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Original Research Article

The cervical spine in juvenile idiopathic arthritis

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Abstract

The objective of this research is to describe the clinical and radiological aspects of cervical spine involvement in Juvenile Idiopathic Arthritis (JIA). Patients fulfilled the ILAR (International League Against Rheumatism) criteria for the diagnosis of the JIA. The diagnosis of JIA course types were used in this study. In all patients conventional X-rays of the cervical spine were obtained and the relationship between clinical status (neck pain, limitation of neck motion and neurological complications) and radiographs findings were studied. Cervical spine disease was assessed according clinical parameters related to JIA. 120 patients were included, mean age 22 years ± 10 [5-49], disease duration of JIA was 12 years ± 9.6 [2-35]. Thirty cases of neck pain were found, 18 patients had an inflammatory cervical pain. Neurological complications (tetraparesia in one case and one case of pyramidal irritation) were noted. In our series no cervical spine surgery was performed. In 27 cases (22.5 %), radiological inflammatory abnormalities were detected in the cervical spine: Apophyseal joint ankylosis was noted in 14 patients, anterior atlantoaxial subluxation in 6 and cervical syndesmophytosis in 5 cases. Cervical spine involvement was more frequent in polyarticular and spondylarthropathy subtypes (p=0.01). Cervical spine changes were common in patients with long lasting (p < 0.001) and severe JIA (p= 0.04). In our series, the most frequent inflammatory change was apophyseal joint ankylosis and anterior atlantoaxial subluxation. Cervical spine changes are common in patients with long lasting JIA.

Keywords: Cervical spine, Juvenile Idiopathic Arthritis

INTRODUCTION

Juvenile idiopathic arthritis is a systemic disease of childhood affecting connective tissue, particularly joints. Cervical spine involvement in JIA (juvenile idiopathic arthritis) was recorded in early description of the disease (Still, 1987). Since then, clinical involvement of the cervical spine has been considered a common and characteristic manifestation of JIA, its frequency ranging from 60 to 70 % (Cassidy, 1982, Ansell and Bywaters, 1963). Radiological evidence of cervical spine abnormalities was found between 27 % and 80 % of different series (Espada et al., 1988). In general, apophyseal disease is the most frequent radiologic change and ankylosis of the zygapophyseal joints between the second and third cervical segments is considered particularly characteristic of JIA. Growth disturbances in the cervical spine are frequently seen in patients with apophyseal joint ankylosis and early disease onset (Espada et al., 1988). Detailed studies of cervical spine abnormalities in a substantial number of patients with JIA are limited in number; furthermore, few reports have correlated clinical features of disease with radiologic changes. The aim of this study was to describe the radiographic features in patients with JIA, and correlate them with clinical finding of the disease.

Table 1. Clinical and radiographic characteristics of population sample

Characteristics	n = 120
Age (years)[1]	22±10.5 (5-49)
Disease duration (years)[1]	10.4±4 (1-16)
Age at onset of JIA (years)[1]	12±9.6 (2-16)
HAQ[1]	1.6±0.80 (0-2.75)
Female[2]	68(57)
Neck limitation[2]	18(15)
Neurological complication[2]	2 (1.6)
One or more radiographic inflammatory abnormality[2]	27(22,5)
vertebral apophyseal joint ankylosis[2]	14(12)
Anterior AAS[2]	6(5)
Growth disturbance[2]	3(2,5)

[1]Median, standard deviation and extremes, [2] Number and percentage.
AAS: atlantoaxial subluxation

MATERIAL AND METHODS

Patients

Patient collection was retrospectively carried out in the out patient hospital and in rheumatology hospital, during the period from 2003-2007 using patient records and radiographs of patients who fulfilled the international league against rheumatism consecutive criteria for JIA and who had cervical spine radiographs available, taken because of neck symptoms or as regular follow up. The patients included in this study were constituted from children and adults. All subtypes of JIA were included. The clinical data relating to JIA were collected (age, sex, age at onset of arthritis, duration of evolution, subtypes. Cervical involvement was clinically assessed by noting the presence of the neck pain, limitation of neck motion and neurological complication.

Roentgengraphic survey

Anteroposterior and or lateral x rays views of the neck were obtained in all cases, the most recent cervical spine radiographs were used in the study. Radiological abnormalities of the cervical spine were tabulated following the classification of Komusi et al (1985) modified to include x-ray finding of childhood disease onset. A diagnosis of anterior atlantoaxial subluxation (AAS) was made if the distance between the anterior aspect of the dens and the posterior aspect of the anterior arch of the atlas was greater than 4 mm in adults and 4.5 mm in children. Posterior AAS was diagnosed if the posterior aspect of the anterior atlas arch was situated posteriorly in relation to the anterior aspect of the dens in lateral view radiographs during extension (Amett et al., 1980). Subaxial subluxation was diagnosed if a vertebra was displaced more than 3 mm in relation to the next lower vertebra when measured from the posterior line of the vertebral bodies. Apophyseal joint ankylosis was recorded when fusion was seen in the joints and the position of the vertebra was unchanged during both flexion and extension. The size of the fourth vertebral body of the cervical spine and the diameter of the sagittal spinal canal of the same vertebra were measured. The fourth vertebral body was considered small or narrow, the so-called juvenile cervical vertebra, when the height of the vertebral body was less than 11 mm or alternatively the sagittal width of the vertebral body was less than 15 mm (Laiho et al., 2002).

Radiographically changes in the hand were assessed in according to the steinbroker grades.

Statistical Methods

Statistical analysis of the difference between patients with cervical involvement and the remainder was calculated by Student's test or Mann-Whitney U test.

RESULTS

A total of 120 patients with JIA (68 female, 52 male) were found for whom a cervical spine radiographs was available; these patients comprised the study group. Mean age at onset of JIA was 12 years ± 9 (range 2 to 16 years), the mean age of the patients at the latest cervical spine radiographs was 22 years ± 10 (range 5 to 49 years), the mean duration at that time was 10 years ± 4 (range 1 to16 years). The median HAQ value was 1.6±0.80 (0-2.75). Clinical characteristics regarding the study group is presented in table 1. For patient subtypes, 19(16 %) systemic subtype, 34(28%) spondyarthropathy subtype, 22(18%) seropositive polyarticular disease, 29(24%) seronegative polyarticular disease, 10(8%) persistent oligoarticular, 6(6%) psoriatic disease (table 2). Twenty seven of our patients (22.5%) manifest at least

Table 2. Distribution of population sample according to JIA subtype

JIA subtype	n = 120
Systemic subtype[1]	19(16)
Spondyarthropathy subtype[1]	34(28)
Seropositive polyarticular disease[1]	22(18)
Seronegative polyarticular subtype[1]	29(24)
Persisitent oligoarticular[1]	10(8)
Psoriatic disease[1]	6(6)

[1]Number and percentage.

Table 3. Characteristics of JIA according to the presence of cervical spine inflammatory changes

Parameters	No cervical spine changes	Cervical spine changes	p
Age (years) [1]	20.5 [5-43]	31 [19-43]	< 0. 001
Age at onset (years) [1]	12 [2-16]	9 [2-16]	0.01
Duration of evolution (years) [1]	10 [3-32]	17.5 [4-35]	< 0. 001
HAQ [1*]	0.87 [0.-2.87]	1.87 [0-2.75]	0. 003
Sex [2]	15(52)	14(48)	0.34
Rhumatoïd Factor [2*]	1 (11.1)	8(88.9)	0.02

[1]Median and extremes, [2]Number and percentage. [*] If data is available.

one radiographic inflammatory abnormality. Fourteen (12%) patients had vertebral apophyseal joint ankylosis. The condition prevailed between C2 and C3 alone in 6 patients and fusion extended caudally to several levels in 8 patients. In 5 patients, bony ankylosis involved the entire cervical spine. Anterior AAS was detected in 6 patients, anterior AAS was in most cases small 4 to 5 mm (4 patients). The maximal atlantodental interval was 9mm. none of patients had posterior AAS. Growth disturbances, with decreased vertical and anteroposterior diameters of the vertebral bodies and decreased height of the adjacent intervertebral discs, were observed in 3 patients. Symptoms and signs of nerve root impingement or myelopathy were seen in 2 cases (one case with tetraperesia, one case with Babinski sign). No patient had undergone surgery on the cervical spine (Table 1).

Polyarticular and spondyarthropathy onset disease was more frequently associated with a higher frequency of cervical involvement (p=0.01). Cervical spine involvement was significantly associated with more advanced structural lesion according to Steinbroker grade (p=0.04). Patients with cervical spine involvement seemed to have an earlier age of disease onset, longer average disease duration.

DISCUSSION

Several clinical features differentiate the juvenile onset from the adult onset form of JIA, among the cervical features; cervical spine involvement has been a characteristically frequent feature of juvenile disease onset (Ansell and Bywaters, 1963; Espada et al., 1988, Ansell, 1977). Radiographic studies of the cervical spine have detected inflammatory abnormalities in 21% to 80 %, (Kjellberg and pavloub, 2011; Martel et al., 1962; Jeremy et al., 1960) of different series depending on patient selection. In this study, the cervical spine was seen in 22.5 % and was significantly related to longer disease duration and earlier onset disease in agreement with a previous report. However its relation to rheumatoid factor (RF) is arguable in literature, in fact, in our study RF was not statistically related to cervical involvement (p= 0.02), these results were not similar to those found by Espada (1988) who noticed a more frequent cervical involvement among the positive RF group, on the other hand Elhai (2013) didn't not find any difference of the cervical spine involvement among the positive RF groups. Radiological findings were evident in patients with polyarticular and spondyarthropathy subtypes, this could be anticipated because remission is most likely in the persistently oligoarticular disease (Elata et al., 1988), whereas the polyarticular subtype shows little tendency to remission. Juvenile ankylosing spondylitis had started at later age than other JIA. The facts that patients with oligoarthritis rarely have cervical spine manifestations have previously been reported by Schaller and Wedgwood (1972). In this study, apophyseal joint ankylosis was detected in 12% of the patients with JIA

and was the most frequent abnormality but less prevalent as has been the case in other studies (39%, 41%, 52% and 63 % (Martel et al.,1962; Laiho et al., 2002; Espada et al., 1988; Laiho et al., 2001). Characteristic of these patients with JIA was that the ankylosis was more frequently present at multiple levels of the cervical spine than at a single apophyseal level. Most commonly, single apophyseal joint involvement was seen at C2 –C3, a fact that confirms the findings of Ansell and kent (1977), and Laiho et al (2002). The upper cervical spine was the second most common site of inflammatory changes. Anterior AAS was mostly mild and detected almost as frequently as in study by Espada et al (1988) (22%) but less often than in the study on refractory JIA (35%) (Laiho et al., 2001). Ansell and Kent (1977) reported an increased frequency of atlanto-axial subluxation in seropositive JIA, this was not confirmed by others investigators (Hensiger et al., 1986). In our study AAS was more frequent in polyarticular subtype. AAI was more frequent in polyarticular disease this find was similar to reported data. Juvenile cervical vertebra was detected in 2,5% of patients with JIA, which is less frequently than those reported by the literature (22%) (Espada et al., 1988) and (26%) (Laiho et al., 1986). In patients with early onset disease, JIA seemed to be more likely associated with growth disturbance of the cervical vertebra (espada et al., 1988; Laiho et al., 2002). Limitation of neck motion was a common clinical finding; this was observed more frequently in patients with radiological evidence of joint fusion. Two patients had symptoms of myelopathy which was be treated by conservative means. In study of Espada et al (1988) no symptoms or signs of nerve root impingement or myelopathy were found, this finding suggest that neurological complications are less likely to develop in juvenile rather than in adult onset RA (salliere et al., 1983).

CONCLUSION

In conclusion, cervical interapophyseal joint fusion was the most frequent and characteristic change of the cervical spine and was usually seen at multiple levels of the cervical spine. The high frequency of AAI indicates the erosive nature of the inflammatory process especially in polyarticular cases.

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