

# Ankylosing spondylitis and a diagnostic dilemma: coccydynia

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## Abstract

### Objective

Coccydynia is defined as pain in or around the tail bone area. The most common cause of coccydynia is either a trauma such as a fall directly on to the coccyx or repetitive minor trauma. The etiology remains obscure in up to 30% of patients. The literature on the contribution of rheumatic diseases to coccydynia is scarce. Our objective was to investigate the prevalence of coccydynia in ankylosing spondylitis (AS) patients.

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### Methods

One hundred and seven consecutive patients with AS were evaluated for coccydynia were enrolled between January and November 2012 for a cross-sectional analysis. Seventy-four consecutive patients were followed for mechanical back pain as controls and the AS patients were interviewed for the presence of coccydynia. The data collected was evaluated on SPSS<sup>®</sup> version 11.5 and Microsoft Excel<sup>®</sup> Programmes.

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### Results

Prevalence of coccydynia in AS (38.3%) was significantly higher than the control group ( $p < 0.0001$ ) in both female and male AS patients (female AS vs. control=40.9% vs. 18.4%,  $p = 0.015$  and male AS vs. control=36.5% vs. 8.0%,  $p = 0.005$ ). Both genders were affected equally in the AS group whereas coccydynia was slightly more frequent in female patients in the control group.

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### Conclusion

Coccydynia is a previously neglected symptom of AS and it is almost three times more common in AS than in non-specific chronic low back pain. Our observation may implicate that inflammatory diseases have a role in the etiology of coccydynia, especially in those without a history of recent or past trauma and coccydynia may be a factor associated with the severity of AS as well.

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### Key words

coccydynia, ankylosing spondylitis, coccyx, spondyloarthropathy

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Received on May 8, 2013; accepted in revised form on November 12, 2013.

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## Introduction

Coccydynia (coccygalgia or coccygodynia) is defined as pain in or around the tail bone area (os coccygis; coccyx). The term coccydynia was first used by Simpson (1) in 1859, but descriptions of pain in the terminal portion of the spine date back to at least 16<sup>th</sup> and 17<sup>th</sup> centuries (2-5). However, despite being recognised for many centuries, coccydynia remains an unsolved mystery because of the perceived uncertainty in identifying the origin of the pain (1). The most common cause of coccydynia is either a trauma such as a fall directly on to the coccyx or due to repetitive minor trauma that occurs due to sitting awkwardly such as during the post-partum period (6). Maigne *et al.* (7) reported that in a series of 51 patients 36 had a history of direct trauma, and Pennekamp *et al.* (8) reported a 50% incidence of direct trauma. Apart from those cases caused by local injury the etiology remains obscure in up to 30% of patients (5, 9, 10). Recently, a more precise classification of coccydynia based on etiology and pathology has been reported in an attempt to include rare causes of coccydynia (Table I) (1).

Coccygeal disorders that could be manifested in coccydynia are injuries (fracture, subluxation, luxation), abnormal mobility (hypermobility, anterior and posterior subluxation or luxation of the coccyx), disc degeneration at sacrococcygeal (SC) and intercoccygeal (IC) segments, coccygeal spicule (bony excrescence), osteomyelitis and tumors. Abnormal mobility of coccyx, which can be seen on dynamic radiograph (lateral x-rays of the coccyx in the standing and sitting position), is the most common pathological finding in patients with coccydynia (70%). It can be a result of injury and chronic static and dynamic overload of the coccyx (obesity, prolonged sitting, cycling, rowing, riding, etc.) (9).

The association of chronic non-specific low back pain with coccydynia has been reported repeatedly in the literature. Postacchini and Massobrio (11) reported that 31% of their patients had coccydynia-associated low back pain and in a different study of Bayne *et al.* (12) 15% of coccydynia patients presented

with preexisting back pain. Attempts to attribute coccydynia to lumbar disk hernia is not always justified since only a small proportion of patients ( $\approx 25\%$ , 13/50) with chronic low back pain have been found to have nerve compression and only a few of these patients had nerve root irritation syndromes when studied specifically (13).

The literature on the contribution of rheumatic diseases to coccydynia is scarce. In 1959, Hart and Robinson reported coccydynia as the initial symptom in 3% of female ankylosing spondylitis (AS) patients (14). Recent publications with MRI of the coccyx defined inflammation in the form of bone oedema and bursitis in adolescents and adults (15, 16). Bone oedema and bursitis on MRI are common findings associated with inflammation of any nature including inflammatory spine diseases such as AS.

Anatomically, os coccyx is the continuum of the vertebral column. Joints between sacrum, coccyx and segments of coccyx are fibrocartilaginous in nature and are comparable to those seen in higher intervertebral spaces. The anterior and posterior sacrococcygeal ligaments are the continuations of the anterior and posterior longitudinal ligaments that stretch along the entire spine. The anterior sacrococcygeal ligament attaches to the front of the first and sometimes the second coccygeal vertebral bodies, blending superiorly with the termination of the anterior longitudinal ligament (17).

Taken together, the close anatomical resemblance, recent MRI findings and the observation of coccydynia in some of the AS patients followed at our outpatient rheumatology clinic led to the following question; what is the contribution of ankylosing spondylitis to the etiology of coccydynia? The aim of this study was to investigate the prevalence of coccydynia in AS patients.

## Methods

### *Patients and data collection*

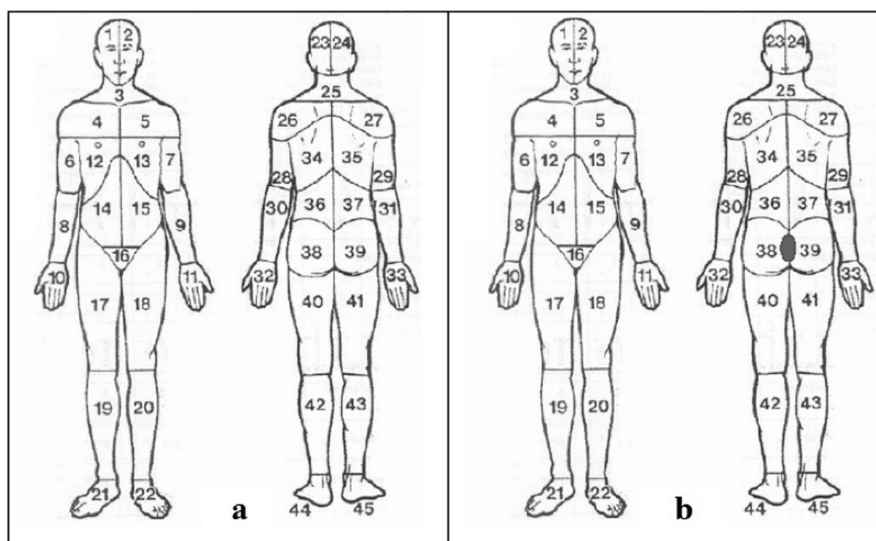
#### *- Study patients*

One hundred and seven consecutive patients with AS diagnosed according to the modified New York criteria (18) were enrolled in the study between

Competing interests: none declared.

**Table I.** Classification of coccydynia based on etiology and pathology.

A: Based on etiology	
	1. Idiopathic
	2. Traumatic
B: Based on pathology	
	1. Degeneration of the sacrococcygeal and intercoccygeal disc and joints
	2. Morphology of the coccyx: type II, III, IV, presence of a bony spicule and coccygeal retroversion
	3. Mobility of the coccyx: hypermobile or posterior subluxation
	4. Referred pain: lumbar pathology or arachnoiditis of the sacral nerve roots, spasm of the pelvic floor muscles and inflammation of the pericoccygeal soft tissues
	5. Others: neoplasm, crystal deposits, infections
C: Somatisation or neurotic	



**Fig. 1.** a. Illustration given to patients to show area of coccydynia. b. Expected area to be marked by patients as the area of coccydynia.

January 2012 and November 2012 for a cross-sectional analysis.

*- Control group*

Seventy-four consecutive patients followed for mechanical back pain (mainly for lumbar hernia, radiculopathy and lumbar strain) at the outpatient clinic of the Department of Physical Medicine and Rehabilitation, Marmara University Faculty of Medicine were enrolled to study as controls.

*- Exclusion criteria*

Patients with a history of trauma or current diagnosis of inflammatory bowel disease or psoriasis among study patients and patients fulfilling the modified New York criteria (19) or patients fulfilling the AS Assessment Study (ASAS) inflammatory back pain criteria

in the control group were excluded (19). The University of Marmara Institutional Review Board approved the study, and all study patients gave informed consent.

*Data collection*

Patients were interviewed face to face using a questionnaire consisting of nine questions regarding demographic data, disease onset and age at diagnosis, current medications and the presence of coccydynia. Figure 1a demonstrates the visual question used to point out the location of pain by the patient. However, in order not to bias the results, the tail bone area was not shown on purpose. Patients reporting coccydynia on questioning were asked to circle the area of pain on the figure to avoid any possible misconception about the “tail

bone area” of the study patients. The expected marking site of patients is shown in Figure 1b.

Collected data was evaluated on SPSS® version 11.5 and Microsoft Excel® Programmes. Data for patients were calculated as mean and standard deviations for continuous variables and as frequencies and percentages for categorical variables. The gender-based differences were analysed with  $\chi^2$  and Student’s *t*-test. For all analyses significance level of *p*-value was accepted as 0.05.

**Results**

One hundred and seven AS and 74 patients with mechanical back were included. Demographic features of AS and control participants are presented in Table II. A substantial proportion of our study patients with AS, but not with non-inflammatory chronic low back pain, defined coccydynia when questioned specifically. Prevalance of coccydynia in AS was 38.3% and was significantly higher in both male and female patients compared to the control group (41/107 in AS vs. 11/74 in control group,  $p<0.0001$ , OR: 2.58; CI:95%; 1.42-4.68). Both genders were affected equally in the AS group whereas coccydynia was slightly more frequent in female patients in the control group (23/63 male AS vs. 18/44 female AS,  $p=0.397$ ; 2/25 male control vs. 9/49 female control,  $p=0.204$ ) (Fig. 2).

The comparison of coccydynia presence according to gender shows significance for AS and control patients in both male (23/63 of male AS vs. 2/25 of male control;  $p=0.005$ ; OR: 4.56; CI:95%,1.16–17.93) and female patients (18/44 of female AS vs. 9/49 of female control;  $p=0.015$ ; OR: 2.27; CI:95%; 1.18–4.43).

In AS patients, disease activity assessed by Bath AS disease activity index (BASDAI) and treatment with biologic agents were compared for coccydynia presence. When each individual domain of BASDAI was evaluated for the affect of coccydynia, the question about the neck, back or hip pain had significantly higher score in patients with coccydynia (Table III). Overall BASDAI score was also higher in this subset of patients.

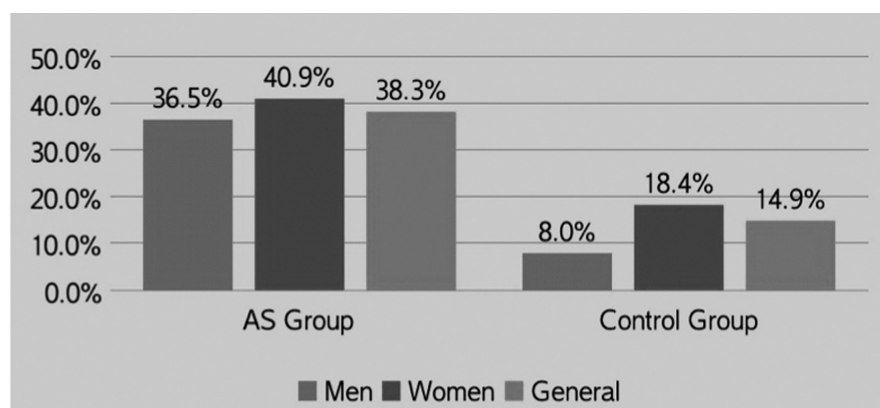


Fig. 2. Comparative proportions of patient with coccydynia.

Table II. Demographics of the study patients.

	AS Group			Control Group		
	Female n, (%)	Male n, (%)	Total n, (%)	Female n, (%)	Male n, (%)	Total n, (%)
Gender	44 (41.1)	63 (58.9)	107	49 (33.8)	25 (66.2)	74
Median age*	39	37	38	45	36	43
Mean age*	38.56	39.35	39.03	46.2	39.1	43.8
Time to diagnosis*	6.47	4.86	5.85	5.0	0.84	3.5
Age at diagnosis*	34.3	32.8	33.4	42.2	38.3	40.8

\*in years.

Table III. Evaluation of individual BASDAI domains according to coccydynia presence.

BASDAI question	Coccydynia present	Coccydynia absent	p-value
How would you describe the overall level of fatigue/tiredness you have experienced?	5.57	4.59	0.110
How would you describe the overall level of AS neck, back or hip pain you have had?	6.02	4.57	0.019
How would you describe the overall level of pain/swelling in joints other than neck, back or hips you have had?	3.75	3.18	0.373
How would you describe the overall level of discomfort you have had from any areas tender to touch or pressure?	4.30	2.93	0.054
How would you describe the overall level of morning stiffness you have had from the time you wake up?	4.75	4.04	0.290
How long does your morning stiffness last from the time you wake up?	3.70	2.44	0.057
Final BASDAI score	4.77	3.70	0.020

Although not significant, the need for anti-tumour necrosis factor  $\alpha$  (Anti-TNF  $\alpha$ ) therapy seemed also higher in patients with coccydynia, suggesting a higher disease severity (31.8% vs. 22.2%,  $p=0.20$ ).

## Discussion

Coccydynia (coccygodynia or pain in the tailbone) is a common, painful condition that is easily diagnosed. Coc-

cydynia resolves either spontaneously or with supportive care over weeks to months. But sometimes it is refractory to treatment and may debilitate the patient.

Most publications on coccydynia are case reports, case series, or reviews, and there are few extensive research studies on etiology. Persistent coccydynia may lead to unnecessary and often not evidence-based diagnostic

tests and interventions. A simple classification based solely on etiology is practical in guiding the management but leaves a substantial proportion of cases without a diagnosis. Contribution of rather infrequent causes, such as inflammatory diseases, on to the etiology of coccydynia is scarcely addressed in the literature. A considerable proportion of our study patients with AS – but not with non-inflammatory chronic low back pain – defined coccydynia when questioned specifically. This observation suggests that cases classified previously as “idiopathic” should carefully be questioned for AS (1).

Ankylosing spondylitis presents with chronic inflammatory back pain (IBP) early in life, generally when patients are in their teens or early to mid-20s (20) and causes considerable functional disability and also the socio-economic impact for the patients, the healthcare system and the society (21). This age of presentation overlaps with that of coccydynia, which also commonly occurs in adolescents and young adults (15, 22). Unfortunately, during the “pre-radiographic” phase, which on average might last 5 to 10 years or longer, AS remains clinically unrecognised (20, 23). The absence of radiographic sacroiliitis may lead to a possible mislabeling of cases especially without an acute or past trauma history as “idiopathic” or “chronic nonspecific back pain” (13). The association of coccydynia with AS is rarely addressed in the literature.

Lately, Maigne *et al.* (16) reported a series of MR imagings in patients with chronic coccydynia. In 70 of the 172 cases (40.7%), the vertebral endplates of the sacrococcygeal or intercoccygeal disc showed a hyperintense signal on T2 and isointense signal on T1-weighted images. In the majority of these cases (38 out of 70), the image was non-homogeneous, moderately intense, poorly delimited and sometimes associated with a slightly increased intradiscal signal on T2. These changes were interpreted as oedema due to joint inflammation, by analogy with the situation described by Modic *et al.* for the lumbar spine (24). Pathogenesis and association of these MRI changes are often not well understood yet (16). Until



recently, imaging of the painful coccyx relied on standard and dynamic radiography. Recent imaging techniques provided important additional data on the inflammation of the coccyx (16). MRI findings such as bone edema and bursitis around the coccyx could provide additional information on the cause of pain that escape detection with x-rays – and can potentially add value to the etiological diagnosis of coccydynia and improve our understanding of the mechanism(s) of pain. BASDAI domain for enthesitis was associated with the presence of coccydynia. This finding may implicate enthesitis as an etiologic factor of coccydynia in AS. Classically, the first coccygeal vertebra articulates with the sacrum via a symphyseal joint containing a thin intervertebral disc of fibrocartilage (25). Coccydynia is encountered predominantly in the female gender (1) while AS is more frequent among the male gender. Equal distribution of coccydynia within genders in AS and the predominance in female patients in the control group patients with chronic non-specific low back pain supports the notion that coccydynia is a previously neglected frequent symptom of AS. Furthermore, need for a more aggressive treatment with anti-TNF- $\alpha$  agents in patients with coccydynia and the association of the second domain of BASDAI which reflects axial involvement in AS suggests that coccydynia is a severity associated feature. The question, whether coccydynia parallels disease activity awaits further studies. Since the first definition of coccydynia in 1859 by Simpson several attempts have been made for classification leaving a substantial proportion of patients labeled as “idiopathic”. In the light of new imaging techniques the observa-

tion of AS patients with coccydynia implicates that inflammatory diseases may have a role in the etiology especially in those without a history of recent or past trauma.

#### Acknowledgements

We would like to thank our valuable colleagues Dr Turkan Cengiz, Dr Ela Karakayali, Dr Hulya Peynirci, Dr Hatice Erkal, Dr Alp Eren Celenlioglu and Dr Mustafa Fahri Kara for their support, and also all our patients for their participation in the study.

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