A rare case of curative colectomy for Takayasu's arteritis

Sirs,

Takayasu's arteritis (TAK) is a large-vessel vasculitis (LVV) characterised by granulomatous inflammation of the aorta and its major branches. Inflammatory bowel disease (IBD) encompasses Crohn's disease and ulcerative colitis (UC). The co-occurrence of TAK and IBD is notably rare with under 200 reported cases (1, 2).

A 52-year-old female of American Indian descent with prior pulmonary embolism and family history of UC presented with pleuritic chest pain, diaphoresis, and rectal bleeding. She initially took 1200 mg of ibuprofen frequently for her chest pain. The patient denied abdominal pain, dizziness, fever, or fatigue. Physical exam demonstrated chest wall tenderness and 2+ peripheral pulses in all extremities. She was febrile to 38.3°C and tachycardic to 201 beats per minute. Initial workup revealed anaemia (10.7 g/dL), leucocytosis (15.7 10^3L), C-reactive protein (CRP) of 338 mg/L, and erythrocyte sedimentation rate (ESR) of 117 mm/hr. Antinuclear antibody titre was 1:1280 with a homogenous pattern. The remaining workup was unrevealing. Computed tomography (CT) pulmonary angiogram demonstrated aortitis in the ascending and descending aorta (Fig. 1A). Temporal artery biopsy was negative. She was initially diagnosed with giant cell arteritis (GCA) and started prednisone 60 mg. Follow-up imaging demonstrated resolution of the aortitis with normalisation of ESR and CRP. She received one month of 40 mg prednisone but self-discontinued thereafter. Despite discontinuing non-steroidal anti-inflammatory drugs (NSAIDS), her haematochezia persisted with progressive anaemia (haemoglobin 8.7 g/dL). Upon prednisone discontinuation, chest pain returned. Repeat imaging demonstrated inflammation of the ascending and descending thoracic aorta, proximal left carotid artery, and brachiocephalic artery, a distribution more consistent with TAK (Fig. 1B). Given ongoing haematochezia, colonoscopy was performed showing severe mucosal changes consistent with UC (Fig. 1C). This patient was then unfortunately lost to follow-up for 11 months before being hospitalised for progressively severe anaemia (haemoglobin 6.5 g/dl), at which time she underwent urgent subtotal colectomy with end ileostomy. Two months after surgery, follow-up positron emission tomography-CT showed complete aortitis resolution, off all immunosuppressive therapy (Fig. 1D). The patient has remained symptom-free without any immunosuppression for either TAK or UC.

This case is valuable as it highlights the rare co-occurrence of TAK and IBD, as well as the novel remission of her disease following colectomy. Due to this patient's family history of UC, her bowel inflammation likely preceded the TAK. Delayed UC diagnosis was likely due to NSAID use confounding the initial rectal bleeding. Additionally, repeated steroid use improved her abdominal symptoms, which caused them to be misattributed to TAK.

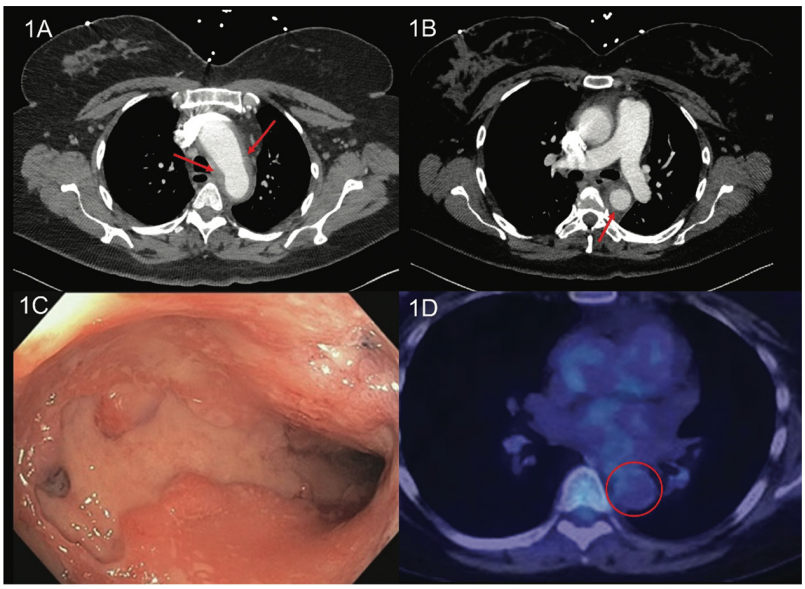

Fig 1. A: CT angiogram, February 2019: Arrows pointing to aortic wall thickening involving the aortic arch and proximal great vessels, which extends into the descending thoracic aorta. **B**: CT angiogram, June 2019: arrow pointing to thickening of the thoracic aorta that extends into the proximal left common carotid artery and right brachiocephalic artery. **C**: Colonoscopy, July 2019: severe mucosal changes and ulcerations. **D**: Positron emission tomography-computed tomography fusion, axial plane, October 2020: red circle indicating the descending thoracic aorta. No evidence of increased fluorodeoxyglucose avidity for any arterial segment with normal uptake in area of prior wall thickening, suggesting resolution of prior aortitis.

The diagnosis of TAK itself was also unique in this case. The patient initially was diagnosed with GCA despite negative temporal artery biopsy. This was reasonable given that temporal artery biopsy has a sensitivity of 77.3% and the accuracy of testing is dependent on the sample obtained, processing, and interpretation (3). In fact, recent guidelines suggest that ultrasound should instead be the primary imaging modality for GCA diagnosis (4). The patient was also of American Indian descent, which is a population infrequently affected by TAK. Generally, TAK occurs in Asian women under the age of 40 (our patient was 52) (5-6). The patient also showed no pulselessness or bruits on exam, so it was not until imaging evidence of LVV that the diagnosis of TAK was made.

Regarding treatment, most patients who experience TAK and IBD require ongoing immunosuppression to manage their vasculitis after colectomy (7-8). Our case did not follow this pattern. Thus, to our knowledge, is the first case in which a colectomy for IBD was curative for TAK. This demonstrates a unique association between IBD and LVV, for which further investigation is warranted to understand possible common aetiopathogenetic mechanisms.

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