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Takayasu's arteritis presenting in a pregnant woman with ankylosing spondylitis: Case report and review of the literature

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ABSTRACT

Introduction: Coincidence of ankylosing spondylitis (AS) and Takayasu arteritis (TA) in single person especially women is rare.

Case report: We report a 31 year old Iranian woman with AS from 5 years and giving history of inflammatory low back pain, morning stiffness, bilateral sacroiliac tenderness, positive Schöber test and enthesitis, negative human leucocytic antigen (HLA-B27) and sacroiliitis on plain X-ray. The patient was under treatment. After 2 years she returned with increased back, hip and entheseal pain as well as claudications in left hand during the eighth week of gestation in her first pregnancy. She discontinued the non-steroidal anti-inflammatory drugs three months before pregnancy and on examination was pulseless on the left side. Color Doppler studies showed segmental stenosis and increased intima thickness in subclavian, axillary and proximal brachial arteries of the left hand with reduced blood flow in those arteries. The right hand was normal. Laboratory result showed microcytic anemia (hemoglobin = 10.8 g/dl, mean corpuscular volume = 66 fL) and erythrocyte sedimentation rate = 104/1st hour. The findings were indicative of TA and she was treated with prednisolone and azathioprine and had successful delivery to full term normal boy at 39th week gestation by Caeserian section. She was followed for two more months after delivery with no complications.

Conclusion: Both TA and AS have no significant adverse effects on pregnancy if diagnosed and timely treated properly. When they occur concomitantly, more attention and care is needed to prevent complications.

Contents

1. Introduction

Ankylosing Spondylitis (AS) is a chronic inflammatory disease mostly affecting the sacroiliac joints, spine and entheses. It is usually accompanied with stiffness and progressive functional limitation of the axial skeleton [1,2] and could also be associated with sleep disturbances [3]. Moreover, subclinical peripheral joint arthritis has been reported in AS patients [4]. The disease commonly develops in men younger than 40 years [1,2]. AS is highly heritable and its risk has been associated strongly with Human leukocyte antigen (HLA) B27 [5]. Non-HLA gene polymorphisms have been reported among Iranian AS patients [6].

Takayasu arteritis (TA) is a rare chronic granulomatous vasculitis generally affecting large vessels including the aorta and its branches [7]. It also leads to narrowing, occlusion, and aneurysms

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of systemic and pulmonary arteries [8,9]. Nailfold changes of the small vessels and capillaries have been reported in Iranian TA patients [10]. Although rare, isolated peripheral pulmonary stenosis as the primary manifestation of TA has also been reported in an Iranian patient [7]. The disease usually affects Asian women in the childbearing age [8,9].

Previous studies indicate that TA disease activity does not increase during pregnancy, while pregnancy-related maternal and fetal complications are increased including uncontrolled hypertension, congestive heart failure, prematurity and fetal death. There is no appropriate treatment approach for TA during pregnancy and variable outcomes were reported [8,11]. Pregnant women with AS would also have different outcomes from normal pregnancy to complicated adverse birth outcomes [12]. Here we report a case of 31 year old women with AS presenting with TA symptoms during the first trimester of pregnancy.

2. Case report

A 31 year old woman was referred to us 5 years ago (by the age of 26) with the complaint of inflammatory low back pain and more than one hour morning stiffness that responded to routine non-steroidal anti-inflammatory drugs (NSAIDs). In the physical examination she had bilateral sacroiliac tenderness, positive Schöber test (2 cm) and enthesitis. Laboratory test showed positive CRP (2+) and negative human leukocytic antigen (HLA-B27), and there was sclerosis in both sacroiliacs in plain radiography. Altogether, she was diagnosed with AS according to the modified New York criteria [13] and treated with indomethacin since then. Her back pain was controlled with no further complaints during the following visits.

The patient stopped doctor's visits for two years and afterwards returned with the complaint of increased back, hip and entheseal pain as well as claudication pain in left hand during the eighth week of gestation in her first pregnancy. She had discontinued the NSAIDs three months before pregnancy. Physical examination showed that she had severe acute AS considering the limited Schöber and occiput to wall measures with no sensible pulse in the left arm and reduced pulse in the right hand. Color Doppler studies showed segmental stenosis and increased intima thickness in subclavian, axillary and proximal brachial arteries of the left hand with reduced blood flow in those arteries. The right hand was normal. Laboratory result showed microcytic anemia (hemoglobin = 10.8 g/dl, mean corpuscular volume = 66 fL) and erythrocyte sedimentation rate (ESR) = 104/1st hour. Other imaging modalities were not performed due to contraindications during the pregnancy.

These findings were indicative of TA and patient received prednisolone 30 mg, azathioprine 100 mg with folic acid and iron supplementation daily. Two months after treatment, the ESR was 33/1st hour, hemoglobin was 12.8 g/dl and her general symptoms were improved. The prednisolone was reduced to 5 mg during two months and azathioprine to 50 mg daily for the rest of the pregnancy. The patient did not develop any complications during pregnancy e.g. new onset of hypertension or cardiac abnormalities with improvement of her symptoms. No maternal or fetal complications occurred during the pregnancy and at the 39th week of gestation, she had Cesarean section and delivered a full term normal boy.

She was followed for two more months after delivery with no complications, but then she moved to other city before we could request further CT angiography or magnetic resonance angiography (MRA) of the left hand.

3. Discussion

Both AS and TA are rare inflammatory diseases and their occurrence at the same time in a single person is rarer. AS usually involves spine, hip and entheses causing restriction in range of motion; on the other hand, TA is inflammatory disease involving mainly aorta and its branches which presents with claudication pain in the limb [1,2,14]. There are case reports regarding coincidence of TA and AS in the same patient [1,2,15–28]. Table 1 shows a comparison of the characteristics of patients with concomitant AS and TA as well as the present case.

Studies have indicated that TA and AS occurrence could be incidental or concurrent due to some underlying factors [1,2]. In such cases, AS precedes TA. They are mostly women and are HLA-B27 negative [5–19], as was our case. While a study by Mielnik and colleagues [2] showed that most cases were males with positive HLA-B27 in 47% of them. It was reported that the age at onset was higher than only AS [14], while our patient was diagnosed at the age of 31. However, in the literature among the reported cases the age range was 14–63 years [15–28]. So both diseases could occur in any age. The important finding is that these patients usually have AS for years and later develop TA. It seems that when these patients are HLA-B27 positive, there is tendency to occur both AS and TA at younger ages. However, none of the recent studies found any relation between TA and AS or other seronegative spondyloarthritides (SpA) with HLA-B27 [28,29], indicative of other possible pathophysiologic mechanisms between these two diseases. On the other hand, a recent study by Kwon et al. [29] reported that extravascular manifestations of TA are rather com-

Table 1
Characteristics of cases reported with concomitant ankylosing spondylitis and Takayasu arteritis.

Case	Age (years)	Sex	HLA-B27	Clinical course	Year	Country
Cowley et al. [15]	Young	F	*−ve*	AS *before* TA	1987	UK
Magaro et al. [16]	32	F	*+ve*	AS *before* TA	1988	Italy
Aoyagi et al. [17]	26	M	*+ve*	UC/AS *before* TA	1998	Japan
Hilário et al. [18]	15	M	*+ve*	AS, CD, TA	1998	Brazil
Schuetz et al. [19]	45	F	*−ve*	TA *before* AS	2002	Swiss
Acar et al. [20]	14	F	*−ve*	*Concurrent* AS/TA	2005	Turkey
Taharboucht et al. [21]	26	F	na	AS *before* TA	2010	Algeria
Abdelghani et al. [22]	41	M	*−ve*	AS *before* TA	2013	Tunisia
Gan et al. [23]	18–35	4 M/2F	*+ve* 4/6	All AS *before* TA	2014	China
Souza et al. [24]	49	F	na	*Concurrent* AS/TA	2015	Brazil
Sattar et al. [25]	54	F	*+ve*	AS *before* TA	2015	USA
Mirfeizi et al. [26]	20	M	*+ve*	AS *before* TA	2015	Iran
Rivière et al. [14]	19–63	9F/2M	*−ve*	AS *before* TA	2017	France
Matsushida et al. [1]	56	M	*−ve*	*Concurrent* AS/TA	2018	Japan
Mielnik et al. [2]	*young*	2 M	na	TA *before* AS	2018	Norway
Montiel-Esparza et al. [27]	35	M	*+ve*	AS *before* TA	2019	Mexico
Esen et al. [28]	21–59	4F/1M	*−ve*	AS *before* TA	2019	Turkey
Present case	31	F	*−ve*	AS *before* TA	2019	Iran

F: female, M: male, HLA: Human leucocytic antigen, AS: ankylosing spondylitis, TA: Takayasu's arteritis, UC: ulcerative colitis, CD: Crohns disease, na: not available.

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mon with axial or peripheral arthritis as the most common findings. This indicated that these two maybe the same disease with different presentations, which needs further evaluations. Sacroiiilitis was present in 7.1% of patients with TA. Gan et al. [23] in a large retrospective study reported that 1.04% of AS patients had TA and 1.27% of TA patients had AS. In all cases, AS was initially diagnosed. AS would present with different cardiovascular complications and chronic inflammation was considered responsible for these findings. The imbalance between inflammatory markers is the cause for arterial inflammation in TA [28].

Adverse pregnancy outcomes have been broadly studied in other rheumatic diseases as rheumatoid arthritis [30], adult-onset Stills [31], systemic lupus erythematosus [32,33], systemic sclerosis [34], idiopathic inflammatory myositis [35], vasculitis [36] and Behcet's disease [37] and such patients should be considered as a high-risk group. The course of AS [12] and TA can affect pregnancy outcome [8,9,11]. As far as we know, our case is the only patient with concomitant TA and AS during pregnancy which had no maternal or fetal complications during the pregnancy. Zhang and colleagues [38] evaluated 13 TA cases occurring during pregnancy and observed successful delivery in 11 cases with normal neonates. They observed no asphyxia or fetal death. Maternal complications on the other hand occurred in 4 cases as preeclampsia or stroke. They believed that timely diagnosis and treatment of the disease would assure good pregnancy outcome for mother and the neonate. Gudbrandsson and colleagues [11] also observed that there was no difference in the rate of miscarriage, abortion or maternal complications in women before or after TA occurrence which indicated that even with TA presence with proper treatment, the outcome will be favorable.

Park et al. [12] evaluated 1293 deliveries in 996 Korean women with AS and observed that the rate of Cesarean section was higher than normal population, but the maternal and fetal complications were similar to the healthy control pregnant subjects. Although AS is associated with reduced quality of life during the pregnancy, but has no significant maternal or neonatal complications [39]. However, a higher rate of Cesarean section and preterm or small-for-gestational-age in neonates has been reported [40].

In conclusion, both TA and AS have no significant adverse effects on pregnancy if diagnosed and timely treated properly. When they occur concomitantly, more attention and care is needed to prevent complications.

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Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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