
INTERNATIONAL JOURNAL OF CURRENT RESEARCH IN BIOLOGY AND MEDICINE

ISSN: 2455-944X

www.darshanpublishers.com

DOI:10.22192/ijcrbm

Volume 2, Issue 5 - 2017

Case Report

DOI: <http://dx.doi.org/10.22192/ijcrbm.2017.02.05.001>

Diffuse goitre in Ankylosing Spondylitis

N.S Neki^{*}, Neeraj Joshi^{**}, Gagandeep Singh Shergill^{**}, Gagandeep Singh^{***},
Khushpreet Singh^{**}, Rubal Sharma^{****}, Gaurav Joshi^{*****}

^{*}Professor, ^{**} Junior Resident Department of Internal Medicine, Government Medical College & Guru Nanak Dev Hospital, Amritsar, Punjab. Pin 143001.

^{***} Research Associate, Department of Neurology, University Hospitals Cleveland Medical center, Cleveland, Ohio (USA)

^{****}Intern Department Of Internal Medicine, Government Medical College & Guru Nanak Dev Hospital

^{*****} Assistant professor Pharmacy Department, DDM Group Of Institutes Una, Himachal Pradesh

Corresponding author: **Dr. N.S. Neki**

88, Street No. 4, Gopal Nagar, Majitha Road, Amritsar, Punjab, India.

Email: drneki123@gmail.com Phone: 09501029128

Abstract

Autoimmune thyroid disease is well known to be associated with various autoimmune diseases, yet it is less clear whether a similar association also exists for ankylosing spondylitis (AS). We are presenting a case of ankylosing spondylitis, which presented with autoimmune hypothyroidism and diffuse neck swelling.

Keywords: Ankylosing spondylitis, Goitre, Autoimmune thyroid disease.

Introduction

Ankylosing spondylitis (AS) is the prototypic form of spondyloarthritis (SPA), typically develops in young adults, with a peak age of onset between 20 and 30 years. It is a group of disorders characterized by inflammation around the entheses (the sites of ligament insertion into bone) as well as an association with the human leukocyte antigen (HLA)-B27, and radiographic sacroiliitis. Extra articular manifestations are common with AS, but no case of Ankylosing spondylitis with diffuse thyroid swelling has been reported in English medical literature so far yet to the best of our knowledge.

Case report

A 20-year-old male patient admitted with H/O joints pain in gluteal region from last 8 month, swelling in neck and difficulty in eating food from last 6 months. Pain in gluteal region was insidious in onset, more at night time, increased during rest and improved as day progress along with stiffness of joints. There was no H/O sore throat and high grade fever (to rule out rheumatic heart disease). After 2 months of joint pain, he started complaining of swelling in neck which was insidious in onset, progressed in size within 2 months and caused marked dysphagia more to solids. He had H/O constipation and lethargy. There was no significant family history. No H/O diabetes mellitus,

hypertension and tuberculosis in the past. On examination neck swelling was diffuse, not associated with pain (Figure 2). For joints pain he took medicine from local practitioner and was in remission as long as he used to take analgesic. General physical examination was normal except for pain in gluteal and lower back with stiffness of joints and swelling in neck. Modified Schober test was done which was highly positive. Blood work-up and X-ray of sacro-iliac region was planned. Complete haemogram, renal and liver profile were normal but thyroid profile was deranged showing high TSH and low Free T3 and T4. Anti TPO antibodies were positive. ESR 115mm/1stHr, ANTI CCP was negative. FNAC was planned which

showed fibro collagenous tissue with entrapped mature adipocytes along with benign looking thyroid follicles lined by low cuboidal epithelium filled with colloid, no evidence of malignancy seen. X-Ray showed fusion in sacro-iliac region (Figure 2). HLA-B27 was also positive. From clinical, blood work up and X-Ray findings, a final diagnosis of ankylosing spondylitis with autoimmune goitre was made. Appropriate anti-inflammatory drugs and levothyroxine was started and patient planned for surgery due to progressive dysphagia in spite of medical management. He underwent B/L thyroidectomy successfully and is under follow-up.

Figure 1: Histopathological specimen of thyroid gland after thyroidectomy.



Figure 2: X-Ray right hip showing joint fusion

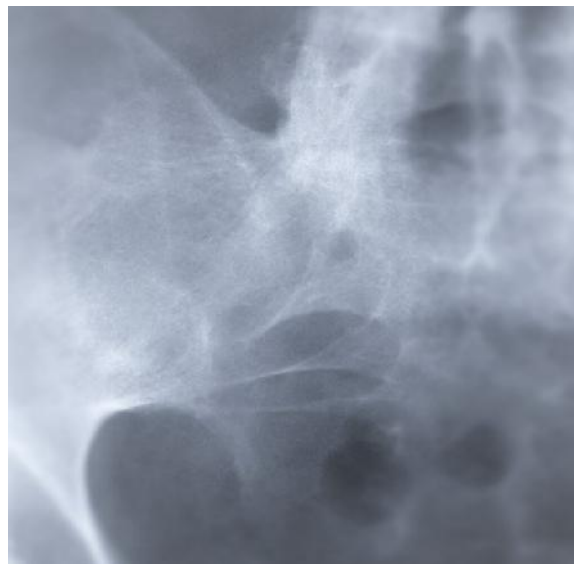
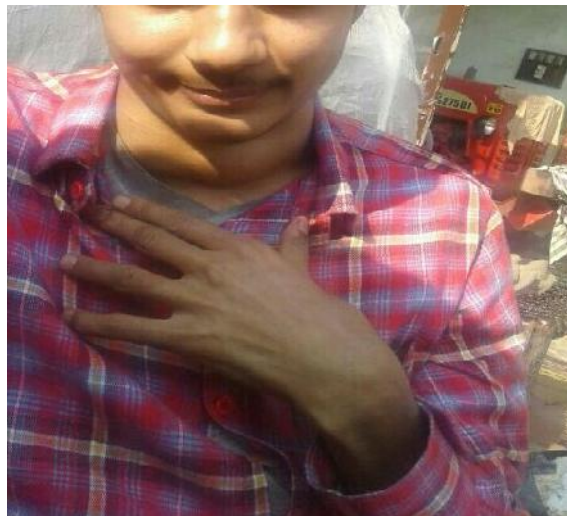


Figure :3 Diffuse goitre in patient with Ankylosing spondylitis.



Discussion

Ankylosing spondylitis(AS) is a member of a family of diseases referred to as spondyloarthritis (SPA). The condition was first fully described in the late 1600s by Bernard Connor (Ref.). It affects young adults with the peak age of onset between 20 and 30 years. Men are more often affected than women, with a ratio of approximately 3:1.¹ About 80% of patients with AS develop the first symptoms before their third decade of life and < 5% present in the fourth decade of life. However, patients with juvenile-onset AS become symptomatic at or before 16 years of age.

In population surveys, AS is present in 1-6% of adults inheriting HLA-B27, whereas the prevalence is 10-30% among B27 positive adult first degree relatives of AS probands².

The exact cause of AS and other spondyloarthropathies is unknown, but genetic and environmental factors play an important role in their pathogenesis. A strong link between AS and the *HLA-B27* gene exists and has been definitively confirmed³. Other MHC genes implicated in AS include HLA-DRB1/MHC Class II alleles, tumor necrosis factor- , IL6R,IL1R2³.

In patients of ankylosing spondylitis despite inflammatory low back pain with or without peripheral arthritis, extra-articular manifestations are also common which include anterior uveitis(40%), IBD (5-10%), psoriasis(10%), aortic valve insufficiency,

cardiac conduction disorders, prostatitis, cauda equina syndrome and apical lung fibrosis(rare late complications).Secondary amyloidosis, though rare, may also be seen.^{4,5}

Autoimmune hypothyroidism may be associated with signs or symptoms of other autoimmune diseases commonly vitiligo, pernicious anemia, Addison's disease, alopecia areata and type 1 diabetes mellitus. Less common associations include celiac disease, dermatitis herpetiformis, chronic active hepatitis, rheumatoid arthritis⁶,systemic lupus erythematosus (SLE),myasthenia gravis, and Sjogren syndrome.^{7,8}Possibly due to a common genetic background, it is less clear whether a similar association may also be present for AS. Autoimmune hypothyroidism, which is common in other autoimmune diseases, but its prevalence is approximately 12.8%⁹ as extra articular manifestation of ankylosing spondylitis .

During literature review, we found few studies demonstrating association of autoimmune hypothyroidism with ankylosing spondylitis, but very less research has been done showing this association with different results, which are mentioned below.

In 1999, Lange et al investigated the frequency of various thyroid disorders in 22 female patients with AS and 22 healthy age matched female controls, and found that the prevalence of anti-thyroid antibodies was significantly higher in the AS group¹⁰.

In 2012, Pérez-Fernández et al¹¹ investigated the frequency of autoimmune thyroid diseases in patients with spondyloarthritis (spa). They initially screened the patients with AS (n=80), PsA (n=31), and uSpA (n=24) by performing thyroid function tests. According to the study protocol, autoimmune thyroid disease was further investigated only in patients having abnormal thyroid function tests. Hypothyroidism of any cause was observed in 14 patients (9.5 %), eight with AS, four with psA, and two with uspa. Among those 14 patients, only five patients, three with AS, one with psA, and one with uspa, met the criteria for autoimmune thyroid diseases. They reported the frequency of autoimmune thyroid diseases in AS, psA, and uspa groups as 3/80 (3.75 %), 1/31 (3.20 %), and 1/24 (4.16 %), respectively.

Most recently, Tarhan et al¹² retrospectively investigated the frequency of thyroid involvement and thyroid dysfunction in 108 patients with AS (M/F=81/27), and detected subclinical hyperthyroidism in three patients and subclinical hypothyroidism in two patients. The frequencies of thyroid nodule, anti-TPO, and anti-TG positivity were reported as 27 (29/108), 29 (32/108), and 11 % (12/108), respectively. They concluded that frequency of autoimmune thyroid disease was higher in the whole group of AS patients than reported in the literature. However, the frequency of thyroid parenchyma hypoechogenicity with USG and the number of patients fulfilling the diagnosis of Hashimoto thyroiditis (HT) were not reported. This study had no healthy control group; however, the subgroup of AS patients with or without anti-TNF treatment was compared with each other. Interestingly, they observed that the frequency of thyroid disorders was significantly lower in the subgroup of patients receiving anti-TNF treatment.

If there is really an association between AS and autoimmune thyroid diseases, this might be due to common genetic pathogenic mechanisms predisposing to these two disorders. Since the frequency of HLA-B27, which has an important role in the pathogenesis of AS, was reported to be four times higher in autoimmune endocrinopathies than in healthy controls¹³, HLA-B27 positivity may also have an additional role in the pathogenesis of autoimmune thyroid disorders. More recently, another molecule known as cytotoxic T-lymphocyte antigen 4 (CTLA-4) was suggested to play a role in the pathogenesis of both AS and autoimmune thyroid disease^{14,15}. Therefore, it may be speculated that both HLA-B27 and CTLA-4 may contribute to the coexistence of

these two diseases, possibly playing a role in the pathogenesis of both AS and autoimmune thyroid disorders.

Some above mentioned recent studies and in our case which showed positive HLA-B 27 and Anti-TPO antibodies with swelling of thyroid, favours that association between two might be present. More prospective controlled studies with larger number of patients, and more better elucidation of the underlying pathogenic mechanisms for both diseases should be done.

Financial support and sponsorship: Nil

Conflicts of interest: there are no conflicts of interest

References

1. Gran JT, Husby G, Hordvik M. Prevalence of ankylosing spondylitis in males and females in a young middle-aged population of Tromsø, northern Norway. *Ann Rheum Dis* 44:359-367, 1985.
2. Joel D.Taurog, John D.Carter. "The spondyloarthritis." *Harrisons principles of internal medicine*. Vol. 2 (19th ed.). McGraw-Hill.p2169.
3. Joel D.Taurog, John D.Carter. "The spondyloarthritis." *Harrisons principles of internal medicine*.Vol.2 (19th ed.).McGraw-Hill. p2170.
4. Zochling J, Smith EU (2010) Seronegative spondyloarthritis. *BestPract Res Clin Rheumatol* 24:747-756.
5. Joel D.Taurog, John D.Carter. "The spondyloarthritis." *Harrisons principles of internal medicine*. Vol. 2 (19th ed.). McGraw-Hill.p2171.
6. Chan AT, Al-Saffar Z, Bucknall RC (2001) Thyroid disease in systemic lupus erythematosus and rheumatoid arthritis. *Rheumatology* 40:353-354.
7. Scofield RH (1996) Autoimmune thyroid disease in systemic lupus erythematosus and Sjogren's syndrome. *Clin Exp Rheumatol* 14:321-330.
8. "Harrisons principles of internal medicine.Vol.2 (19th ed.).McGraw-Hill. p2291.
9. Serda Em, Mehtap Bozkurt, Mehmet Ça layan, Pelin Oktayo lu, Mehmet Karakoç. The evaluation of comorbid diseases in patients with ankylosing spondylitis. doi: 10.5798/diclemedj. 2014; 41 (4): 662-666.

10. Lange U, Boss B, Teichmann J, Klett R, Stracke H, Bretzel RG et al (1999) Thyroid disorders in female patients with ankylosing spondylitis. *Eur J Med Res* 22:468–474.
11. Pérez-Fernández OM, Mantilla RD, Cruz-Tapias P, Rodríguez- Rodríguez A, . (2012) Spondyloarthropathies in autoimmune diseases and vice versa. *Autoimmune Dis.* doi: 10.1155/2012/736384.
12. Tarhan F, Oruk G, Niflioglu O, Ozer S. (Thyroid involvement in ankylosing spondylitis and relationship of thyroid dysfunction with anti-TNF treatment. *Rheumatol Int*(2013) 33:853–857.
13. Hrdá P, Sterzl I, Matucha P, Koriath F, Kromminga A (2004) HLA antigen expression in autoimmune endocrinopathies. *Physiol Res* 53(2):191–7.
14. Lee WY, Chang YH, Lo MK, Chang CP, Yang SC, Yang TP et al. (2010) Polymorphisms of cytotoxic T lymphocyte-associated antigen-4 and cytokine genes in Taiwanese patients with ankylosing spondylitis. *Tissue Antigens* 75:119–126.
15. Kavvoura FK, Akamizu T, Awata T, Ban Y, Chistiakov DA, Frydecka I et al. (2007) Cytotoxic T lymphocyte associated antigen 4 gene polymorphisms and autoimmune thyroid disease: a meta analysis. *J Clin Endocrinol Metab* 92:3162–3170.

Access this Article in Online	
	Website: www.darshanpublishers.com
Quick Response Code	Subject: Medicine

How to cite this article:

N.S Neki, Neeraj Joshi, Gagandeep Singh Shergill, Gagandeep Singh, Khushpreet singh, Rubal Sharma, Gaurav Joshi. (2017). Diffuse goitre in Ankylosing Spondylitis. *Int. J. Curr. Res. Biol. Med.* 2(5): 1-5.
 DOI: <http://dx.doi.org/10.22192/ijrbm.2017.02.05.001>