Relationship between ossification of the stylohyoid ligament and enthesopathy: A comparative study

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Abstract

Objective

Stylohyoid apparatus might be an anatomic region in the cervical spine involved by enthesopathy. The aim of this study was to assess the elongation and/or ossification at the stylohyoid apparatus in the degenerative or inflammatory diseases such as ankylosing spondylitis (AS), psoriatic arthropathy (PsA) and cervical spondylo-arthrosis (CS) in which cervical spine involvement can be seen.

Methods

Twenty-eight patients with AS, 25 patients with PsA, 31 patients with CS and 50 controls who did not have any complaints or symptoms related with elongated styloid process (SP) were included in the study. On the lateral cervical radiographs, the anterior and posterior aspects of each vertebral body and intervertebral disk were carefully evaluated for the presence and severity of syndesmophytes, osteophytes or ossification of the adjacent longitudinal ligaments in the patient group. The entire osseous length of the SP was measured on the lateral and lateral oblique mandibular or cervical views or the anteroposterior views radiographs in the patients and controls.

Results

There were statistically significant differences between the patients with AS and control group and between the patients with PsA and control group with respect to the length of SP. The dimension of syndesmophytes or ligamentous ossification of the cervical spine involving C5-6 intervertebral disk level were significantly correlated with length of SP in the patients with AS.

Conclusion

Elongated SP might be another manifestation of enthesopaty in cervical spine of the spondyloarthropathies.

Key words

Enthesopathy, spondyloarthropathy, stylohyoid ligament ossification.

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Introduction

Styloid process (SP) and stylohyoid ligament (SHL) are originated from the second branchial arch cartilage. In man the ceratohyal element degenerates in time. However, its fibrous sheath persists as the SHL, which contains cartilaginous and osseous potential (1-7). Some authors state that the function of SHL is important to establish the centric relation of the mandible to the maxilla, since this is a ligamentous position. Others believe that SHL seems to have no function in determining centric relation or mandibular movement (4).

Elongation or mineralization of the stylohyoid apparatus is a relatively common occurrence that may produce a variety of clinical symptoms and radiographic appearances (1-3, 5, 7-12). Elongation of the stylohyoid apparatus causes irritation of the trigeminal, facial, glossopharyngeal or vagus nerves. This leads to symptoms of recurrent nonspecific throat discomfort, foreign body sensation, dysphagia, facial pain, increased salivation. Also, carotidynia may result from impingement of the styloid process on the carotid artery producing regional tenderness or headaches. These symptoms complex is known Eagle's Syndrome (7-12). The three theories of developmental ossification were proposed that could explain ossification or calcification of the stylohyoid apparatus. The "Theory of reactive hyperplasia" proposes that ossification would occur at the terminal end of the process. The "theory of reactive metaplasia" involves ligamentous sections of the stylohyoid apparatus to undergo metaplastic changes. These two theories could explain the marked ossification of the stylohyoid apparatus secondary to traumatic events. The third theory called the "theory of anatomic variance" involves the stylohyoid process and/or the SHL as ossified structures that develop in the early, formative years after birth (5).

Enthesis is a site of insertion of a tendon, ligament or articular capsule into bone. Endochondral ossification at this site is termed as enthesophyte. Enthesopathy may be developed in many disorders including traumatic, degenerative, diffuse idiopathic skeletal hyperostosis (DISH), inflammatory and metabolic diseases (13). Guo *et al.* (9) reported 4 patients with DISH who had ossification of the SHL in association with clinical findings compatible with Eagle's syndrome.

Stylohyoid apparatus might be an anatomic region in the cervical spine involved by enthesopathy. The aim of this study was to assess the elongation and/or ossification at the stylohyoid apparatus in the degenerative or inflammatory diseases such as AS, PsA and CS in which cervical spine involvement can be seen.

Material and methods

Twenty-eight patients with AS [5 women, 23 men, aged 19-57 years (mean 38.2 years)], 25 patients with PsA [18 women, 7 men, aged 19-65 years (mean 46.1 years)], 31 patients with CS [23 women, 8 men, aged 33-68 years (mean 49.3 years)] and 50 controls who did not have any complaints or symptoms related with elongated SP [29 women, 21 men, aged 18-65 years (mean 41.8 years)] were included in the study. The data from the patients with PsA were compared to the data from 50 control patients, but suitable control groups were created among these subjects for AS and CS groups.

AS was diagnosed by the modified New York criteria (14). Cervical spondylitis was diagnosed with clinical (inflammatory neck pain and pain during the movement) and/or radiological features of cervical involvement (15).

Clinical and radiological involvement of spinal and peripheral joint was assessed in the patients with psoriasis. Radiographs of both hands, feet, cervical, lumbar spine, and sacroiliac joints were obtained. The present study included those patients who showed clinical and/or radiological evidence of spinal or peripheral joint disease (16-20). Particular care was given to exclude other possible causes of articular symptoms, such as fibrositis and degenerative disc disease. PsA was classified into 7 categories according to Gladman *et al.* (21). The skin and nails were examined by a dermatologist.

Current severity of psoriasis was graded according to the Psoriasis Area Severity Index (PASI) score (22). Each finger nail was assessed for pitting, onycholysis, hyperkeratosis and dystrophy.

Cervical radiologic involvement was classified based on cervical spine radiographs of the patients with AS and PsA according to Salvarani *et al.* (15). Narrowing of the intervertebral disc space especially if accompanied by osteoarthrosis was not accepted as a positive finding (16). Lateral views of both feet were examined for the presence of calcaneal enthesopathy in the patients with AS and PsA.

All of the patients with CS had cervical pain and typical spondylarthrotic degenerative changes on the lateral and anteroposterior cervical radiographs.

On the lateral cervical radiographs, the anterior and posterior aspects of each vertebral body and intervertebral disk were carefully evaluated for the presence of syndesmophytes, osteophytes or ossification of the adjacent longitudinal ligaments in the patient group. Two examiners (ST, ZU) developed a semiquantitative scale to evaluate the bone excrescences. Bone excrescences were scored as grade 0: absent, grade 1: mild, grade 2: moderate and grade 3: large and completely fused with adjacent vertebras on lateral cervical radiographs. Narrowing of the intervertebral disc space especially if accompanied by osteophytosis was attributed to coincident osteoarthrosis and not accepted as a positive finding in the patients with AS and PsA.

The entire osseous length of the SP was measured as a unit using a transparent milimeter radiology ruler on the lateral and lateral oblique mandibular or cervical views, the anteroposterior views radiographs in the patients and controls (5, 23). Initially, films were evaluated by one radiologist blinded to the patients' clinical status. To confirm the lenght of SP, the same radiologist and a physiatrist together reviewed again all radiographs from patients and controls four months after the first evaluation blinded to the previous assessments. Interobserver or intraobserver variability assessments were not done. There was complete agreement between the examiners on the films at the final review. The best visualised and longer SP was evaluated for the analysis. We excluded all radiographs that were difficult to interpret for technical or anatomic reasons from the study. We consider the SP to be elongated (abnormal) when the entire osseous length of the bony process and/or the mineralized portion of the SHL exceeds 25 mm on the radiograph (3, 6, 8, 10, 24). For brevity the term elongated SP will be used in this report, encompassing the truly elongated SP and the ossified or calcified SHL.

Elongated SP was classified according to Langlais *et al.* (8) for the types of radiographic appearance and patterns of ossification.

Statistical analysis was performed using the SPSS package. Means were tested with the Mann-Whitney U test, Pearson's and Spearman correlation analysis and proportions by chi-square test. Statistical significance was set at p£ 0,05.

Results

Table I shows the clinical features of the patients with AS and PsA. Cervical spine involvement was slightly more frequent in the patients with PsA than AS. In both of the patients with AS and PsA calcaneal enthesopathy was found in similar frequency (Table I). According to the Gladman's classification the findings of PsA were as follows; oligooarthritis: 2 patients, polyarthritis: 5 patients, lumbar spine only: 12 patients, lumbar spine + oligoarthritis: 1 patient, lumbar spine + polyarthritis: 1 patient.

Radiological abnormalities of the cervical spine and the pattern of cervical spine involvement in the patients with AS and PsA are shown in Table II. Radiological evidence of cervical spine involvement was seen more often in patients with PsA (88%) than in patients with AS (71.4%). No patients had vertical penetration of the odontoid process. We found a significant difference in the length of SP between the patients with ankylosing pattern cervical involvement (mean 25.6 mm) and no cervical involvement (mean 15.8 mm) in the AS group ($p = 0.01$).

The prevalence of elongated SP was found 31.3% on the neck or mandibular radiographs in a review of 134 patients and controls. Elongated SP which the length of SP was greater than 25 mm was found in 9 (32.1%) of 28 patients with AS, in 15 (60%) of 25 patients with PsA, in 9 (29%) of 31 patients with CS and in 9 (18%) of 50 controls. Figures 1 a, b and c show the elongated SP in patients with AS, CS and a control subject.

Most of the elongated SPs were classified as elongated and pattern of the calcification was partially calcified. We did not detect completely calcified pattern in any of the patients.

There were statistically significant differences between the patients with AS and the control group and between the patients with PsA and the control group with respect to the length of SP ($p = 0.003$, 0.000, respectively). There was no statistically significant difference in the length of SP between the patients with CS and the control group ($p = 0.72$).

The dimension of syndesmophytes or ligamentous ossification of the cervical spine involving C5-6 intervertebral disk level were significantly correlated with the length of SP in the patients with AS ($p = 0.02$). The number of cer-

Table I. Clinical features of the patients with AS and PsA

	AS	PsA
Mean (SD) duration of arthritis (years)	11.8 (8.9)	3.6 (5.0)
Cervical spondylitis (%)	82.1	88.0
Calcaneal enthesopathy (%)	64.3	64.0
Mean (SD) duration of psoriasis (years)	-	10.5 (11.7)
Mean (SD) PASI score	-	7.3 (6.4)
Nail lesions (%)	-	32.0

(a)

(b)

(c)

Fig. 1. **(a)** Elongated SP classified as elongated and partially calcified pattern (**black arrowhead**) in a patient with ankylosing spondylitis. **(b)** Elongated SP process classified as segmented and calcified outline pattern (**black arrowhead**) in a patient with cervical spondylosis. **(c)** Elongated SP classified as elongated and partially calcified pattern (**black arrow**) in a control subject.

vical vertebral bodies involved by ligamentous ossification or syndesmophytes was borderline significantly correlated with length of the SP in the patients with AS ($p = 0.05$). There was no relationship between the syndesmophytes or ligamentous ossification of the cervical spine and the length of SP in the patients with PsA. The dimension of osteophytes of the cervical spine involving C4-5 intervertebral disk level and the number of cervical vertebral bodies involved by osteophytes had significant negative correlation with length of SP in the patients with CS ($p = 0.03$, 0.05, respectively). The total score of the bone excrescences was the highest in the AS group compared with PsA and CS.

There was no correlation between the patient's age and disease duration and length of the SP in the patients group. We did not detect any significant difference between the patients with or without cervical spondylitis or calcaneal enthesopathy with the length of the SP in the patients with AS and PsA. In addition there was no relationship between the severity of sacroiliac involvement and length of the SP in these groups. There was no significant difference between the patients with or without nail involvement with the length of the SP in the patients with PsA. The PASI scores did not have a significant correlation with length of the SHL.

Discussion

The origin of the elongated SP is a debatable issue. Apart from being a secondary elongation caused by merging with the confining ossified portion of the SHL, the SP may grow as a reactive response to certain stimuli, such as trauma. In such cases, it has been suggested that a real metaplasia takes place – that is, a direct transformation from the stylohyoid fibrocartilaginous tissue into osseous tissue. Some authors discuss degeneration of the SHL with the deposition of calcium salts in the fibrous tissue (4-6, 24).

Ossification or calcification of the SHL is a widespread phenomenon. Trying to compare our results with the data available from literature, we found very different values ranging from 2% to 84.4% (4, 5, 24, 25). Our result (31.3%) cannot be directly compared, and the discrepancies we met certainly rise from different radiographic criteria and patient selection. We agree with the concept of "mineralization in the stylohyoid ligament complex" as proposed by Ferrario *et al.* (24), since it is often impossible to distinguish calcification or ossification of the ligament and elongation of the SP.

The prevalence of styloid process-stylohyoid ligament apparatus abnormalities in patients with AS, PsA and CS has not been studied. Guo *et al.* (9) stated that various types of ossification as well as enlargement of the SP and SHL correlate significantly with ligamentous ossification or osteophytes of the cervical spine, and such a correlation could explain the presence of clinical findings compatible with Eagle's syndrome among some patients with DISH involving the cervical spine. They did not find a significant difference in the prevalence of various types ossification of SHL between the patients with and without thoracic spine DISH. We found that the length of SP was significantly longer in the AS and PsA groups than the controls. Stylohyoid apparatus abnormalities seen in our patients with AS and PsA represent an association of these alterations and spondyloarthropathies, or elongated SP might have been another manifestation of enthesopathy of the spondyloarthropathies.

The embryologic development of enthesis is characterized by the primitive tendon or ligament attaching to the cartilage. Metaplasia of fibroblasts at this attachment site results in the formation of fibrocartilage. This process extends further into the tendon or ligament, while the remainder of the cartilage undergoes endochondral ossification. Inflammation at fibrocartilaginous enthesis and subsequent fibrous and bony ankylosis is a characteristic feature of spondyloarthropathies (26). Enthesitis seems to play major role in spinal involvement. This may depend on the content and composition of proteoglycan and type II collagen or mechanic factors.

An alternative hypothesis proposes autoimmunity to cartilage, especially fibrocartilage of certain joints and enthesis (27). Remus *et al.* (28) found that ossification of the posterior longitudinal ligament associated with more severe cervical spine disease in the patients with AS. They suggested that ossification of the posterior longitudinal ligament might be another manifestation of enthesopathy of the spondyloarthropathies. We might speculate that the enthesopathy possibility is more probable. Because it is of cartilaginous origin, the SHL retains the potential to ossify.

The percentage of elongated SP was detected to be increased in patients with PsA (60%). Radiological evidence of the cervical spine being affected by the disease is a relatively common finding in patients with PsA, occuring in 35-75% of patients (15). In our study 88% of the patients with PsA had radiological abnormalities in the cervical spine. The greater prevalence of elongated SP might be related with the cervical spine abnormalities that may become striking in patients with psoriasis (29).

In general, a pattern similar to AS with syndesmophytes, apophyseal joint involvement, and ligamentous calcification is the most frequent radiological change in patients with PsA (15). Ankylosing disease subset of cervical spine involvement is the more frequently reported pattern which is similar to that seen in AS (15). Jenkinson *et al.* (16) revealed that no patients had vertical penetration of the odontoid process in patients with PsA. Our results were in agreement with these findings (Table II). In this study elongated SP was found to be associated with radiological features indicating more severe bone outgrowths or ankylosis at C5-6 intervertebral disc level and total number of involved vertebra in patients with AS. Total scores of dimension of syndesmophytes or ligamentous ossifications were also higher in the AS group than the patients with PsA. The severity and distribution of bone outgrowths in psoriatic spondylitis are different from in AS. Although facet joint space narrowing, sclerosis,

Table II. Radiological abnormalities of the cervical spine and pattern of the cervical spine involvement in the patients with AS and PsA.

	AS patients		PsA patients	
Radiological abnormality	no.	(%)	no.	(%)
Atlantoaxial subluxation	1	(3.5)	3	(12.0)
Odontoid erosion	13	(46.4)	10	(40.0)
Vertical penetration	0		0	
Facet joint narrowing	14	(50.0)	11	(44.0)
Facet joint fusion	14	(50.0)	8	(32.0)
Subaxial fusion	0		2	(8.0)
Subaxial narrowing	1	(3.5)	1	(4.0)
Subaxial subluxation	0		1	(4.0)
Subaxial syndesmophytes	11	(39.2)	11	(44.0)
Ligamentous calcification anterior	3	(10.7)	12	(48.0)
Ligamentous calcification posterior	7	(25.0)	11	(44.0)
Ligament calcification anterior and post.	1	(3.5)	8	(32.0)
Pattern of cervical spine				
Inflammatory	2	(7.1)	4	(16.0)
Ankylosing	18	(64.3)	18	(72.0)
Normal	8	(28.6)	3	(12.0)

and bone ankylosis may be seen, the severity of these findings is much less than in AS (29). Severity of SHL involvement might be related with ankylosing in cervical spine in spondyloarthropathies.

Our results revealed that there was no significant difference between the patients with CS and the controls with respect to length of SP. There was a possibility that asymptomatic radiologically degenerative structural changes also could occur at the cervical spine in the control group. Conversely, we found that osteophytes correlates with a decreased length of SP (negative correlation). This suggests that osteophytes of the cervical spine and styloid process-stylohyoid ligament apparatus abnormalities in patients with CS behave independently from each other. Correlation between DISH and ossification of the posterior longitudinal ligament, as well as ossification of the sacrotuberous ligament, has been reported (30). Ossification of the posterior longitudinal ligament was found to be associated with radiological features of sacroiliitis grade in the patients with AS (28). In our study the length of the SP was not associated with radiological sacroiliitis or calcaneal enthesopathic involvement in the patients with AS and PsA. Severity of skin disease did not predict development of PsA or cervical spine involvement (16, 17). We did not observe the relationship between the elongated SP and severity of skin disease.

Considering the extent of ossification versus age, there are very different conclusions. It was stated that elongated SP occur more frequently in older patients (4,24). This is explained by degeneration of the ligamentous apparatus linked to a general tendency toward the deposition of calcium salts (31). However, the study by Omnell et al (6) revealed that ossified sites of the SHL in most patients are established during childhood and adolescence. After age 20 the increase seemed nonlinear and occurred at a slower rate. Guo *et al.* (9) found that the older age of the patient correlates with a decreased frequency of certain types of ossification of the SP and SHL (negative correlation). In our study the length of the SP was not found to be associated with patients' age and the duration of the disease in all groups of patients.

In conclusion elongated SP might be another manifestation of enthesopaty in cervical spine of the spondyloarthropathies (AS and PsA). The prevalence of elongated SP was found to be much greater in PsA patients whereas the length of SP was found to be associated with more severe cervical spine ankylosis which was more prominent in patients with AS. Whether elongated SP may produce clinical findings compatible with Eagle's syndrome in these patients will require further study.

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