous solid mass with intralesional vascularisation in the right buccal involving the right parotid gland. The right submandibular gland was enlarged with heterogeneous parenchyma. The right neck lymph nodes were also enlarged **[Figure 2]**. No abnormalities were found in the chest radiograph and abdominal ultrasonography. Neck MRI revealed a T1-T2WI iso-hyperintense lobulated solid mass, heterogeneously contrast-enhanced with diffusion restriction in the right buccal involving the parotid gland. The lesion extended to the right zygomatic-buccal-mandibular subcutaneous tissue, bordering the submandibular gland and nearby musculatures, without signs of bone infiltration. A lesion with similar characteristics and smaller size was also found in the left zygomatic-buccal subcutaneous tissue **[Figure 3]**.

Histopathology

Histopathology consistent with Kimura's disease. No malignant signs were seen **[Figure 4]**.

Figure 1

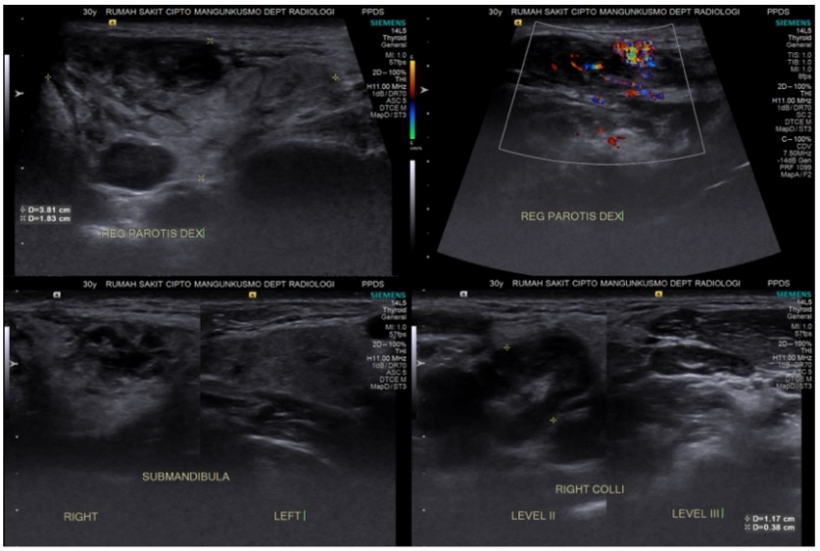

Figure 2

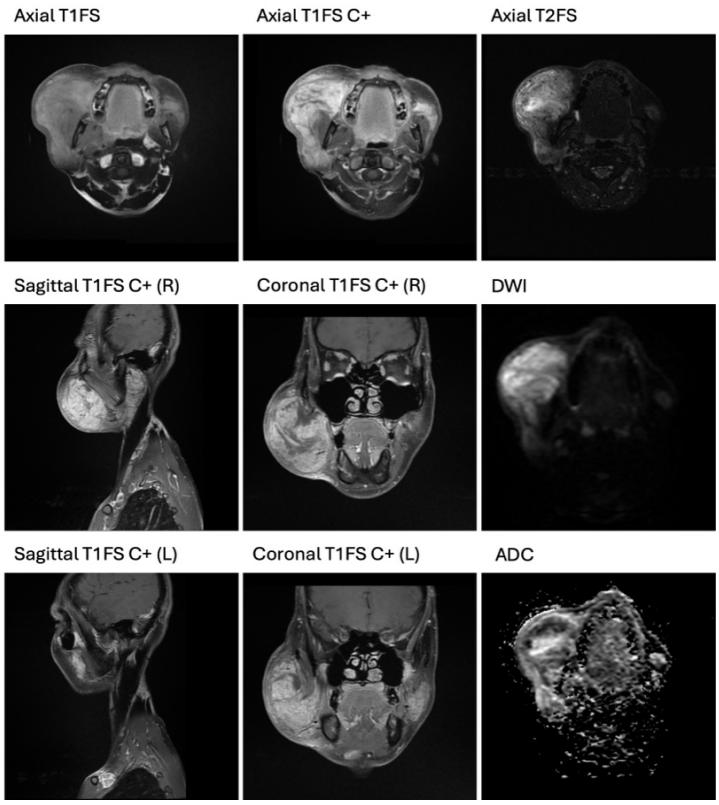

Figure 3

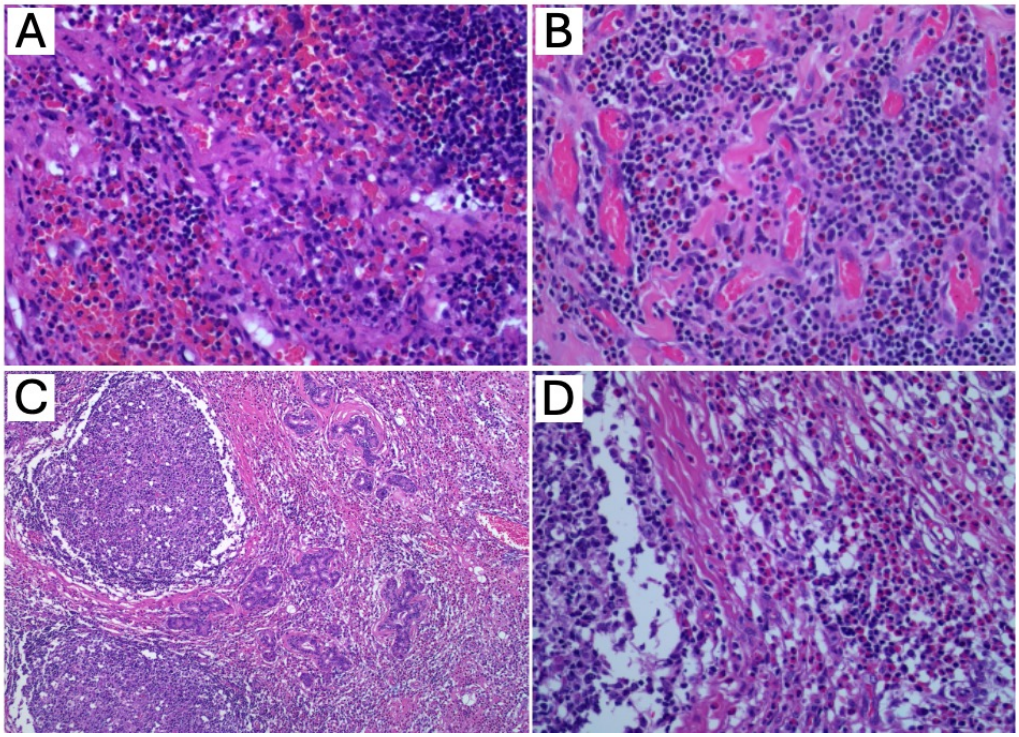

Figure 4.

Management and Follow-Up Imaging

After mass excision and tapering off methylprednisolone, our patient underwent a one-month follow-up CT. An irregular, ill-defined, non-contrast-enhanced solid lesion in the right buccal subcutis involving masticator space and parotid bed. A similar lesion in the left buccal subcutis slightly involved the left masticator space **[Figure 5, Figure 6]**. Our patient underwent further chemoradiotherapy for these postoperative residuals.

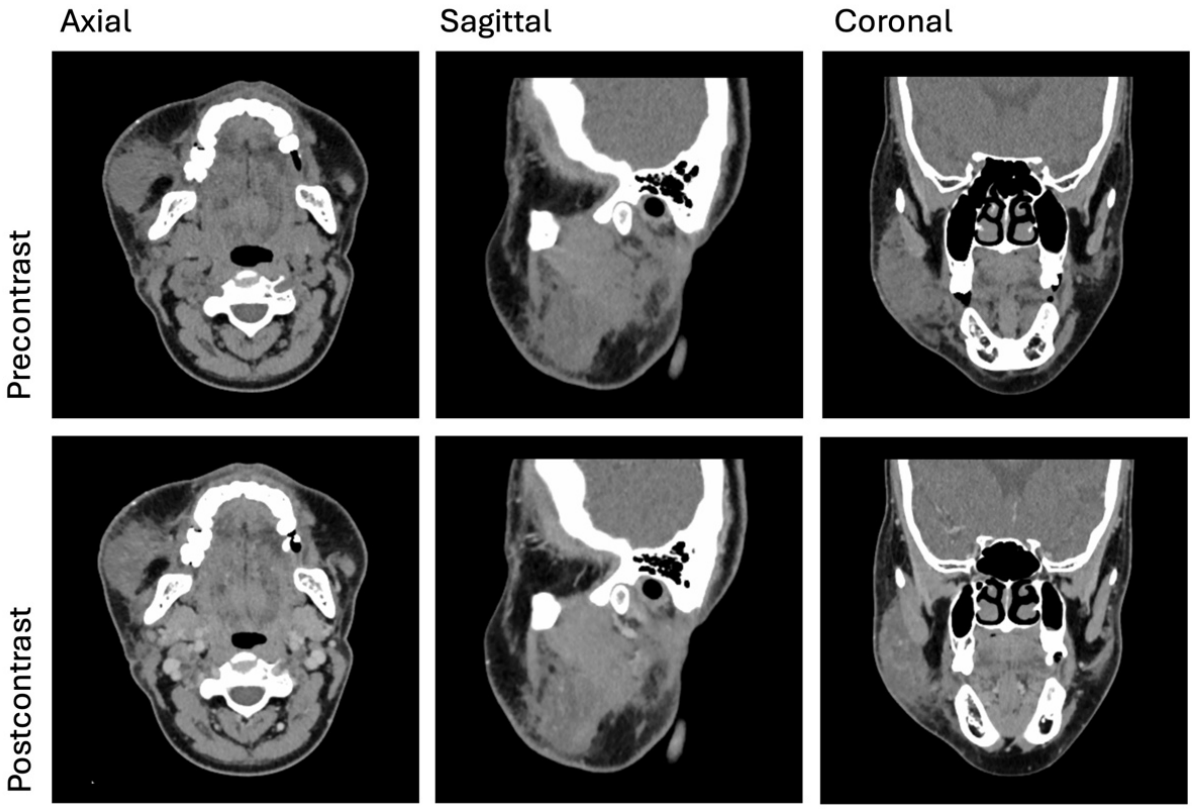

Figure 5.

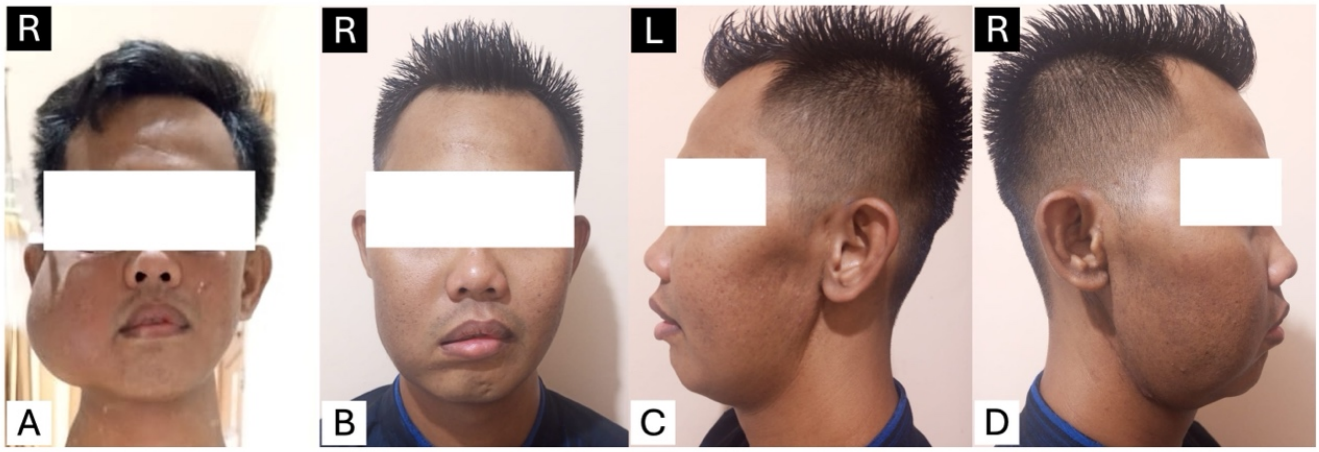

Figure 6.

Discussion

Kimura's disease in Indonesia is rare. Our patient demographics are consistent with the majority of reported cases as a progressive, painless lump in head-neck subcutaneous tissue in Asian males aged 10-40 years (peak in the third decade).[2,8] Our patient had the disease recurrent over 16 years, as some cases of recurrence have been reported over three decades.[9] This history of disease helped lead to the diagnosis.

Imaging characteristics are non-specific. In ultrasonography, salivary glands may appear enlarged and heterogeneous, resembling neoplasm, especially lymphoma. Involved lymph nodes also enlarged with hilar vascularization.[10] On CT, enlarged salivary glands appear hypodense and heterogenously contrast-enhanced. Enlarged lymph nodes show hypodense post-contrast.[11] Our patient had no involvement in other body regions. Some literature reports that KD may involve the axilla, inguinal, extremities, and abdomen.[12] MRI becomes the modality of choice. Lesions may appear well-circumscribed/infiltrative, iso-/hyperintense on T1-T2WI, and homo-/heterogeneous postcontrast. Lymph nodes show homogenous invasion without necrosis.[13,14] Despite similar characteristics, diagnosis should not be based on imaging alone. Histopathology remains necessary to exclude malignancy.[15]

The main management is surgery and conservative constant low-dose steroids to reduce the size of enlarged lymph nodes. Lesions usually re-expand after the steroid is stopped.[16] Radiotherapy is useful for lesions unresponsive to steroids or recurrences after surgery.[15]

Conclusion

Clinical examination with supporting radiology and histopathology is required for KD recurrence diagnosis. A combination of ultrasound and MRI/CT may show mass expansion in the salivary gland, lymph nodes, subcutaneous tissue, and possible residuals.

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