Clin Rheumatol (2003) 22: 94–98
DOI 10.1007/s10067-002-0681-2

ORIGINAL ARTICLE

A. El Maghraoui · R. Bensabbah · R. Bahiri
A. Bezza · N. Guedira · N. Hajjaj-Hassouni

Cervical spine involvement in ankylosing spondylitis

Received: 13 May 2002 / Accepted: 16 October 2002

Abstract Our objective was to study cervical spine involvement in a Moroccan population of ankylosing spondylitis (AS) patients and evaluate correlations with disease symptomatic and structural severity. Patients were prospectively enrolled for a 1-year period. Clinical, biological, and radiological data were collected. The risk of cervical spine involvement was estimated using the Kaplan–Maier method. Sixty-one patients were enrolled: 38 males (62.2%) and 23 females of mean (SD) age 35.1 years [11] (range 17–66). The mean disease duration was 10.6 years [7] (0.5–30). Forty-three patients (70.4%) had a history of neck pain. Radiological involvement was present in 33 cases (54%). The concordance between clinical and radiological involvement was statistically significant ($\kappa = 0.49$; $P < 10^{-6}$). The risk of cervical spine involvement with regard to disease duration showed that 19.6% of patients had radiological involvement after 5 years, 29.9% after 10 years, 45.1% after 15 years and 70.0% after 20 years. Comparison between patients with and without cervical spine radiological involvement showed no difference in age of onset or sex. There was statistical difference in symptomatic severity parameters (Schöber, chest expansion, BASMI, BASFI, BASDI, BASG) and structural severity parameters (lumbar syndesmophytes score, BASRI). Our study confirms the greater severity of AS in North African countries. Cervical spine involvement increases with age and disease duration in AS and is more frequent in symptomatic and structural severe forms of the disease.

Keywords Ankylosing spondylitis · Cervical spine

A. El Maghraoui · R. Bensabbah · R. Bahiri · A. Bezza
N. Guedira · N. Hajjaj-Hassouni
El Ayachi Hospital, Rabat-Salé,
Ibn Sina University Centre, Morocco

A. El Maghraoui (✉)
Internal Medicine Department,
Military Hospital Mohamed V, Rabat, Morocco
Tel.: 212 61 54 71 90
Fax: 212 37 71 70 32
E-mail: a_elmaghraoui@hotmail.com

Introduction

Ankylosing spondylitis (AS) is a chronic rheumatic disorder characterised by inflammation of the enthesis and sometimes the joints, which may lead to ankylosis. Neck involvement is classically thought to be common, especially in women and in patients with a long duration of disease [1,2]. Patients are at risk of atlantoaxial subluxation, cervical spine fractures and cervical spinal stenosis.

Previous studies conducted in North African countries showed that spondyloarthropathies in these countries are more severe than those observed in France and are characterised by more frequent hip involvement [3,4].

This study was conducted to determine the prevalence of clinical and radiological cervical spine involvement in Moroccan AS patients, their chronology in the history of the disease, and their correlations with indexes of disease symptomatic and structural severity.

Material and methods

Consecutive patients with a diagnosis of AS according to the modified New York criteria [5] who attended our department during 1 year were included in the study. Our department is one of the three rheumatological departments in Rabat where patients from all the north of Morocco are referred. Consent was obtained for all patients. All patients had a prospective rheumatologic assessment conducted by two rheumatologists (AEM and RB) using a structured questionnaire. This included demographic data (age, gender and duration of disease, defined as the time between the date of first symptoms and patient enrolment). Disease and neck pain history were registered from direct interview. Inflammatory neck pain was defined as night cervical pain associated with morning stiffness. Clinical examination included lumbar flexion measured by Schöber index, and chest expansion measured at the level of the xiphisternum. Cervical spine mobility was estimated using different distances measured with a tape: chin-to-chest distance, measuring the decrease of the distance between the chin and jugulum in maximal cervical flexion; chin–acromion distance, measuring the decrease of the distance between the chin and the acromial process in maximal cervical rotation on both sides (left

and right); and tragus–acromion distance, measuring the decrease of the distance between the tragus and the acromial process in maximal lateral bending of the head on both sides (left and right). Disease symptom severity was measured by the Bath Ankylosing Spondylitis Metrology Index (BASMI) [6], the Bath Ankylosing Spondylitis Functional Index (BASFI) [7], and the Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) [8]. Global evaluation of disease handicap was evaluated by Bath Ankylosing Spondylitis Global Score (BAS-G) [9].

Radiological assessment

Recent (within 6 months) X-rays of the pelvis, lumbar and cervical spine were obtained for all patients and read blind by the same observer (AEM). Intraobserver variability was good (correlation coefficient ranged from 0.92 to 0.98). Nobody had been excluded because of missing X-rays. Sacroiliitis and hip involvement were assessed on anteroposterior pelvic X-rays and graded respectively on New York [10] and BASRI scales [11–13]. Cervical spine involvement was assessed on anteroposterior and full flexion lateral views. Squaring was defined as present if the surface of the vertebra was convex, or if a line could fictively be drawn with a transparent ruler from the upper to the lower border of a vertebral body, and if this line overlapped 50% or more with the surface of the vertebra. Discitis was defined by the presence of disc space narrowing, with or without vertebral erosions or the absence of osteophytes. Radiological anterior atlantoaxial subluxation was diagnosed if the distance between the anterior aspect of the odontoid and the posterior aspect of the anterior arch of the atlas was 3 mm or greater in the lateral maximal flexion view. Finally, cervical spine involvement was defined by the presence of at least one of the following radiographic lesions: squaring, vertebral erosions, syndesmophytes, discitis, or facet joint involvement.

Lumbar spine syndesmophytes were assessed on posteroanterior and lateral lumbar spine standard radiography. Syndesmophyte score was obtained after grading on a scale of 0–3 (already used in many previous studies [14–19]), with 3 being equal to complete bony bridging (bamboo spine), 2 to more than three syndesmophytes bridges, and 1 to incipient syndesmophytes. Global radiological involvement was measured by the Bath Ankylosing Spondylitis Radiological Score (BASRI) [11–13].

Biological assessment

Erythrocyte sedimentation rate (ESR) was measured by the Westergren method.

Statistical analysis

The study was conducted in different steps:

1. The description of the study population.
2. Measurement of the prevalence of cervical spine involvement (clinically and radiologically), and the concordance between clinical and radiological cervical spine involvement using the κ test.
3. A description of the radiological features.
4. Estimation of the risk of cervical spine involvement with regard to disease duration, using the Kaplan–Meier method. For this analysis the beginning of cervical spine involvement was defined as the date of the first clinical symptoms of cervical spine disease, and the date of the onset of disease was defined as the date of the first occurrence of symptoms related to spondyloarthropathy.
5. Comparison of the patients with and without cervical spine involvement using the χ^2 test for qualitative variables and Student's *t*-test for quantitative variables.

Results

The main characteristics of the patients are summarised in Table 1. Sixty-one consecutive patients were seen in the 1-year recruitment period, 38 men (62.2%) and 23 women. Their mean age was 35.1 years $\pm$ 11 (17–66) and mean disease duration 10.6 years $\pm$ 7 (0.5–30).

Forty-three patients (70.4%) had a history of inflammatory neck pain with limitation of range of motion on at least one of the tests used in clinical examination. Radiological involvement was present in 33 patients (54%). The concordance between the clinical presentation (presence of history of neck pain) and the radiological lesions (presence of radiological lesions) showed $\kappa = 0.49$ ($P < 10^{-6}$). However, 12 patients with neck pain had no X-ray involvement and two patients with X-ray involvement did not complain of neck pain.

The different radiological features observed are summarised in Table 2. The most commonly observed lesion was the vertebral squaring (49.1%), followed by facet joint involvement (47.5%), syndesmophytes (26.2%), vertebral body erosions (21.3%) and discitis (16.3%). None of the patients presented with anterior C1–C2 subluxation or vertebral fracture.

Calculating the risk of cervical spine involvement with regard to disease duration using the Kaplan–Meier method showed that 19.6% of patients had radiological involvement after 5 years, 29.9% after 10 years, 45.1% after 15 years and 70.0% after 20 years (Fig. 1).

Comparison between males and females showed no significant statistical difference in any of the tested variables (data not shown).

Table 1 Demographic, clinical, biological and radiological characteristics of the study population. Results are expressed as mean (SD) for quantitative variables and *n* (%) for qualitative variables

		Range
Age: yrs, mean (SD)	35.3 (11.5)	17–66
Sex M/F	38/23	
Disease duration: yrs, mean (SD)	10.5 (7.3)	0.5–30
Clinical presentation		
Inflammatory back pain, *n* (%)	54 (88.5)	
Buttock pain, *n* (%)	57 (93.4)	
Neck pain, *n* (%)	45 (73.7)	
Peripheral arthritis, *n* (%)	33 (45.9)	
Enthesiopathy, *n* (%)	28 (45.9)	
Uveitis, *n* (%)	6 (9.8)	
Psoriasis, *n* (%)	3 (4.9)	
IBD, *n* (%)	6 (9.8)	
Biology		
ESR (mm), mean (SD)	41.5 (31.1)	2–105
Radiology		
Sacroiliitis, mean (SD)	3.0 (0.8)	2–4
BASRI, mean (SD)	5.4 (3.5)	2–12
Cervical spine radiological lesions, *n* (%)	33 (54)	
Cervical spine facet joint score, mean (SD)	4.7 (6.7)	0–18
Hip involvement, *n* (%)	38 (62.3)	

Table 2 Radiological features of cervical spine involvement in 61 patients with ankylosing spondylitis

	n	%
Normal X-rays	28	46
Squaring	30	49.1
Vertebral body erosions	13	21.3
Syndesmophytes	16	26.2
Discitis	10	16.3
C1–C2	3	4.9
C2–C3	2	3.2
C3–C4	1	1.6
C4–C5	6	9.8
C5–C6	3	4.9
C6–C7	2	3.2
Facet joint involvement	29	47.5
C2–C3	21	34.4
C3–C4	21	34.4
C4–C5	20	32.7
C5–C6	21	34.4
C6–C7	21	34.4
C7–D1	19	31.1

A comparison between patients with and without cervical spine radiological involvement is shown in Table 3. The patients with cervical spine involvement were older and had a longer duration of disease, but the difference is not statistically significant (37.3 years vs 32.8, $P = 0.12$, and 12.2 years vs 8.5, $P = 0.051$, respectively). Analysis showed no difference in age of onset or sex. There was a statistical difference in symptomatic severity parameters (Schöber, chest expansion, BASMI, BASFI, BASDAI, BASG) and structural severity parameters (lumbar syndesmophyte score, BASRI). Patients with cervical spine involvement had a more severe disease (lower Schöber index and chest expansion, and higher BASMI, BASFI, BASDAI and BASG). Patients with cervical spine involvement had a tendency towards more hip involvement, but the difference was not statistically significant (72.7% vs 50%; $P = 0.069$).

Four male patients had cervical spine syndesmophytes without lumbar spine syndesmophytes. Their mean age was 33.5 years (3.4) (29–37) and their mean disease duration was 11.0 years (8.2) (2–19). All of them had severe disease, with mean BASMI 7.0 (2.0) and BASFI 87.7 (12.3).

Discussion

This study confirmed the high prevalence of cervical spine involvement in AS in Morocco, as was also the case for hip involvement. However, the female predominance indicated in the literature was not observed. It confirmed the greater frequency of cervical spine involvement with increasing disease duration. Cervical spine involvement reduces neck motion and produces progressive neck kyphosis, which may progress until the patient is unable to look forward and has a chin-on-chest deformity. It also showed that the risk of cervical spine involvement is more common in symptomatic and structurally severe forms of AS.

Hip involvement is also associated with a more severe disease [20]. However, in contrast to cervical spine involvement, hip arthritis occurs at a young age and previous studies have shown that patients who did not develop hip disease after 10 years of AS have a great chance of never developing it at all [21–23].

As cervical spine involvement increased with disease duration, we chose a life-table analysis to estimate the risk of cervical spine involvement. The main methodological point that can be debated is the choice of accepting the patient's memory as sufficient to determine the disease onset and the onset of cervical spine involvement. However, the magnitude of this potential error in date is slight, and we think that this choice is more valid than using the date of first consultation as

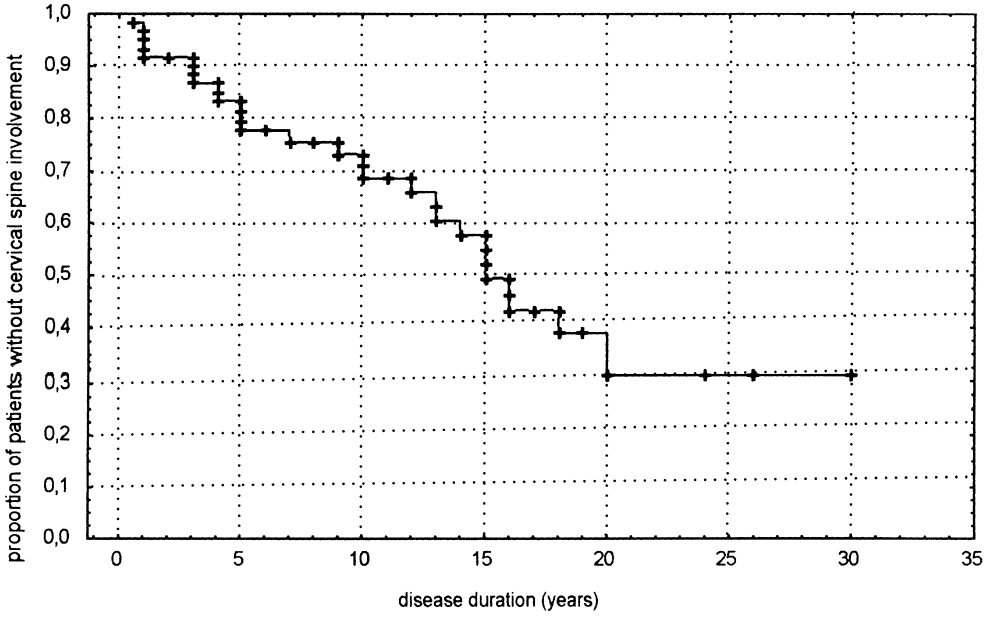

Fig. 1 Risk of cervical spine involvement during the course of 61 patients with ankylosing spondylitis

Table 3 Comparison between patients with and without cervical spine radiological involvement

	Presence of cervical spine radiological lesions (n = 33)	Absence of cervical spine radiological lesions (n = 28)	P
Age: yrs, mean (SD)	37.3 (11.9)	32.8 (10.6)	NS
Age of onset: yrs, m (SD)	24.5 (12.0)	25.1 (11.3)	NS
Sex: males, n (%)	20 (60.6)	18 (64.2)	NS
Disease duration: yrs, m (SD)	12.2 (7.8)	8.5 (6.1)	NS
Schöber: cm, m (SD)	1.7 (1.7)	3.1 (1.8)	0.03
Chest expansion: cm, m (SD)	3.7 (2.0)	5.1 (1.8)	0.008
BASMI: 0–10, m (SD)	5.1 (3.3)	2.2 (1.8)	0.0001
BASFI: 0–100, m (SD)	57.8 (31.5)	29.7 (32.1)	0.001
BASDAI: 0–100, m (SD)	45.9 (26.3)	25.6 (18.3)	0.001
BASG: 0–100, m (SD)	73.5 (29.3)	44.7 (30.8)	0.0004
BASRI: 2–12, m (SD)	7.4 (3.1)	3.0 (2.1)	<0.00001
Lumbar spine syndesmophyte score: 0–3, m (SD)	1.2 (1.5)	0.3 (1.9)	0.001
Hip involvement: n (%)	24 (72.7)	14 (50)	NS

disease onset, or the date of first abnormal X-ray as the onset of cervical spine involvement.

Different cervical spine radiological lesions are observed. In our study, the commonest feature was the vertebral squaring (which can be considered as the first stage of anterior vertebral body enthesopathy), followed by facet joint involvement. Syndesmophytes and disc involvement were observed in 16 (26.2%) and 10 (16.3%) cases, respectively. Recently, de Vlam et al. [24] showed that the facet joint involvement and the presence of bridging syndesmophytes were related, and suggested that the facet joint was primarily involved in AS. Our study confirms the high prevalence of facet joint involvement but did not find any interrelation with syndesmophytes.

None of our patients had anterior atlantoaxial subluxation or cervical spine fracture. In the literature [25,26], the prevalence of atlantoaxial subluxation varies from 6.8% to 21%, and seems to be more frequent in peripheral forms of spondyloarthropathy. This could reflect differences in patient selection or study design, or perhaps differences in clinical expression due to ethnicity. Moreover, atlantoaxial subluxation is not associated with disease duration and in many cases has been reported as a presenting feature of the disease [27, 28].

Finally, none of our patients underwent surgical treatment of the cervical spine. Unlike total hip arthroplasty, spinal surgery has the additional risk of important neurological and vascular complications owing to the close proximity of vital neurovascular structures [29]. About 0.5% of patients with AS will require cervical spine surgery at some stage. The results seems to be good, especially as regards neck pain, but the majority of patients in Koh's series [29] reported that neck stiffness was the same or worse after surgery.

In summary, our study confirms the greater severity of AS in North African countries. In Morocco AS is characterised by frequent hip and cervical spine involvement. The latter increases with disease duration and is more frequent in symptomatic and structurally severe forms of the disease, whereas hip involvement seems to be an independent feature, starting usually at a younger age. However, longitudinal studies including a higher number of patients are required to further evaluate the clinical significance and natural history of cervical spine involvement in AS.

References

1. Meijers KAE, Heerma van Voss SFC, Francois RJ (1968) Radiological changes in the cervical spine in ankylosing spondylitis. Ann Rheum Dis 27: 333–338
2. Resnick D, Dwosh IL, Goergen TG et al. (1976) Clinical and radiographic abnormalities in ankylosing spondylitis: a comparison of men and women. Radiology 119: 293–297
3. Hajjaj-Hassouni N, Maetzel A, Dougados M, Amor B (1993) Comparative study of patients evaluated for spondyloarthropathy in France and Morocco. Rev Rhum (Engl Ed) 60: 341–346
4. Claudepierre P, Gueguen A, Ladjouze A et al. (1995) Predictive factors of severity of spondyloarthropathy in North Africa. Br J Rheumatol 34: 1139–1145
5. Van der Linden S, Valkenburg HA, Cats A (1984) Evaluation of diagnostic criteria for ankylosing spondylitis. A proposal for modification of the New York Criteria. Arthritis Rheum 4: 361–368
6. Jenkinson TR, Mallorie PA, Whitelock HC, Kennedy LG, Garrett SL, Calin A (1994) Defining spinal mobility in ankylosing spondylitis. The Bath Ankylosing Spondylitis Metrology Index (BASMI). J Rheumatol 21: 1694–1698
7. Calin A, Garrett S, Whitelock HC et al. (1994) A new approach to defining functional ability in ankylosing spondylitis: the development of the Bath Ankylosing Spondylitis functional index. J Rheumatol 21: 2281–2285
8. Garrett S, Jenkinson T, Kennedy LG, Whitelock HC, Gaisford P, Calin A (1994) A new approach to defining disease status in ankylosing spondylitis: the Bath Ankylosing Spondylitis Disease Activity Index. J Rheumatol 21: 2286–2291
9. Jones SD, Steiner A, Garrett S, Calin A (1995) The Bath Ankylosing Spondylitis Patient Global Score [abstract]. Br J Rheumatol 34 (Suppl 1): 114
10. Bennet PH, Burch TA (1967) New York symposium on population studies in the rheumatic diseases: new diagnostic criteria. Bull Rheum Dis 28: 453–458
11. MacKay K, Mack C, Brophy S, Calin A (1998) The Bath Ankylosing Spondylitis Radiology Index (BASRI): a new, validated approach to disease assessment. Arthritis Rheum 41: 2263–2270
12. Kennedy LG, Jenkinson TR, Mallorie PA, Whitelock HC, Garrett S, Calin A (1995) Ankylosing spondylitis: the correla-

tion between a new metrology score and radiology. Br J Rheumatol 34: 767–770
13. Calin A, Mackay K, Santos H, Brophy S (1999) A new dimension to outcome: application of the Bath Ankylosing Spondylitis Radiology Index. J Rheumatol 26: 988–992
14. El Maghraoui A, Borderie D, Edouard R, Roux C, Dougados M (1999) Osteoporosis, body composition and bone turnover in ankylosing spondylitis. J Rheumatol 26: 2205–2209
15. Maillefert JF, Aho S, El Maghraoui A, Dougados M, Roux C (2001) Changes in bone density in patients with Ankylosing spondylitis: a two year follow-up study. Osteoporosis Int 12: 605–609
16. El Maghraoui A, Bezza A, Tabache F et al. (2000) A controlled study of sacroiliitis in Behçet's disease. Clin Rheumatol 20: 189–191
17. El Maghraoui A, Dougados M, Freneaux E, Chaussade S, Amor B, Breban M (1999) Concordance between abdominal scintigraphy using technetium-99m hexamethylpropylene amine oxime-labelled leucocytes and ileocolonoscopy in patients with spondyloarthropathies and without clinical evidence of inflammatory bowel disease. Rheumatology 38: 543–546
18. El Maghraoui A, Chaouir S, Bezza A et al. (2003) Thoracic high-resolution CT in patients with ankylosing spondylitis and without respiratory symptoms. Ann Rheum Dis 62: 185–186
19. El Maghraoui A, Chaouir S, Bezza A et al. (2002) Bone mass evaluation using computerized tomography in ankylosing spondylitis. Ann Rheum Dis 6: SAT173
20. Amor B, Silva Santos R, Nahal R, Listrat V, Dougados M (1994) Predictive factors of the long term outcome of spondyloarthropathies. J Rheumatol 10: 1883–1887
21. Khan MA (1994) Ankylosing Spondylitis: clinical features. In: Klippel J, Dieppe P, eds. Rheumatology. Mosby-Year Book Europe, 3: 25.1–25.10
22. Calin A, Elswood J (1998) The relationship between pelvic, spinal and hip involvement in ankylosing spondylitis. One disease process or several? Br J Rheumatol 27: 393–395
23. Carette S, Graham D, Little H, Rubenstein J, Rosen P (1983) The natural disease course of ankylosing spondylitis. Arthritis Rheum 26: 186–190
24. de Vlam K, Mielants H, Veys EM (1999) Involvement of the zygapophyseal joint in ankylosing spondylitis: relation to the bridging syndesmophyte. J Rheumatol 26: 1738–1745
25. Ramos-Remus C, Gomez-Vargas A, Guzman-Guzman JL et al. (1995) Frequency of atlantoaxial subluxation and neurologic involvement in patients with ankylosing spondylitis. J Rheumatol 22: 2120–2125
26. Suarez-Almazor ME, Russel AS (1988) Anterior atlantoaxial subluxation as a complication of early ankylosing spondylitis. J Rheumatol 15: 973–975
27. Sorin S, Askari A, Moskowitz RW (1979) Atlantoaxial subluxation as a complication of ankylosing spondylitis. Two case reports and review of the literature. Arthritis Rheum 22: 273–276
28. Hamilton MG, MacRae ME (1993) Atlantoaxial dislocation as the presenting symptom of early ankylosing spondylitis. Spine 18: 2344–2346
29. Howe Koh W, Garrett S, Calin A (1997) Cervical spine surgery in ankylosing spondylitis: is the outcome good? Clin Rheumatol 16: 466–470