CASE REPORT

Takayasu's or tuberculous arteritis?

Carlos Moura,¹ Mauricio Antonio Aquino,¹ Jose Rocha Filho,¹ Mittermayer Santiago²¹Hospital Santa Izabel, Salvador, Brazil²Serviços Especializados em Reumatologia, Salvador, Brazil**Correspondence to** Professor Mittermayer Santiago, mbsantiago2014@gmail.com

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SUMMARY

Takayasu's arteritis (TA) is a large vessel vasculitis of unknown aetiology characterised by involvement of the aorta and its major branches. Tuberculous arteritis of the aorta is an uncommon condition usually secondary to the dissemination of *Mycobacterium tuberculosis* infection from the mediastinum and/or lung to the adjacent aorta; this may mimic TA. We report a case of a 23-year-old woman with cutaneous granulomatous vasculitis and saccular aneurysmal dilation of the aorta and large vessels, and highlight the findings shared by TA and tuberculous arteritis.

BACKGROUND

Takayasu's arteritis (TA) is a large vessel vasculitis of unknown aetiology characterised by a chronic granulomatous panarteritis of the aorta and its major branches. It preferentially affects women before age 40 years.¹ In a genetically predisposed patient, the pathogenesis of TA involves large vessel wall infiltration, predominantly by T and B lymphocytes and dendritic cells. Infectious diseases, particularly *Mycobacterium tuberculosis*, can be a trigger for the development of TA^{2 3} as a hypersensitivity reaction.⁴ In contrast, inflammation of the aorta may be secondary to the direct invasion of the artery by the *M. tuberculosis*.

We describe a case in which the distinction between TA and tuberculous arteritis was not clear, and discuss the relationship between these two conditions.

CASE PRESENTATION

A 23-year-old woman presented to our outpatient clinic with a 2-year history of nodular, painless, hyperchromic lesions on her lower legs associated with symmetric polyarthralgia (ankles, knees, wrists and elbows) and intermittent fever of non-specific pattern. She had previously had a skin biopsy showing leucocytoclastic vasculitis with granulomatosis without Acid-fast bacillus in the specimen. Her medical history was remarkable for the diagnosis of prolactinoma in 2013, secondary to hypophyseal microadenoma, for which she was taking cabergoline (1 mg/week orally). She reported an improvement in serum prolactin and decrease in the microadenoma after initiation of the therapy. She was also taking prednisone (20 mg/day) for her cutaneous and joint manifestations for the last 6 months, with partial response. On examination, she was in good condition with a blood pressure of 120/80 mm Hg at the right upper limb and 90/70 mm Hg at the left upper limb. A systolic murmur was audible at left sternal border. Cutaneous nodular hyperchromic lesions were noted on her

legs (figure 1). The rest of the physical examination was unremarkable.

INVESTIGATIONS

The initial investigational work up was within normal range for the patient's complete blood count, liver enzymes, electrolytes, renal function, blood cultures (pyogenic, mycobacteria and fungi), C3 and C4 fractions, rheumatoid factor, hepatitis serology (B and C) and urinalysis. Sputum bacilloscopy was negative. The erythrocyte sedimentation rate was 100 mm/h; C reactive protein was 7.6 mg/dL (normal limit 6 mg/dL); skin tuberculin test was strongly positive (23 mm); chest X-ray showed a dilation of the aortic arch. CT of the thorax revealed findings that could suggest the presence of pulmonary tuberculosis (TB) such as small airway disease, and mucoid bronchiolar impaction in the anterior segment of the upper left lobe and in the apicoposterior segment, extending to the aortic arch, indicating the probable dissemination route to the aortic arch (figure 2). A computed angiotomography of the aorta and its large branches showed the following findings: saccular aneurysms in aortic isthmus (measuring 6.2 cm in maximum length), origin of the brachiocephalic artery trunk (2.2×2.2×2.2 cm), origin of the left common carotid artery (1.6×1.2×1.2 cm), occlusion of the left subclavian artery due to a thrombus, luminal narrowing of the left pulmonary artery due to aortic aneurysm, compression and thrombosis of the left lower lobar artery with partial recanalisation by collaterals (figure 3). Thoracic MRI showed mild aortic wall oedema, indicating an inflammatory process. Transthoracic echocardiogram showed a saccular aneurysm in the aortic isthmus with thickening in the wall, suggesting aortitis.

DIFFERENTIAL DIAGNOSIS

Considering the patient's initial clinical manifestation (leucocytoclastic vasculitis), a range of

Figure 1 Cutaneous nodular hyperchromic lesions.

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Figure 2 (A) Signs of small airway disease and mucoid bronchiolar impaction in the anterior segment of the upper left lobe (arrow) suggesting tuberculosis. (B) The same finding in the apicoposterior segment, extending to the aortic arch, indicating the probable dissemination route to the aortic arch.

Figure 3 (A) Thickening of the aortic wall (straight arrow) and pseudoaneurysm in the aortic arch (curved arrow). (B) Occlusion of the lower lobe branch of the right pulmonary artery.

diagnostic possibilities exists including primary vasculitis (eg, antineutrophil cytoplasmic autoantibody-associated) and hidden infectious processes (eg, hepatitis C). However, the combination of these skin lesions with involvement of large vessels, strong purified protein derivative positivity and possible TB on imaging examination narrows the differential diagnosis to two possibilities: TB with vascular inflammatory activity and TA.

TREATMENT

The patient underwent empiric therapy for TB with rifampicin, isoniazid, pyrazinamide and ethambutol for 6 months, and began taking prednisone (1 mg/kg/day). Three months after starting drug therapy, she was submitted to a bypass surgery with placement of a Y Dacron tube from the ascendant aorta to the right brachiocephalic trunk and left common carotid, associated with an implantation of an endoprosthesis in the ascending aorta subsequent to the bypass towards the descending thoracic aorta with aneurysm exclusion.

OUTCOME AND FOLLOW-UP

The patient has been clinically well and asymptomatic, and the dose of prednisone has been tapered to 5 mg/day.

DISCUSSION

The relationship between TB and TA is well known. Some patients with TA have had previous contact with *M. tuberculosis*, shown either by the positivity of the skin tuberculin test^{5 6} or by the detection of specific gene sequences of the bacillus in aortic tissue.⁷ The present case is intriguing because her manifestations did not allow for the distinction of whether or not it was a true TA in a patient previously exposed to *M. tuberculosis* or a tuberculous arteritis itself. From the clinical point of view, joint manifestation can be a feature of TA but can also be a finding of a TB infection as a reactive process known as Poncet's arthritis in a patient with high levels of immunity to *M. tuberculosis*.⁸

Another confounding factor was the patient's cutaneous manifestation. The finding of granulomatous vasculitis in a skin biopsy can present in both TA and tuberculous arteritis. Though a large vessel vasculitis, TA may have its inflammatory process

generally begin in the *vasa vasorum*; this could also explain the presence of small vessel vasculitis in the dermal tissue in some cases of TA.⁹ Alternatively, the patient's cutaneous granulomatous vasculitis may be part of the spectrum of tuberculids associated with an *M. tuberculosis* infection.^{10 11}

Regarding the imaging findings, the presence of saccular aneurysms could suggest infectious disease (TB) as a cause of the vasculitic process,¹² however, saccular aneurysms have also been described in TA.¹³ Moreover, the presence of multiple lesions in different arterial areas would not be expected in classical aortic tuberculous aneurysms; however, the presence of multiple aorta tuberculous pseudoaneurysms have also been described previously.¹⁴ Unfortunately, during the surgical procedures, we could not retrieve any material for pathological analysis.

Of note, the pituitary lesion presented by our patient was considered as incidental. Despite the reported cases of pituitary tumours secondary to inflammatory disease and mycobacterial infections,¹⁵ the patient had no other evidence of hypophysitis, and she had a good therapeutic response to cabergoline. This medication has been associated with abdominal periaortitis and retroperitoneal fibrosis,¹⁶ but to the best of our knowledge, there is no report on its induction in aortic multiple aneurysms.

In conclusion, this case illustrates the difficulty in precisely defining the clinical entity as TA or tuberculous arteritis. Specifically, we cannot exclude the possibility that TB was indeed a trigger for that autoimmune event (TA) or even that it simply represents a coincident of contact with TB and TA because both conditions have a high prevalence in the Brazilian population.

Learning points

- ▶ *Mycobacterium tuberculosis* can be a trigger for the development of Takayasu's arteritis.
- ▶ Takayasu's arteritis and tuberculous arteritis share clinical and radiological features.
- ▶ Before starting treatment for Takayasu's arteritis, infection by *M. tuberculosis* should be excluded.

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Competing interests None declared.

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