

# Poster Round

## 海報目錄

時間：114年11月23日(星期日)14:25-15:10

地點：台中林酒店 3F 世紀廳海報區

主持人：陳信華醫師

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TCR 60	Drug repurposing: Hydroxychloroquine Reduces the Risk of Hepatocellular Carcinoma in Patients with Hepatitis B Virus Infection 老藥新用: 煙氫奎寧降低B型肝炎病毒感染患者罹患肝細胞癌之風險性	張克宇 <sup>1</sup> 李向嚴 <sup>1</sup> 林韋睿 <sup>1</sup> 楊詠婷 <sup>1</sup> 朱有晨 <sup>1</sup> 林子閔 <sup>1,2</sup> 沈佑銓 <sup>3</sup> 張棋楨 <sup>1,2</sup>
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## Impact of COVID-19 Vaccination on Juvenile Idiopathic Arthritis-Propensity Score-Matched Cohort Study from the TriNetX Global Collaborative Network

Shih-Tsung Fu<sup>1</sup>, Su-Boon Yong<sup>2,3</sup>, Ping-Hao Chiang<sup>4</sup>, Chia-Jung Li<sup>5</sup>, Chih-Wei Tseng<sup>6,7</sup>, Shioh-Ing Wang<sup>8</sup>, James Cheng-Chung Wei<sup>8-10</sup>, Jiu-Yao Wang<sup>2,3</sup>

<sup>1</sup>Department of Education, China Medical University Hospital, Taichung, Taiwan

<sup>2</sup>Department of Allergy and Immunology, China Medical University Children's Hospital, Taichung, Taiwan

<sup>3</sup>Research Center for Allergy, Immunology, and Microbiome (A.I.M.), China Medical University Hospital, Taichung, Taiwan

<sup>4</sup>School of Medicine, Chung Shan Medical University, Taichung, Taiwan

<sup>5</sup>Department of Obstetrics and Gynecology, Kaohsiung Veterans General Hospital, Kaohsiung, Taiwan

<sup>6</sup>Division of Allergy, Immunology and Rheumatology, Department of Internal Medicine, Taichung Veterans General Hospital, Taichung, Taiwan

<sup>7</sup>Department of Public Health, College of Medicine, National Cheng Kung University, Tainan, Taiwan

<sup>8</sup>Institute of Medicine, Chung Shan Medical University, Taichung, Taiwan

<sup>9</sup>Center for Health Data Science, Department of Medical Research, Chung Shan Medical University Hospital, Taichung, Taiwan

<sup>10</sup>Department of Allergy, Immunology & Rheumatology, Chung Shan Medical University Hospital, Taichung, Taiwan

### COVID-19 疫苗接種對青少年特發性關節炎病患的影響—來自 TriNetX 全球健康研究網絡平台的傾向分數配對世代研究

傅仕聰, 楊樹文, 江秉豪, 李佳榮, 曾智偉, 王秀英, 魏正宗, 王志堯

中國醫藥大學附設醫院教學部, 中國醫藥大學兒童醫院兒童過敏免疫風濕科, 中國醫藥大學附設醫院過敏免疫及微菌叢研究中心, 中山醫藥大學醫學院, 高雄榮民總醫院婦女醫學部, 台中榮民總醫院內科部過敏風濕免疫科, 國立成功大學醫學院公共衛生學系, 中山醫學大學醫學研究所, 中山醫學大學醫學研究部健康數據科學中心, 中山醫學大學過敏免疫風濕科

**Background:** Our study aimed to determine the impact of COVID-19 vaccination on medical service use and mortality in juvenile idiopathic arthritis (JIA) patients.

**Methods:** We retrieved data from the US Collaborative Network in TriNetX between January 1st, 2020 and June 30th, 2023. We investigated the incidence of COVID-19, medical utilization, and all-cause mortality in JIA patients with or without COVID-19 vaccination. The hazard ratio (HR) and 95% confidence interval (95% CI) of the outcomes were calculated between propensity score-matched (PSM) 1:1 groups. A total of 652 vaccinated and 652 unvaccinated JIA patients were enrolled. Sensitivity analyses and subgroup analyses were also conducted.

**Results:** The vaccinated group demonstrated an elevated risk of infection (HR = 2.285, 95% CI: 1.219-4.282) than the unvaccinated group, and a notably higher overall use of medical services (HR = 2.112, 95% CI: 1.081-4.129). There was no difference in all-cause mortality between the two groups. As for sensitivity analysis, the risk of outcomes did not significantly differ between the COVID-19 vaccine group and the influenza vaccine group. When the Delta variant prevailed, vaccination was associated with a statistically non-significant rise in COVID-19 risk (HR = 1.402, 95% CI: 0.599-28.60); in the era of Omicron, there was increased overall medical service use (HR = 2.326, 95% CI: 0.884-6.121).

**Conclusions:** JIA patients with COVID-19 vaccination had a higher risk of COVID-19 infection, leading to increased medical service use but no significant rise in mortality. These results warrant further investigation into the impact of the vaccine and its immunological responses.

## 海報摘要 TCR59

### Drug repurposing: Hydroxychloroquine Reduces the Risk of Hepatocellular Carcinoma in Patients with Hepatitis C Virus Infection

Wei-Jui Lin<sup>1</sup>, Ke-Yu Chang<sup>1</sup>, Hsiang-Gyen Lee<sup>1</sup>, Yung-Ting Yang<sup>1</sup>, Yu-Chen Chu<sup>1</sup>, Tzu-Min Lin<sup>1,2</sup>, Chi-Ching Chang<sup>1,2</sup>

<sup>1</sup>Division of Allergy, Immunology and, Rheumatology Taipei Medical University Hospital

<sup>2</sup>Division of Allergy, Immunology and Rheumatology, Department of Internal Medicine, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan

**老藥新用: 氫氯奎寧降低C型肝炎病毒感染患者罹患肝細胞癌之風險性**

林韋睿<sup>1</sup> 張克宇<sup>1</sup> 李向嚴<sup>1</sup> 楊詠婷<sup>1</sup> 朱有晨<sup>1</sup> 林子閔<sup>1,2</sup> 張棋楨<sup>1,2</sup>

台北醫學大學附設醫院過敏免疫風濕科<sup>1</sup>

台北醫學大學醫學院醫學系內科學科過敏免疫風濕學科<sup>2</sup>

#### Abstract

**Background:** Hydroxychloroquine (HCQ) has been suggested to exert anticancer effects through anti-inflammatory, autophagy-related, and non-autophagy-related mechanisms. This study investigated the association between HCQ use and hepatocellular carcinoma (HCC) risk in patients with chronic hepatitis C virus (HCV) infection.

**Methods:** We enrolled patients with HCV infection from Taiwan's National Health Insurance Research Database, covering the period from January 1, 2006, to December 31, 2016. Kaplan–Meier analysis and Cox proportional hazards regression were used to analyze HCC risk in HCQ users and nonusers. We evaluated the association of HCC risk with concurrent medication use and comorbidities.

**Results:** We included 139,263 patients with newly diagnosed HCV infection. During the follow-up period, 1037 patients developed HCC. In the HCQ user group (n =1598), 62 patients had HCC, whereas 975 of the 15,980 HCQ nonusers developed HCC. Patients with chronic HCV infection who used HCQ had a significantly lower risk of HCC than did those who did not use HCQ (adjusted hazard ratio [aHR], 0.68; 95% CI, 0.51–0.92). No dose–response relationship was observed between the HCQ user and nonuser groups. The use of concurrent medications, such as antihistamines (aHR, 0.74; 95% CI, 0.59–0.93), statins (aHR, 0.38; 95% CI, 0.32–0.46), nonsteroidal anti-inflammatory drugs (aHR, 0.60; 95% CI, 0.48–0.76), and aspirin (aHR, 0.82; 95% CI, 0.72–0.94), was associated with a lower risk of HCC.

**Conclusion:** HCQ use may reduce HCC risk in patients with HCV infection. The results provide key insight into the potential benefits of HCQ use in preventing HCC in patients with chronic HCV infection. Future studies should determine the mechanisms underlying the anticancer effect of HCQ.

## 海報摘要 TCR60

### Drug repurposing: Hydroxychloroquine Reduces the Risk of Hepatocellular Carcinoma in Patients with Hepatitis B Virus Infection

老藥新用: 煙氣奎寧降低 B 型肝炎病毒感染患者罹患肝細胞癌之風險性

Ke-Yu Chang<sup>1</sup>, Hsiang-Gyen Lee<sup>1</sup>, Wei-Jui Lin<sup>1</sup>, Yung-Ting Yang<sup>1</sup>, Yu-Chen Chu<sup>1</sup>, Tzu-Min Lin<sup>1,2</sup>, Yu-Chuan Shen<sup>2,3</sup>, Chi-Ching Chang<sup>1,2</sup>

<sup>1</sup>Division of Allergy, Immunology and, Rheumatology Taipei Medical University Hospital

<sup>2</sup>Division of Allergy, Immunology and Rheumatology, Department of Internal Medicine, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan

<sup>3</sup>Division of Rheumatology, Immunology and Allergy, Taipei Medical University Wang Fang Hospital

張克宇<sup>1</sup> 李向嚴<sup>1</sup> 林韋睿<sup>1</sup> 楊詠婷<sup>1</sup> 朱有晨<sup>1</sup> 林子閔<sup>1,2</sup> 沈佑銓<sup>3</sup> 張棋楨<sup>1,2</sup>

台北醫學大學附設醫院過敏免疫風濕科<sup>1</sup> 台北醫學大學醫學院醫學系內科學科過敏免疫風濕學科<sup>2</sup>

台北醫學大學萬芳醫院過敏免疫風濕科<sup>3</sup>

#### Abstract

**Background:** Hydroxychloroquine (HCQ) may protect against cancer, but its effects in patients with hepatitis B virus (HBV) remain unexplored. We investigated the association of HCQ use with hepatocellular carcinoma (HCC) risk in patients with HBV. **Methods:** This population-based cohort study selected the data of patients with HBV infection during 2006–2016 from Taiwan's National Health Insurance Research Database. We performed multivariate and stratified analyses using the Kaplan–Meier method and Cox proportional hazards regression to evaluate the association of HCQ use with HCC risk in the HBV cohort.

We included 688,295 patients with newly diagnosed HBV infection. During follow-up, patients with HBV who used HCQ had a significantly lower HCC risk than those who did not use HCQ (adjusted hazard ratio [aHR], 0.47; 95% confidence interval [CI], 0.32–0.69). Furthermore, we stratified the patients into subgroups according to

cumulative defined daily dose (cDDD) and average cDDD per year. We observed that compared with those obtained for HCQ nonusers (cDDD < 28), the aHRs obtained for HCQ users in the 28 ≤ cDDD < 89.92, 89.92 ≤ cDDD < 293.41, and cDDD ≥ 293.41 subgroups were 0.42 (95% CI, 0.23–0.78), 0.55 (95% CI, 0.32–0.95), and 0.44 (95%

CI 0.23–0.83), respectively. Similarly, compared with those obtained for HCQ nonusers, the aHRs obtained for the average cDDD < 18.49, 18.49 ≤ average cDDD < 61.40, and average cDDD ≥ 61.40 subgroups were 0.32 (95% CI, 0.16–0.62), 0.55

(95% CI, 0.32–0.94), and 0.61 (95% CI 0.34–1.10), respectively.

**Conclusion:** Among patients with HBV infection, HCQ use was associated with a reduced HCC risk. Further research should elucidate the underlying mechanisms.

## 海報摘要 TCR61

以關節炎及卵巢輸卵管膿瘍表現的古德氏症候群--- 個案報告

A Case of Good Syndrome with Presentation of Tubal-ovarian Abscess and Arthritis—A Case Report

葉宏明\* 林靖麒 林科名

台南市立醫院內科\* 嘉義長庚醫院內科部風濕過敏免疫內科

A 50-year-old previously healthy woman presented with fever, chills, and lower abdominal pain in April 2021, diagnosed as sepsis due to a ruptured tubo-ovarian abscess. She underwent laparoscopic surgery, including bilateral salpingo-oophorectomy and appendectomy, with postoperative treatment for a suspected fungal infection. Imaging incidentally revealed a mediastinal mass (**Fig. 1**), later identified as a type AB thymoma (Modified Masaoka Stage I), which was surgically removed without complications.

Seven months later, she developed left foot pain without systemic autoimmune or infectious symptoms. Laboratory tests were unremarkable, and autoimmune markers were negative. Ultrasound of the foot suggested talonavicular arthritis (**Fig. 2**). Initial treatment with colchicine and steroids was ineffective. Further immunological workup revealed profound hypogammaglobulinemia and a marked reduction in B and CD4 T cells, confirming a diagnosis of Good syndrome—a rare immunodeficiency associated with thymoma. She began monthly intravenous immunoglobulin (IVIG) therapy. Her thymoma remained stable, and her arthritis gradually improved with treatment.

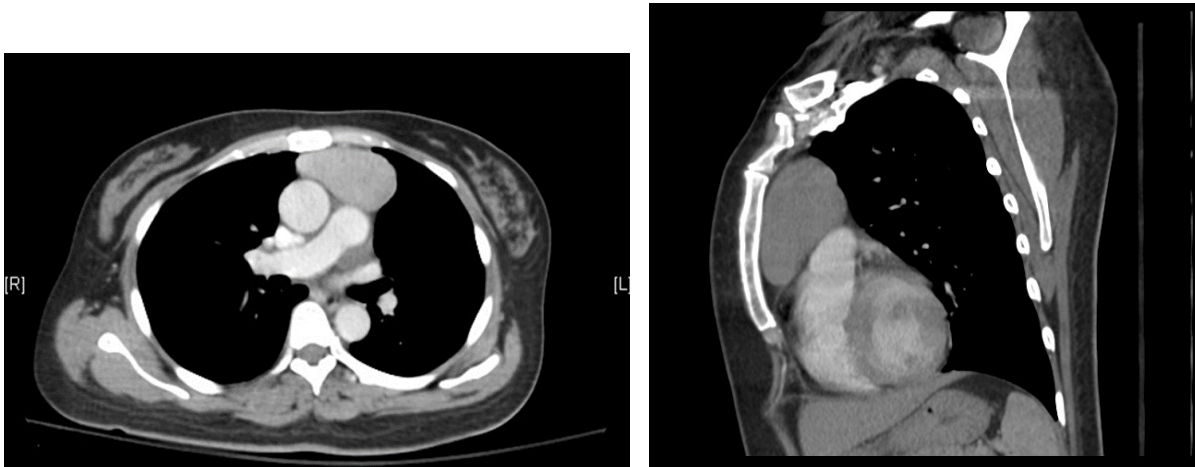


Figure.1

High attenuation mass in anterior mediastinum, up to 7.7cm with poorly enhancement and no enlarged lymph node noted.

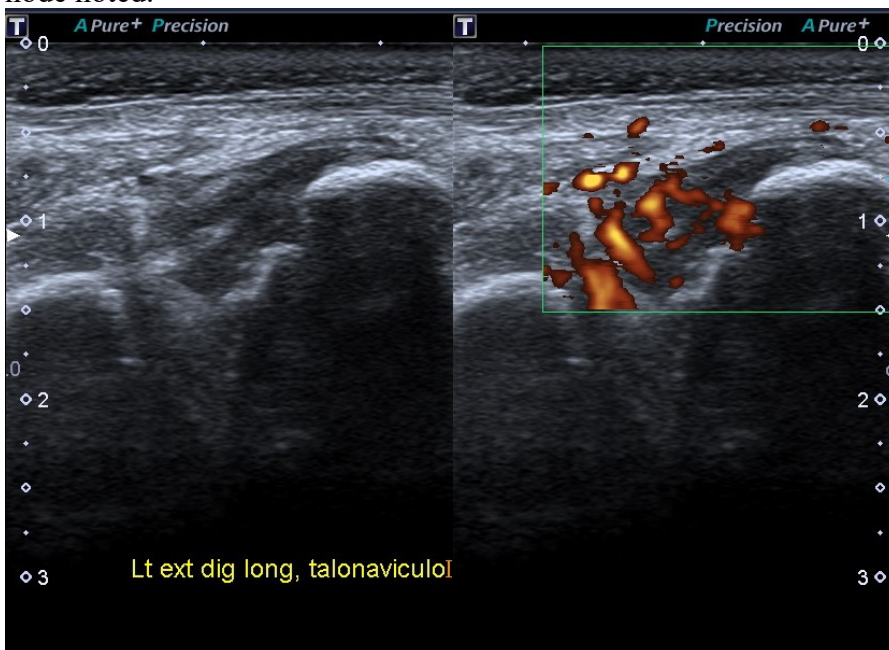


Figure 2.

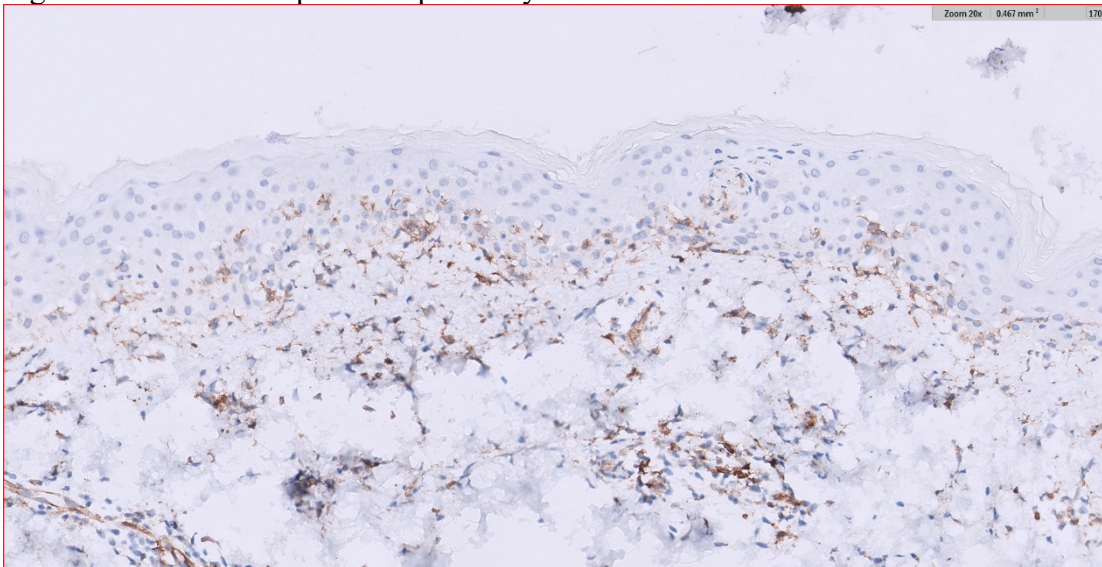
Small amount of homogeneous fluid accumulation is noted over left talonavicular joint with hypervascularity detected.

Clinical Images: Rowell syndrome

Figure 1



Figure 2 CD123-Intraepidermal plasmacytoid dendritic cells



This 60-year-old man developed a mildly pruritic skin eruption shortly after taking moxifloxacin. The rash initially appeared on the forearms and progressively spread to the face, trunk, and lower limbs, presenting as well-defined, erythematous, edematous macules and papules that coalesced into confluent plaques, sparing the palms and soles (Figure 1). Laboratory results revealed positive rheumatoid factor (15 IU/mL), anti-dsDNA (17 IU/mL), markedly elevated anti-SSA (120 U/mL), speckled-pattern ANA (1:1280, AC-4/5), elevated IgG (2370 mg/dL), and hypocomplementemia (C3 70.1, C4 6.1 mg/dL). Skin biopsy confirmed the diagnosis of **Rowell syndrome** (Figure 2), a rare condition characterized by lupus erythematosus with erythema multiforme-like lesions and a specific immunologic profile including speckled ANA, positive anti-Ro/SSA or anti-La/SSB, and positive RF.

Hung-Cheng Tsai, MD  
蔡弘正

Division of Allergy, Immunology, and Rheumatology,  
Department of Internal Medicine, Taipei Veterans General Hospital.  
台北榮民總醫院過敏免疫風濕科

## A Challenging Case of Multicentric Lymphadenopathy: Idiopathic Multicentric Castleman Disease Mimicking IgG4-Related Disease

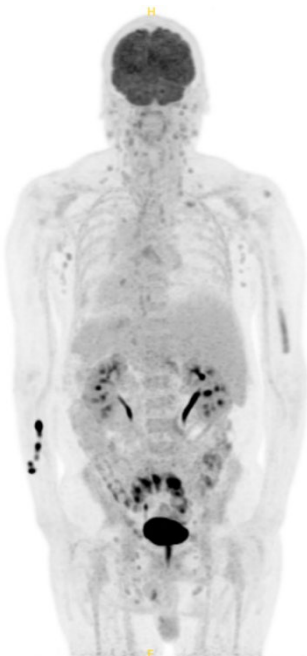
We present a 56-year-old man with a three-year history of systemic symptoms and generalized lymphadenopathy, ultimately diagnosed with idiopathic multicentric Castleman disease—not otherwise specified (iMCD-NOS), following an initial misdiagnosis of Immunoglobulin G4-related disease (IgG4-RD).

He initially presented with submandibular lymphadenopathy, fever, night sweats, and weight loss. Laboratory data revealed polyclonal hypergammaglobulinemia and an elevated serum IgG4 to IgG ratio. Lymph node biopsy revealed dense plasma cell infiltration, abundant IgG4-positive plasma cells, and an elevated IgG4/IgG ratio. Given overlapping histopathologic features, a definitive diagnosis was not possible, and IgG4-RD-directed therapy was initiated with limited response. Despite rituximab treatment, inflammatory markers remained elevated.

Follow-up evaluation revealed persistent hypergammaglobulinemia, elevated interleukin-6 and C-reactive protein (CRP) levels, thrombocytosis, and widespread hypermetabolic lymphadenopathy on positron emission tomography/computed tomography (PET/CT). A repeat biopsy showed sheets of polytypic plasma cells, a lower IgG4/IgG ratio, and absence of defining features of IgG4-RD. A final diagnosis of iMCD-NOS, plasmacytic variant, was made based on international consensus criteria. This case may represent the idiopathic plasmacytic lymphadenopathy (IPL)-type of iMCD—a proposed but unvalidated subtype increasingly recognized in recent literature.

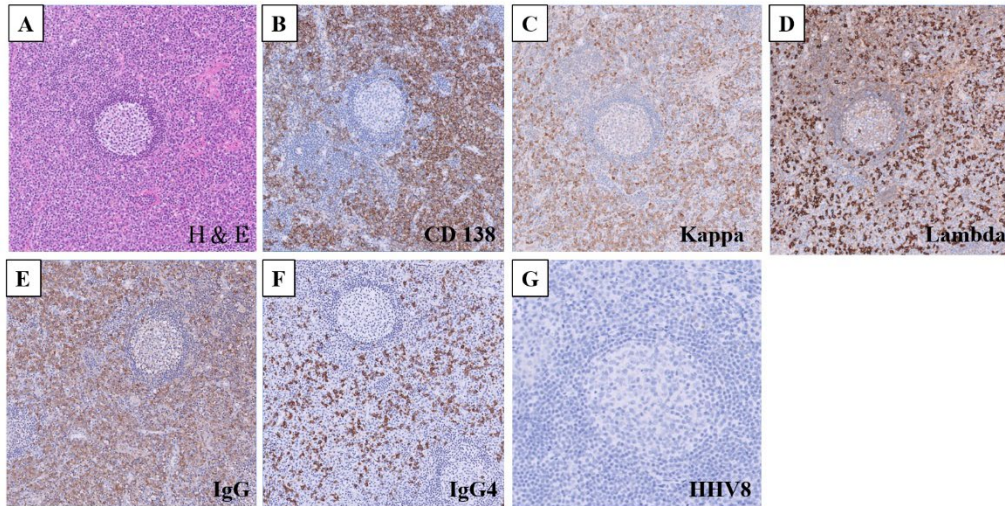
### Figure 1 FDG PET/CT Findings in iMCD

Fluorodeoxyglucose (FDG) PET/CT demonstrates diffusely hypermetabolic lymphadenopathy involving cervical, supraclavicular, axillary, mediastinal, gastrohepatic, retroperitoneal, iliac, and inguinal regions. These findings are consistent with a systemic lymphoproliferative disorder, as seen in idiopathic multicentric Castleman disease (iMCD).



## Figure 2 Histopathological and Immunohistochemical Features of Lymph Node Biopsy in iMCD

(A) Hematoxylin and eosin (H&E) staining shows sheets of grade 3 polyclonal plasma cell proliferation without regressed germinal centers, prominent follicular dendritic cells (FDCs), or increased vascularity. (10x, H&E)  
(B–D) Immunohistochemistry(IHC) highlights dense CD138-positive plasma cells with a Kappa/Lambda ratio of approximately 2:1, supporting polyclonal proliferation. (10x, IHC)  
(E–F) IgG4 immunostaining reveals up to 200 IgG4-positive plasma cells per high-power field (HPF) and an IgG4/IgG-positive cell ratio of 20%. (10x, IHC)  
(G) Human herpesvirus 8 (HHV-8) staining is negative. (20x, IHC)



Wang, Pin-Hsuan<sup>1</sup>, Tsai, Hung-Cheng<sup>1,2</sup>

王品軒, 蔡弘正

<sup>1</sup> Division of Allergy, Immunology and Rheumatology, Department of Medicine, Taipei Veterans General Hospital, Taipei, Taiwan.

<sup>2</sup> Faculty of Medicine, School of Medicine, National Yang Ming Chiao Tung University, Taipei, Taiwan

<sup>1</sup> 臺北榮民總醫院過敏免疫風濕科

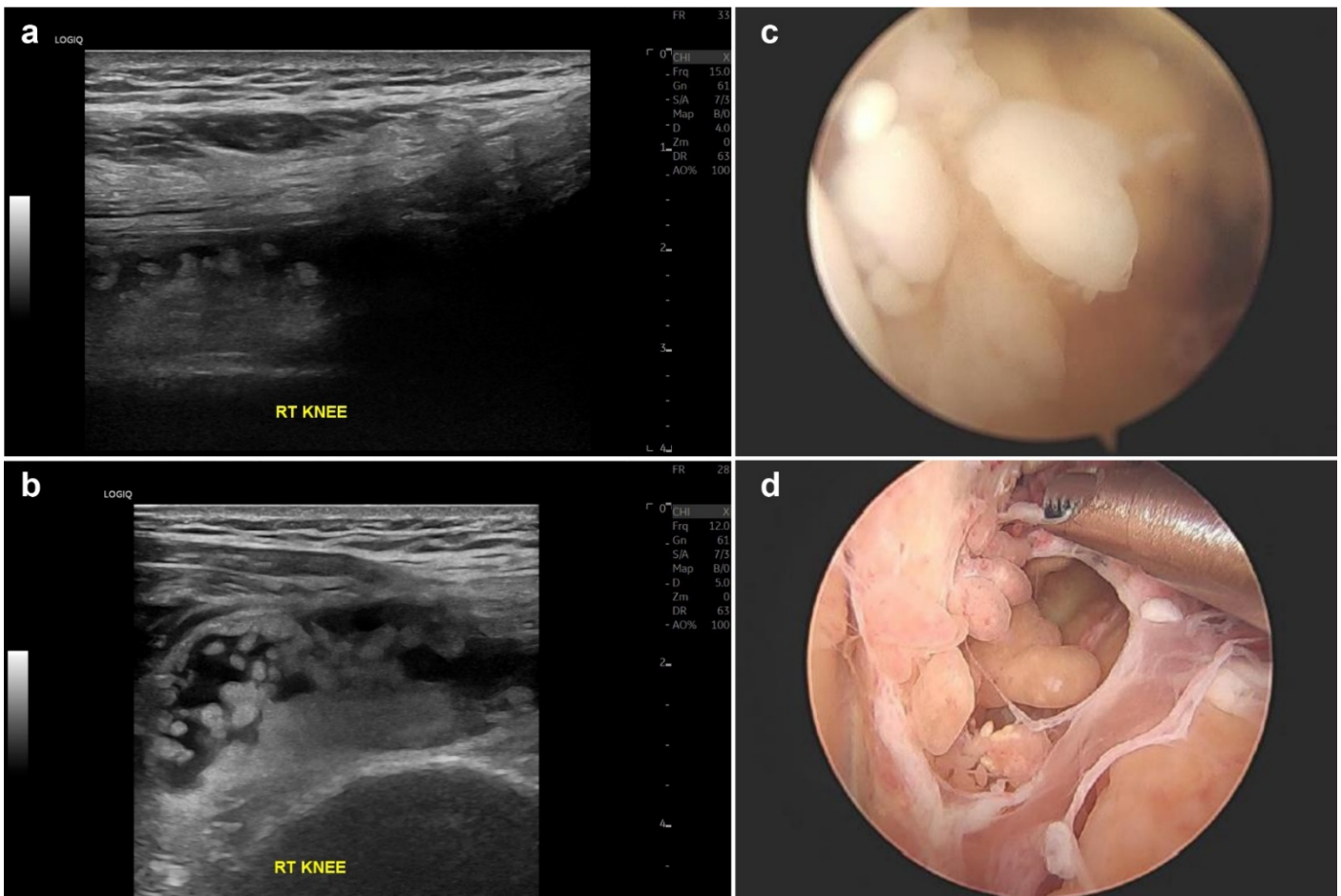
<sup>2</sup> 國立陽明交通大學醫學系

**Clinical Images: Lipoma arborescens masquerading as inflammatory synovitis**

A 62-year-old man reported recurrent swelling and soreness of the right knee for more than 6 months, with symptoms worsening after prolonged activity. Physical examination revealed mild fullness of the right suprapatellar pouch without erythema, warmth, or tenderness. Plain radiography showed mild osteoarthritis and no erosions. Inflammatory markers were only mildly abnormal (CRP 1.74 mg/dL, ESR 9 mm/hr); ANA, CTD screen, RF-IgM, and anti-CCP antibodies were negative. Musculoskeletal ultrasound demonstrated multiple iso- to hyperechoic frond-like villi and floating within an increased volume of synovial fluid (**Figures 1a, 1b**). Power Doppler imaging revealed no hypervascularity, favoring a diagnosis of lipoma arborescens rather than inflammatory synovitis. The patient underwent arthroscopic excision of the villous adipose tissue (**Figures 1c, 1d**). Histopathology confirmed synovial lipomatosis, comprising mature adipocytes surrounded by delicate fibrous septa. He has remained symptom-free since surgery.

Lipoma arborescens is a rare but characteristic, idiopathic villous lipomatous proliferation of the synovium that most often presents as a chronic, unilateral suprapatellar knee effusion. On musculoskeletal ultrasound it appears as mobile frond-like projections that show absent or negligible power Doppler flow, a finding that contrasts sharply with the hypervascular pannus of inflammatory synovitides such as rheumatoid arthritis. Recognizing these signatures enables timely, curative arthroscopic synovectomy and prevents unnecessary immunosuppressive therapy.

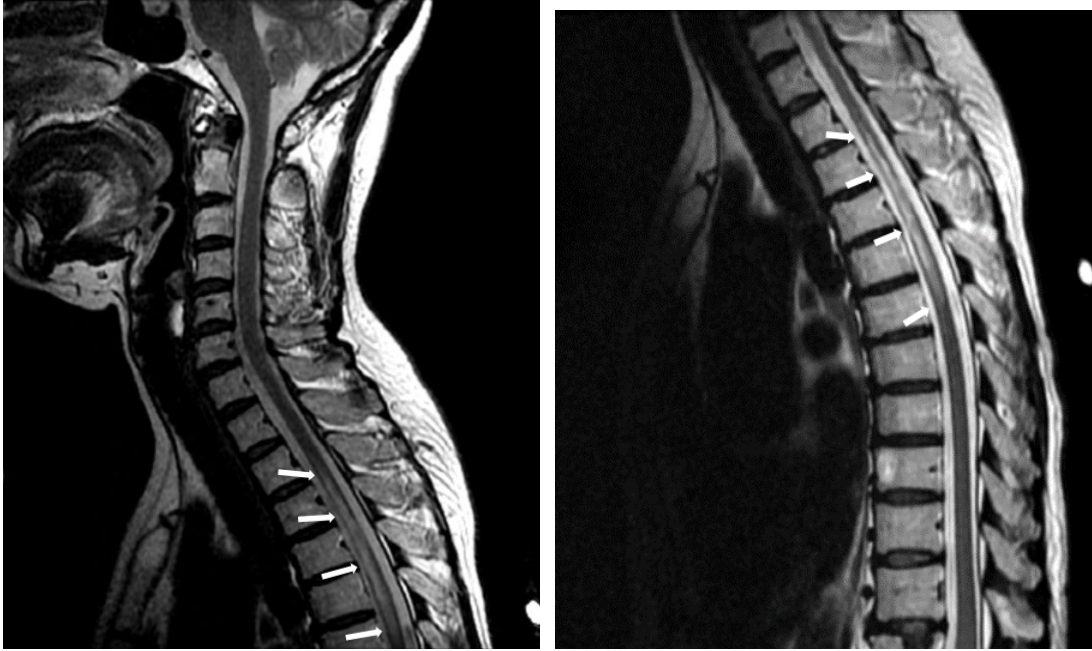
**Figure 1**



Kai-Chun Wang  
王愷君

Division of Allergy-Immunology-Rheumatology, Department of Medicine,  
Kaohsiung Veterans General Hospital, Taiwan  
高雄榮民總醫院過敏免疫風濕科

Clinical Images: Neuromyelitis Optica Spectrum Disorders in Relapse



▲ Figure A: Spine MRI 9 years ago (in June, 2016) during initial diagnosis, myelitis lesion over T2 to T6 level was labelled by arrowhead.



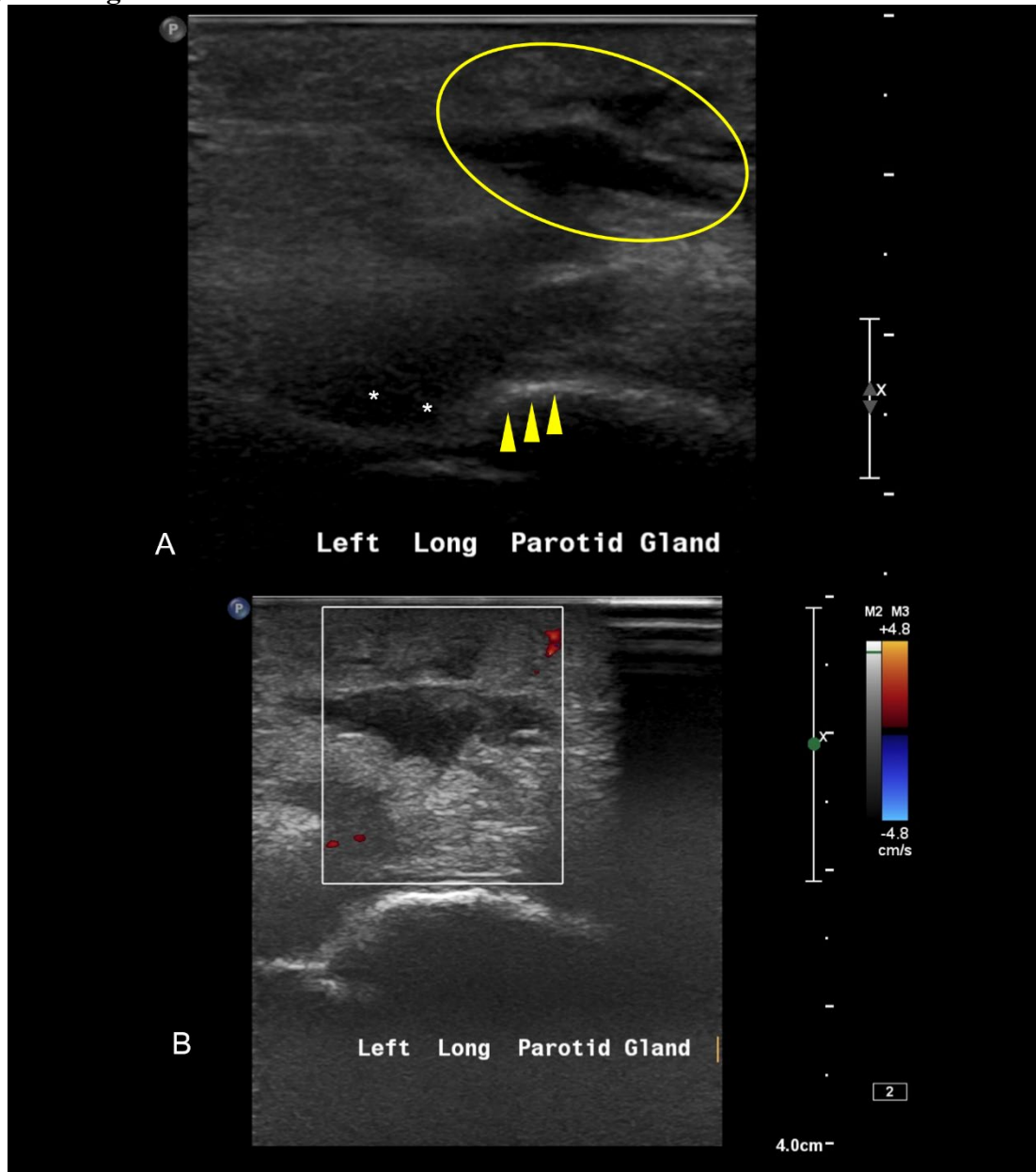
▲ Figure B: Spine MRI during this episode of relapse (in June, 2025), myelitis lesion over C2 to C6 level was labelled by arrowhead.

A 66-year-old woman with a history of multiple autoimmune diseases—including systemic lupus erythematosus (SLE), antiphospholipid syndrome, Sjögren’s syndrome, and Hashimoto’s thyroiditis—presented with a relapse of neuromyelitis optica spectrum disorder (NMOSD), which had been initially diagnosed 9 years earlier based on positive serum aquaporin-4 (AQP4) antibodies and acute myelitis. At that time, spinal MRI revealed longitudinally extensive T2-weighted hyperintense signal changes extending from T2 to T6 (Figure A). She had been maintained on oral methylprednisolone and mycophenolate mofetil since that initial episode.

The current relapse was characterized by a one-month history of numbness starting in the neck and anterior chest, gradually progressing to the trunk and all four limbs, and accompanied by gait instability prior to hospitalization. Neurological examination revealed mild proximal weakness in the lower limbs bilaterally, generalized hyperreflexia of deep tendon reflexes, preserved Babinski reflexes, impaired tandem gait, and bilateral lower limb paresthesia. Cervical spine MRI showed hyperintense T2-weighted signal changes from C2 to C6 with faint enhancement in the posterior columns, consistent with longitudinally extensive transverse myelitis. Additionally, chronic T2 hyperintensities from T2 to T5 were observed, suggesting sequelae of prior myelitis (Figure B). Serum AQP4 antibodies remained positive. The patient was treated with high-dose intravenous methylprednisolone pulse therapy without clinical improvement. She was subsequently managed with plasmapheresis followed by rituximab infusion. This case highlights that NMOSD in a patient with SLE can relapse at different levels of the spinal cord. Regular follow-up and close monitoring of neurological symptoms are essential for timely diagnosis and management.

Cheng-Han Chung, Wei-Sheng Chen  
鍾承翰, 陳瑋昇  
Taipei Veterans General Hospital, Taipei, Taiwan  
臺北榮民總醫院

**Clinical Images: Non-bacterial osteomyelitis over left mandible bone with surrounding parotitis in a 16-year-old girl**



The patient, a 16-year-old girl without underlying disease, was admitted for persistent left pre-auricular area swelling for 6 months with recent progression. She had been visited the department of dentistry, where oral antibiotic (Curam<sup>R</sup> 1g Q12H) was prescribed. Although antibiotic therapy was administered, the symptoms progressed. On physical exam, a firm mass (5x5 in size) with swelling, local erythema, tenderness was noticed over right pre-auricular area. She had difficulty opening mouth, which lead to decreased intake, while there's no fever, no weight loss nor trauma history. Lab data showed leukocytosis (11100/uL), anemia (10.8g/dL) and mild elevated CRP levels (17.9 mg/L). Ultrasound examination of the parotid gland showed heterogenous hyper-echogenicity with cystic formation (Fig. A circle) and surrounding fibrotic changes with positive PD signal over left parotid gland (Fig. B). Hyperechogenicity over the ramus part of left mandible (Fig. A arrow heads) with hypoechoic changes adjacent to the cortex (Fig. A asterisks) was also noticed. CT scan demonstrated ground-glass appearance of the left mandibular body/ramus and periosteal reaction with lytic foci as well as enlarged left parotid gland with increased enhancement as well as inflammation of subcutaneous soft tissue. CT-guided biopsy of left mandible was performed and the pathology showed no evidence of granuloma nor malignancy but fibrosis with inflammatory cells infiltration. The patient was diagnosed with chronic nonbacterial osteomyelitis and treated with NSAIDs, colchicine and prednisolone. After 3 months of treatment, her symptoms improved a lot and follow-up ultrasound showed normalized appearance of left parotid gland and mandible, while fibrotic changes and biopsy scarring over left mandible cortex.

### Conclusion:

Ultrasound serves as a valuable tool assessing and visualizing inflammatory changes over parotid glands as well as mandible bone<sup>1-3</sup>. Besides, it is non-invasive without the concerns of radiation exposure and sedation among pediatric patients. Finally, we can use ultrasound to follow up patient's inflammation conditions of the parotid gland, mandible bone after the treatment.

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Chi-Teng, Tseng, MD-MPH;

Yeong-Jian, Jan Wu, MD-PhD;

Jing-Long, Huang, MD-PHD

曾吉騰, 吳詹永嬌, 黃璟隆

Chang Gung Memorial Hospital, Keelung

Chang Gung Memorial Hospital, Linkou, Taiwan

基隆長庚紀念醫院, 林口長庚紀念醫院

## 海報摘要 TCR68

### Granulomatosis with Polyangiitis Presenting as Hypertrophic Pachymeningitis

#### 肉芽腫性多血管炎：以肥厚性硬腦膜炎為臨床呈現

Hsiang-Yen Lee<sup>1,2</sup>, Kai-Leun Tsai<sup>2</sup>, Yu-Chen Chu<sup>1</sup>, Wei-Jui Lin<sup>1</sup>, Ke-Yu Chang<sup>1</sup>, Tzu-Min Lin<sup>1,3</sup>, Chi-Ching Chang<sup>1,3</sup>

李向嚴<sup>1,2</sup> 蔡凱倫<sup>2</sup> 朱有晨<sup>1</sup> 林韋睿<sup>1</sup> 張克宇<sup>1</sup> 林子閔<sup>1,3</sup> 張棋楨<sup>1,3</sup>

1 Division of Allergy, Immunology, and Rheumatology Taipei Medical University Hospital

2 Division of Allergy, Immunology, and Rheumatology Taipei Hospital, Ministry of Health and Welfare

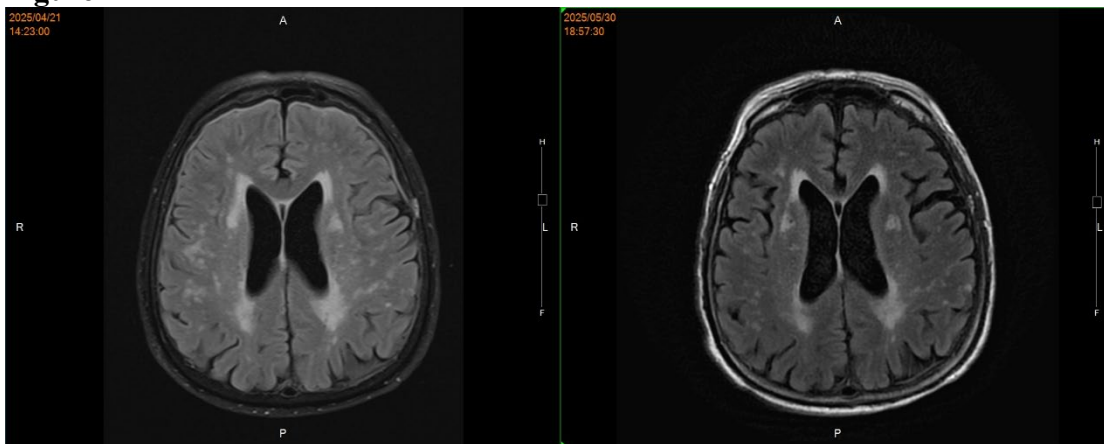
3 Division of Allergy, Immunology and Rheumatology, Department of Internal Medicine, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan

1 台北醫學大學附設醫院過敏免疫風濕科

2 衛生福利部臺北醫院過敏免疫風濕科

3 台北醫學大學醫學系過敏免疫學科

### Figure 1



(1a) Pre-treatment MRI demonstrating hypertrophic pachymeningitis. There is diffuse pachymeningeal thickening and homogeneous enhancement along the cerebral convexities and skull base. Notably, the dura of the left tentorium cerebelli and left frontotemporal region is markedly thickened and enhanced, indicating extensive pachymeningeal involvement. No discrete parenchymal mass is evident. These imaging findings were consistent with an aseptic meningitic process (hypertrophic pachymeningitis) rather than an encapsulated abscess or neoplasm.

(1b) Follow-up MRI after rituximab therapy showed marked radiologic improvement. The pachymeningeal enhancement has substantially regressed; the previously thickened dura exhibits only faint residual enhancement and reduced thickness. Comparison of the pre- and post-treatment images demonstrates near-complete resolution of the dural inflammatory changes, especially at the tentorium and frontal dura. This correlates with a successful therapeutic response. Such radiologic resolution of hypertrophic pachymeningitis after therapy has been reported in the literature. In our patient, the resolution of dural enhancement paralleled his clinical improvement.

Figure 2

Figure 2: Histopathology of the dural biopsy (H&E, 400×). Granulomatous inflammation with central necrosis (N) and surrounding chronic inflammatory cells is seen. A multinucleated giant cell is present at the periphery of a necrotic focus, and adjacent reactive lymphocytes and plasma cells form a rim around the necrotic center. The arterial wall shows fibrinoid necrosis and inflammation, indicative of a vasculitic process. These features of necrotizing granulomas and vasculitis are characteristic of GPA. Immunohistochemistry further supported the diagnosis: CD68 stain highlighted abundant macrophages within the granulomas (confirming the histiocytic component), and IgG4 immunostaining demonstrated only rare IgG4-positive plasma cells. The IgG4+/IgG+ plasma cell ratio was approximately 20%, which is well below the 40% threshold considered suggestive of IgG4-related disease. The presence of neutrophils and necrosis on the biopsy, together with the low IgG4/IgG ratio, favored GPA over IgG4-related pachymeningitis

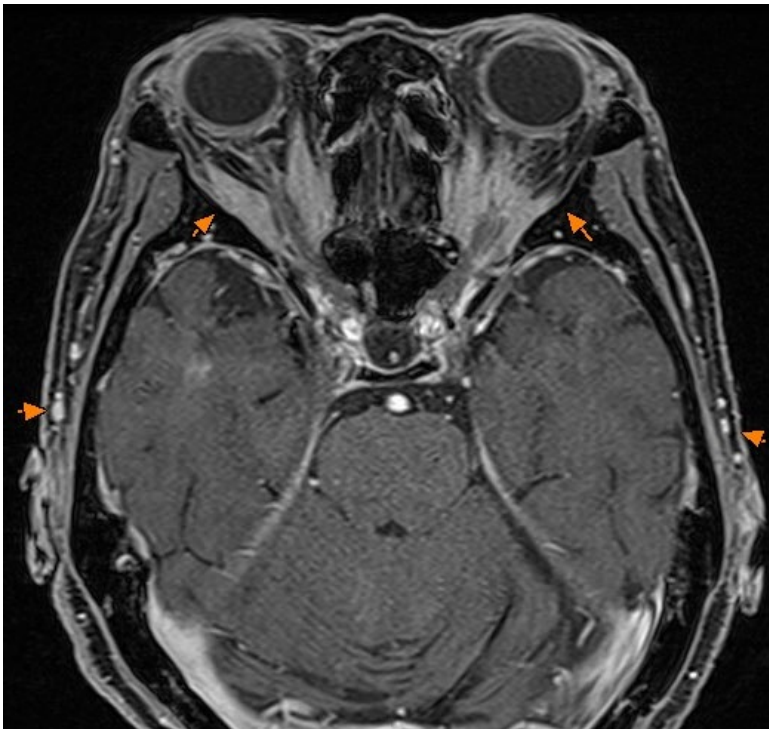
海報摘要 TCR69

**Clinical Images: Acute vision loss due to inflammatory-mediated immune disease**

A. Patient with giant cell arteritis (GCA), complicated with arteritic anterior ischemic optic neuropathy (AAION)



B. Patient with Graves' disease, complicated with dysthyroid optic neuropathy (DON)



Acute vision loss is a medical emergency frequently encountered in rheumatologic practice. Patient A, an 84-year-old woman, presented with new-onset right temporal pain followed by sudden right eye vision loss, later progressing to blurred vision in the left eye. Patient B, an 80-year-old woman, had progressive right eye vision loss for 5 months, followed by left eye involvement. Both were referred by ophthalmologists. Examinations revealed optic disc edema and relative afferent pupillary defect (RAPD), suggestive of arteritic anterior ischemic optic neuropathy (AAION) or optic neuritis. Patient A was diagnosed with giant cell arteritis; MRI showed mural enhancement and wall thickening of bilateral superficial temporal arteries with mildly enlarged extraocular muscles. Patient B was diagnosed with Graves' disease with dysthyroid optic neuropathy; MRI showed mild thickening of the superficial temporal arteries and asymmetric enlargement with enhancement of extraocular muscles. Both received IV methylprednisolone 500 mg/day for 3 days. Patient A was continued on tocilizumab, and patient B on rituximab. Both regained left eye vision, and light perception returned in the previously blind right eye.

Shih-Hsun, Lan, MD

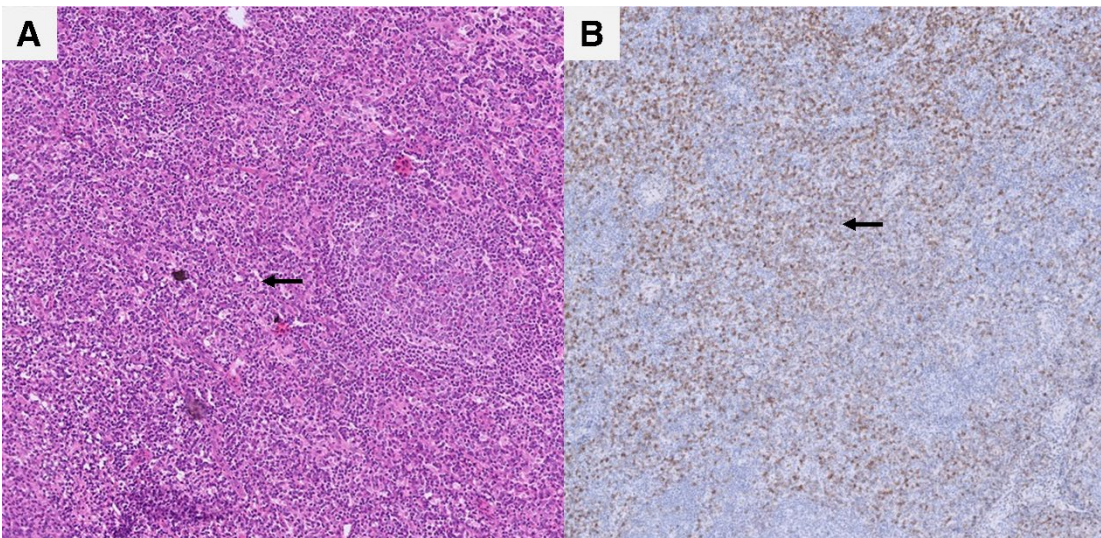
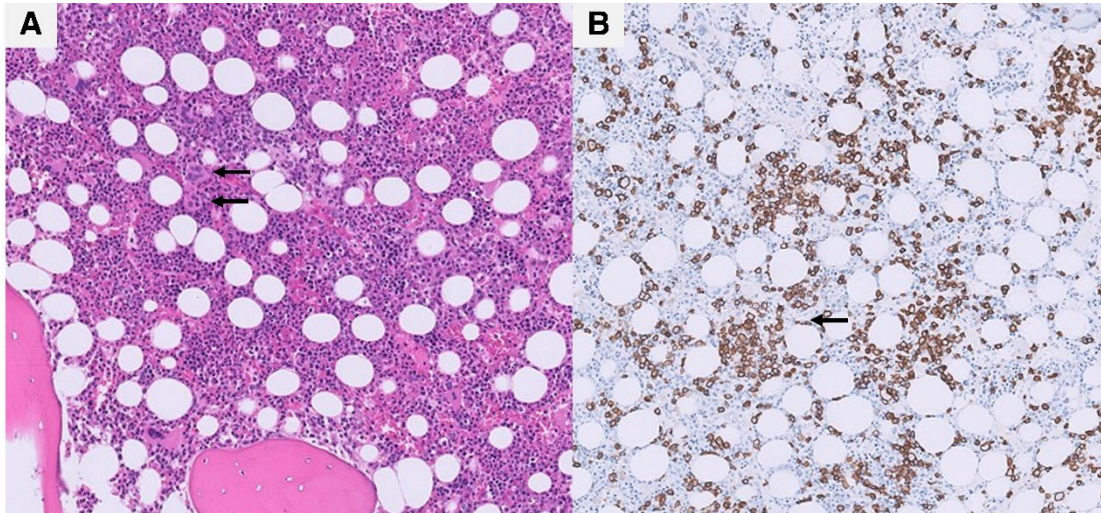
藍士勛

National Taiwan University Hospital

Yunlin Branch, Taiwan

台灣大學附設醫院雲林分院

Clinical Images: idiopathic multicentric Castleman disease



A 57-year-old man presented with asymptomatic non-palpable purpura over both legs since May 2024. He was treated with oral prednisolone, colchicine, and topical corticosteroids but subsequently developed poor appetite, unintentional weight loss (5 kg in 10 days), night sweats, dyspnea on exertion, and diarrhea. Laboratory evaluation revealed hypocomplementemia (low C3/C4) and a monoclonal protein pattern (IgG/Kappa) on serum immunoelectrophoresis, while autoimmune serologies (ANCA, ANA, myositis panel) and hepatitis studies were negative. Bone marrow examination showed hypercellularity with plasmacytosis and granulomas without evidence of neoplasm (black arrow, Figure 1). A skin biopsy of the right leg demonstrated leukocytoclastic vasculitis. CT-guided biopsies of abdominal lymph nodes performed in July 2024 and March 2025 revealed plasmacytosis without malignancy. Excisional biopsy of a right neck lymph node showed plasmacytosis with up to 200 IgG4-positive cells/HPF and an IgG4/IgG ratio of 25% (black arrow, Figure 2), fulfilling the minimum histopathological criteria for idiopathic multicentric Castleman disease (iMCD), though other inflammatory disorders could not be completely excluded. After multidisciplinary review and exclusion of alternative diagnoses, a final diagnosis of iMCD was established, and IL-6 inhibitor therapy is pending approval.

Ping-Hsuan Kuo, MD, Chien-Chih Lai, MD, Ph.D., Yen-Po Tsao, MD, Ph.D., Wei-Sheng Chen, MD, Hsien-Tzung Liao, MD, Ph.D., Ming-Han Chen, MD, Ph.D. Yi-Syuan Sun, MD, Taipei Veterans General Hospital, Taipei, Taiwan.

郭蘋萱, 蔡弘正, 賴建志, 曹彥博, 陳瑋昇, 廖顯宗, 陳明翰, 孫易暄  
臺北榮民總醫院

## 海報摘要 TCR71

### Auto-antibody testing facilitating diagnosis of oral-limited pemphigus: a case report.

#### 自體抗體檢測協助診斷口腔侷限型天皰瘡：病例报告

Li-heng Huang<sup>1</sup>

<sup>1</sup> Department of internal medicine, division of allergy, immunology and rheumatology,  
National Taiwan University Hospital Yunlin branch

黃立恒<sup>1</sup>

<sup>1</sup> 台大醫院雲林分院風濕免疫科

### Case presentation

An elderly man was referred to our rheumatology clinic by an otolaryngologist for evaluation of painful oral and buccal ulcers accompanied with a few lip vesicles for two-months. Physical examination revealed no Nikolsky sign, palpable cervical lymphadenopathy or other skin eruption. Laryngoscopy showed no evidence of mass lesion. Laboratory results demonstrated elevated inflammatory markers: C-reactive protein was 4.4 mg/dL, and erythrocyte sedimentation rate was 73 mm/hr. Antinuclear anti-body (ANA) testing was positive at the titer of 1:1280 with a homogeneous (AC-1) pattern. Microbiological cultures, serum IgG, extractable nuclear antigen panel (anti-ENA), anti-dsDNA, intercellular substance (ICS) antibodies, and anti-basement membrane zone antibodies were all within normal limits or non-reactive.

Because of his financial constraints, concerns regarding glucocorticoid usage and hesitation toward tissue sampling, therapeutic trial with Colchicine, Hydroxychloroquine (400mg daily) and Methotrexate (10mg weekly) were first initiated. The response, however, was limited. Following multi-disciplinary discussion, additional advanced autoantibody testing was pursued, and an incisional biopsy was performed by the referring ENT specialist. Three weeks later, anti-desmoglein-3 (anti-DSG3) antibody returned positive (59.2RU/mL, reference <20). Anti-DSG1 and anti-BP180 antibody were negative. Histopathology revealed neutrophil-rich debris, although direct immunofluorescence cannot be performed due to severe destruction. Subsequently, he agreed to initiate a moderate dose of glucocorticoid. His lesions gradually resolved, and inflammatory markers improved.

### Discussion

Oral ulcers are frequently encountered in our clinic. This case highlights that certain patients with bullous diseases may have lesions confined solely to the oral cavity (with or without vesicles on the lips), in the absence of extra-oral involvement. Nikolsky sign may not be present in such cases. Multidisciplinary collaboration is essential for comprehensive assessment of clinical, serological, and pathological aspects. When pursuing biopsy, performing it at an earlier stage may increase diagnostic yield. Additionally, anti-desmoglein antibody testing can provide supportive evidence in establishing the diagnosis.

## Upadacitinib as Rescue Therapy for Refractory Hepatitis in Adult-Onset Still's Disease: A Case Report

Yeong-Jang Lin, Hung-An Chen, Chao-Yu Chen, Yu-Ting Tseng

Division of Allergy, Immunology, and Rheumatology, Chi Mei Medical Center

Upadacitinib 作為成人型史迪爾氏症難治性肝炎的救援治療：病例報告

林永章，陳宏安，陳昭宇，曾毓婷

奇美醫學中心過敏免疫風濕科

### Case Presentation

A 42-year-old man with a history of hypertension was admitted with daily spiking fevers (up to 40 °C) accompanied by chills for one month. He reported a transient, non-pruritic pink rash over the trunk and extremities, which appeared during febrile episodes. Ten days prior to admission, he developed a sore throat. He denied weight loss, night sweats, recent travel, chest or abdominal pain, arthralgia, dyspnea, or dysuria. Previous evaluations at other institutions failed to establish a diagnosis; tests for COVID-19, influenza, and dengue fever were negative.

On physical examination, mild oropharyngeal erythema without exudate was observed. There was no lymphadenopathy or hepatosplenomegaly. Laboratory tests revealed: WBC 13,300/ $\mu$ L (neutrophils 80.5%), hemoglobin 12.7 g/dL, platelets 202,000/ $\mu$ L, CRP 95.1 mg/L, ferritin >40,000 ng/mL, AST 396 U/L, and ALT 210 U/L. ANA, rheumatoid factor, and blood cultures were negative. Chest radiography was unremarkable.

A diagnosis of adult-onset Still's disease (AOSD) was made according to the Yamaguchi criteria, fulfilling three major criteria (fever  $\geq 39$  °C for  $\geq 1$  week, evanescent rash, neutrophilic leukocytosis) and three minor criteria (sore throat, abnormal liver function tests, negative ANA and RF). Infectious, neoplastic, and alternative autoimmune etiologies were excluded through comprehensive investigations.

The patient was started on intravenous methylprednisolone (40 mg every 8 hours), which resulted in rapid resolution of fever, rash, sore throat, and CRP elevation. However, despite clinical improvement, his transaminase levels progressively increased, with AST rising to 2,103 U/L and ALT to 3,409 U/L. Jaundice developed, with total bilirubin peaking at 18.9 mg/dL. Ferritin levels remained >40,000 ng/mL. Abdominal ultrasonography showed no biliary tract dilatation. Extensive workup—including Weil-Felix, CMV, EBV, and toxoplasma serologies, anti-smooth muscle, anti-parietal cell, anti-mitochondrial antibodies, viral hepatitis markers, HIV, and ceruloplasmin—was unremarkable. Liver biopsy demonstrated active hepatitis without features suggestive of autoimmune hepatitis or other specific etiologies.

Due to persistent hepatic dysfunction, high-dose pulse methylprednisolone (750 mg/day for 3 days) was administered without improvement. Subsequently, upadacitinib (15 mg/day) was initiated. This JAK inhibitor led to gradual improvement in liver enzymes and ferritin levels, with sustained clinical and biochemical stabilization on follow-up.

## Immunomodulatory Effects of Molecular Hydrogen Therapy in Complex Autoimmune Diseases: A Case Study on Changes in Tr1 Cells, Breg Cells, and TIM3 Expression

Jing-Yuan Chen<sup>1</sup>, Shan-Wen Lui<sup>2</sup>, Ting-Yu Hsieh<sup>3</sup>, Yi-Jung Ho<sup>4</sup>, Hsin-Ling, Hsieh<sup>5</sup>, Feng-Cheng Liu<sup>5</sup>

陳靖元<sup>1</sup> 呂善玟<sup>2</sup>, 謝庭仔<sup>3</sup>, 何怡蓉<sup>4</sup> 謝心玲<sup>5</sup>, 劉峰誠<sup>5</sup>

<sup>1</sup> Department of Internal Medicine, Tri-Service General Hospital, National Defense Medical Center, Taipei 114, Taiwan, R.O.C.

<sup>2</sup> Department of Internal Medicine, Linkou Chang-Gung Memorial Hospital, Taoyuan, Taiwan, R.O.C.;

<sup>3</sup> Department of Internal Medicine, Taichung Veterans General Hospital, Taichung, Taiwan, R.O.C.;

<sup>4</sup> School of Pharmacy, National Defense Medical Center, Taipei 114, Taiwan, R.O.C.

<sup>5</sup> Rheumatology/Immunology/Allergy, Tri-Service General Hospital, National Defense Medical Center, Taipei 114, Taiwan, R.O.C.

國防醫學院三軍總醫院風濕免疫過敏科

### Background:

Aneurysmal subarachnoid hemorrhage (SAH) complicated by autoimmune diseases presents significant therapeutic challenges, particularly when conventional treatments fail to achieve adequate disease control. Molecular hydrogen therapy has emerged as a promising adjuvant treatment modality due to its anti-inflammatory and immunomodulatory properties. This study aims to investigate the immunomodulatory effects of molecular hydrogen therapy on regulatory immune cell populations in a complex case of aneurysmal SAH with concurrent rheumatoid arthritis (RA) and systemic lupus erythematosus (SLE).

### Materials and Methods:

A 44-year-old female patient with aneurysmal SAH, pre-existing RA, and newly diagnosed SLE complicated by acute ischemic infarction was treated with daily molecular hydrogen capsules (170 mg hydrogen-rich coral calcium equivalent to  $1.7 \times 10^{21}$  hydrogen molecules per capsule) for six months. Comprehensive flow cytometric analysis was performed to evaluate changes in immune cell phenotypes, including T regulatory type 1 (Tr1) cells, regulatory B (Breg) cells, and TIM3 expression on cytotoxic T cells (Tc cells). Clinical parameters, autoantibody titers, and neurological assessments were monitored throughout the treatment period.

### Results:

Following six months of molecular hydrogen therapy, significant improvements were observed in both clinical outcomes and immune markers. The patient demonstrated progressive neurological recovery with Glasgow Coma Scale improvement from E3M2VT to E4M6VT, and muscle strength enhancement across all limbs. Flow cytometric analysis revealed increased TIM3<sup>+</sup> expression on Tc cells (baseline: 8.2% to 6-month: 15.7%), elevated Breg cell populations (baseline: 2.1% to 6-month: 4.8%), and restoration of Tr1 cell levels that correlated with clinical improvement. Anti-dsDNA antibody converted from positive to negative, and RA remained well-controlled despite discontinuation of tofacitinib. No adverse effects were reported during the treatment period.

### Conclusion:

Molecular hydrogen therapy demonstrates significant potential as an adjuvant treatment for complex autoimmune conditions complicated by neurological injury. The therapy effectively modulated key regulatory immune cell populations, particularly Tr1 cells and Breg cells, while enhancing TIM3 expression on cytotoxic T cells, suggesting improved immune tolerance and anti-inflammatory responses. These findings warrant larger-scale clinical trials to validate the therapeutic efficacy of molecular hydrogen in autoimmune diseases with neurological complications.

## 海報摘要 TCR74

### A Case of Primary Antiphospholipid Syndrome Complicating Budd-Chiari Syndrome Presenting as Acute Abdomen and Fever

K.S. Tseng, C.C. Wang,

Tao-Yuan General Hospital, Division of Rheumatology, Immunology and Allergy

原發抗磷脂症候群併發巴德-吉亞利症候群呈現腹痛和發燒

曾國森、王淳峻

衛生福利部桃園醫院 風濕免疫過敏科

**Introduction:** Antiphospholipid syndrome (APS) can cause arterial or venous thrombosis and complications of pregnancy. Budd-Chiari syndrome (BCS) is a rare complication in APS. We encountered an APS patient presented with fever and acute abdomen, BCS was suspected from the findings on MRI study with elevation of thrombotic marker. Early initiation of anticoagulant therapy achieved a good outcome.

**Case presentation:** A 24-year-old male, was admitted for investigation of fever and abdomen pain intermittently for three months. PE, temperature 38.9°C, no icteric sclera, no palpable LNs, tenderness in the epigastric region. Labs revealed WBC  $4.8 \times 10^9/L$ , Hgb: 11.7 g/dL, platelet was  $117 \times 10^9/L$ , PT:14.5sec (N)/APTT:111.7sec, D-Dimer:3.37 ug/ml (<0.5), CRP: 5.5mg/dl. Biochemistry :normal liver enzymes with no kidney dysfunction. ANA 1:640 (speckled), lupus anticoagulant LA1/LA2 ratio :3.05(H), cardiolipin antibodies and anti- $\beta_2$  GPI antibody(-). Cultures were negative. MRI reported compatible with budd-chiari syndrome. Anticoagulant therapy with warfarin was prescribed, then fever and abdominal pain resolved. After discharge follow up labs showed normal D-Dimer /CRP and free of symptoms under warfarin use alone.

**Conclusion:** Our patient had fever and evidence of marked inflammation. It was ruled out etiologies such as infection, BCS was difficult to diagnose. It is suggested that extensive thrombosis and vascular endothelial damage induced inflammatory reaction. Our experience suggests that anticoagulant therapy can be effective in the early stage of BCS associated with APS. If BCS is suspected in patients with APS, contrast-enhanced MRI of the hepatic/portal veins should be performed promptly and followed by early initiation of adequate anticoagulant.

## Diagnostic and therapeutic Challenges in Autoimmune Hemolytic Anemia with Underlying Systemic Autoimmunity and Dual Malignancies

Shih-Hsun, Lan

Division of Rheumatology, National Taiwan University Hospital Yunlin Branch

潛藏系統性自體免疫與雙重惡性腫瘤之自體免疫性溶血性貧血的診斷與治療挑戰

藍士勛

風濕免疫科, 台灣大學附設醫院雲林分院

### Background:

Autoimmune hemolytic anemia (AIHA) may arise from systemic autoimmunity or malignancy. While bilirubin elevation is common, total bilirubin (T-bil) >40 mg/dL is rare and may reflect both hemolysis and biliary obstruction.

### Case Presentation:

A 64-year-old woman presented with mixed-type AIHA and persistent jaundice. Workup revealed positive ANA, low complements, and antiphospholipid antibodies. She denied any other discomfort, including changes in bowel habits, vaginal spotting, weight loss, or night sweats. She received mycophenolate and belimumab for suspected systemic lupus erythematosus (SLE). She had one episode of right flank pain and admitted for further evaluation. Cecal cancer was later found and resected, with stable disease control. She developed another episode of right flank pain and marked T-bil elevation (peak at 63 mg/dL). Imaging revealed a common bile duct (CBD) stone and suspicious local recurrence of cecal cancer. Endoscopic retrograde cholangiopancreatography (ERCP) with stent placement and high-dose steroids led to rapid resolution of jaundice and hemolysis. Biopsy of a suspicious local recurrence tumor later confirmed low-grade B-cell lymphoma. Rituximab (R)-mini-CHOP chemotherapy achieved remission.

### Discussion:

This case underscores the importance of recognizing dual causes of severe hyperbilirubinemia in AIHA. Timely ERCP and immunosuppression reversed both hepatic and hematologic dysfunction. The sequential discovery of two malignancies highlights the need for thorough evaluation in atypical AIHA. Multidisciplinary care was essential for a favorable outcome.

