



## ORIGINAL ARTICLE

# Coccydynia in patients with axial spondyloarthritis: Reflection of enthesitis?

*Aksiyal spondiloartritli hastalarda koksadini entezitin yansıması mıdır?*

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## Summary

**Objectives:** In this study, we aimed to compare the Maastricht Ankylosing Spondylitis Enthesitis Score (MASES) values of patients with axial spondyloarthritis (ax-SpA) with and without coccydynia.

**Methods:** We included 42 cases between the ages of 18 to 65 that were admitted to our clinic between August 1, 2019 and April 20, 2020 with the diagnosis of ax-SpA. The first group consisted of 13 patients with coccydynia and the second group consisted of 29 patients without coccydynia. Besides the demographic data Ankylosing Spondylitis Disease Activity Score-C-reactive protein (ASDAS-CRP), Bath Ankylosing Spondylitis Functional Index (BASFI), and MASES values of the patients were recorded.

**Results:** We found that 13 (29.5%) of 42 patients with ax-SpA had coccydynia. While the rate of female patients in the coccydynia group was 46.15%, in the group without coccydynia, this rate was 31.03%. The mean of MASES, ASDAS-CRP, and BASFI values of the coccydynia group was statistically significantly higher than the group without coccydynia. We found that the BASFI was the most effective factor affecting the presence of coccydynia.

**Conclusion:** Our study supports the increased prevalence of coccydynia in patients with ax-SpA. In this study, we found that the presence of coccydynia may be associated with hypomobility rather than enthesitis.

Keywords: Axial spondyloarthritis; coccydynia; enthesitis.

## Özet

**Amaç:** Koksidini olan ve olmayan aksiyal spondiloartritli hastaların MASES (Maastricht Ankylosing Spondylitis Enthesitis Score) sonuçlarının karşılaştırılmasıdır.

**Gereç ve Yöntem:** 01/08/2019–20/04/2020 tarihleri arasında polikliniğimize başvurmuş, 18–65 yaş arası aksiyal spondiloartritli hastalar çalışmaya dahil edildi. Birinci grupta koksidini olan 13 hasta, ikinci grupta koksidini olmayan 29 hasta bulunmaktaydı. Hastaların demografik verileri ile beraber ASDAS-CRP, BASFI ve MASES sonuçları kaydedildi.

**Bulgular:** Aksiyal spondiloartritli 42 hastanın 13'ünde (29,5%) koksidini olduğu saptandı. Erkek hastaların daha ağırlıklı olduğu bu çalışmada koksidini olan grupta kadın hasta sayısı daha fazla idi. Koksidini olan grupta MASES, ASDAS-CRP ve BASFI skorlarının hepsi, koksidini olmayan gruba göre anlamlı derecede yüksekti. Bu parametrelerden koksidini varlığı ile ilişkisi en yüksek olan skorlama BASFI olarak bulundu.

**Sonuç:** Sonuç olarak çalışmamız, aksiyal spondiloartritli hastalarda artmış koksidini sıklığını desteklemektedir. Öte yandan çalışmamızda bulunan BASFI ile koksidini ilişkisi, entezitten çok hipomobilitate hipotezini desteklemektedir.

Anahtar sözcükler: Aksiyal spondiloartrit; koksidini; entezit.

## Introduction

Coccydynia is defined as pain felt around the tail bone area.<sup>[1]</sup> The term is first described by Simpson in 1859.<sup>[2]</sup> Although many years have passed since its identification, coccydynia remains an unsolved mystery due to some uncertainty in understanding the origin of pain. While external direct trauma or repetitive minor trauma are the most frequent

causes in etiology, chronic coccydynia with exacerbation and relief episodes without trauma is also a considerable subtype.<sup>[3]</sup>

Coccydynia may be related to post-traumatic fracture, luxation or subluxation, and abnormal mobility (hypermobility, hypomobility). Abnormal mobility can be assessed by dynamic lateral radiographs

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(standing and sitting).<sup>[4]</sup> Maigne et al.<sup>[5]</sup> reported a study of 51 patients, 36 had a history of direct trauma. Coccydynia is seen above 1% among spinal disorders.<sup>[6]</sup> The data about coccydynia in rheumatological diseases is quite limited. Hart and Robinson reported in 1959, that women with ankylosing spondylitis may have coccydynia as an initial symptom.<sup>[7]</sup> Deniz et al.<sup>[8]</sup> reported that coccydynia was observed 3 times more in patients with ankylosing spondylitis than patients with chronic mechanical low back pain. In studies conducted with magnetic resonance imaging (MRI), it has been reported that bone marrow edema and bursitis can be seen around the coccyx. These findings are similar to the inflammatory changes that can be seen in the spine in patients with spondyloarthritis.<sup>[9,10]</sup> The anterior and posterior sacrococcygeal ligaments are similar to the anterior and posterior longitudinal ligaments at the spinal level, which come from the sacrum and adhere to the coccyx.<sup>[11]</sup> Due to this close anatomical relationship and intense ligamentous structure we hypothesized that coccydynia might be a reflection of enthesitis in patients with axial spondyloarthritis (ax-SpA). In this study, we aimed to compare the Maastricht Ankylosing Spondylitis Enthesitis Score (MASES) values of patients with ax-SpA with and without coccydynia.

## Material and Methods

A cross-sectional and non-interventional trial was conducted in physical medicine and rehabilitation outpatient clinic. The study included 42 cases between the ages of 18 to 65 that were admitted to our clinic between August 1, 2019 and April 20, 2020 with the diagnosis of ax-SpA. Patients with a history of trauma or current diagnosis of inflammatory bowel disease, parturients, and patients with psychotic disorders were excluded. The cases were evaluated in two groups. The first group consisted of 13 patients with coccydynia and the second group consisted of 29 patients without coccydynia.

Besides the demographic data of the patients, the disease activity was assessed in subjects with axial SpA through the Ankylosing Spondylitis Disease Activity Score (ASDAS) including C-reactive protein (CRP) (ASDAS-CRP).<sup>[12]</sup> Functional status was assessed through the Bath Ankylosing Spondylitis

Functional Index (BASFI).<sup>[13]</sup> The enthesitis were assessed through the MASES which records tenderness with palpation as either present (1) or absent (0) on each enthesial site. MASES analyses 13 sites: the fifth lumbar spinous process and six entheses on both sides (first and seventh costochondral joints, posterior superior iliac spine, anterior superior iliac spine, iliac crest, and Achilles tendon insertion).<sup>[14]</sup>

## Statistical Analysis

Statistical analyzes were done with Number Cruncher Statistical System 2007 Statistical Software (Utah, USA) package program. In the evaluation of the data, besides descriptive statistical methods (mean, standard deviation), Shapiro - Wilk normality test was used to analyze the variables, independent t-test was used for comparisons of variables with normal distribution, and Chi-square test was used to compare qualitative data. Logistic regression analysis was performed to determine the factors affecting the presence of coccydynia. The results were evaluated at the significance level of  $p < 0.05$ .

## Results

It was determined that 13 (29.5%) of 42 patients with ax-SpA had coccydynia. While the rate of female patients in the coccydynia group was 46.15%, in the group without coccydynia, this rate was 31.03%. There was no statistically significant difference between the mean age, gender distribution, biological agent use rates, and disease duration between the two groups ( $p > 0.05$ ) (Table 1).

In both groups, patients generally had high disease activity (ASDAS-CRP  $> 2.1$ ). The mean of MASES, ASDAS-CRP, and BASFI of the coccydynia group was statistically significantly higher than the group without coccydynia (Table 2).

To determine the factors affecting the presence of coccydynia, MASES, ASDAS-CRP, and BASFI variables were evaluated by using logistic regression analysis, and the most effective factor was determined as BASFI ( $p = 0.018$ ) (Table 3). The positive predictive and negative predictive value of MASES with a cut-off value 4, was found 71.4, and 89.3 respectively. Likelihood ratio was 5.58.

**Table 1.** The distribution of the demographical features among groups

	Coccydynia (+) n=13		Coccydynia (-) n=29		p
	n	%	n	%	
Age	48.31±11.94		43.14±10.73		0.171
Gender					
Female	6	46.15	9	31.03	0.344
Male	7	53.85	20	68.97	
Biological agent treatment	4	30.7	12	34.8	2.225
Disease duration	8.85±5.32		6.83±4.92		0.238

**Table 2.** The distribution of the clinical features among groups

	Coccydynia (+) n=13	Coccydynia (-) n=29	p
MASES	4.92±1.32	3.14±1.48	0.001
ASDAS-CRP	3.02±0.6	2.31±0.63	0.002
BASFI	4.62±0.87	2.89±1.08	0.0001

MASES: Maastricht Ankylosing Spondylitis Enthesitis Score; ASDAS-CRP: Ankylosing Spondylitis Disease Activity Score-C-reactive protein; BASFI: Bath Ankylosing Spondylitis Functional Index.

**Table 3.** Logistic Regression Analysis of the indexes

	B	p	OR	95% CI for OR	
				Lower	Upper
MASES	0.15	0.756	1.16	0.46	2.93
ASDAS-CRP	0.53	0.611	1.71	0.22	3.38
BASFI	1.44	0.018	4.21	1.28	8.86

OR: Odd ratios; CI: Confidence interval; MASES: Maastricht Ankylosing Spondylitis Enthesitis Score; ASDAS-CRP: Ankylosing Spondylitis Disease Activity Score-C-reactive protein; BASFI: Bath Ankylosing Spondylitis Functional Index.

## Discussion

In this study, we aimed to investigate the MASES values of the axial-SpA patients with and without coccydynia. Patients with coccydynia were found to have significantly higher MASES values than the group without coccydynia ( $p=0.001$ ). In addition, we found the frequency of coccydynia to be 29.5% in patients with axial SpA, seems higher when compared to general population.<sup>[15–17]</sup> We found that BASFI and ASDAS-CRP scores of patients with coccydynia were also higher than the group without coccydynia. When we examine the factors affecting the presence of the coccydynia with logistic regression analysis, BASFI was found as the most effective factor ( $p=0.018$ ).

Coccydynia is a common and unpleasant condition which can be easily diagnosed. Patients' main complaint is pain, which typically is triggered by sitting position, especially on hard surfaces. Furthermore, the pain may increase with sexual intercourse and during defecation. Axial trauma and the childbirth are known as the most common causes of coccydynia which may lead to abnormal mobility of the coccyx.<sup>[18]</sup> An important part of coccydynia is still classified as idiopathic.<sup>[1]</sup> There is limited data available about coccydynia in rheumatologic diseases.<sup>[7]</sup> Deniz et al.<sup>[8]</sup> reported that the prevalence of coccydynia in ankylosing spondylitis was significantly higher than the mechanical back pain group. They also assessed the disease activity in the study group using the Bath Ankylosing Spondylitis disease activity index and they found that the patients with coccydynia had significantly higher scores. Our study supports that patients with coccydynia classified as 'idiopathic' should carefully be examined for axial SpA.

The coccyx is surrounded by many ligaments including the sacrococcygeal ligaments, the sacrospinous ligament, the sacrotuberous ligament. These ligaments help to support the pelvic floor. Anterior and posterior sacrococcygeal ligaments have similar function to the anterior and posterior longitudinal ligaments at the spinal level, which attach the sacrum to the coccyx.<sup>[11]</sup> The Romanus lesion is one of the earliest signs in the spine of ax-SpA. The lesion is an enthesitis limited to the junction of the anterior longitudinal ligament and annulus fibrosus.<sup>[19]</sup> The lesion begins with acute inflammatory process and erosion at the vertebral body corners (low T1 and high T2 signal) where the ALL attaches. Then, the process goes fatty replace-

ment (high T1 and T2 signal) and finally sclerosis (low T1 and T2).<sup>[20]</sup> Maigne et al.<sup>[10]</sup> first described the soft tissue lesions around the coccyx with an MRI study. The soft tissue lesions, i.e., bursitis, and also bone marrow edema seem similar to the spinal MRI findings of patients with ax-SpA. In this study, in terms of coccydynia, the positive predictive and negative predictive value of MASES with a cut-off value 4, was found 71.4 and 89.3, respectively. Likelihood ratio was 5.58 which means the patients with a MASES value >4, have the risk of coccydynia 5.58 times more than a patient with a lower score.

The majority of coccydynia cases are associated with subluxation or hypermobility. The general opinion is that instability may lead to chronic inflammatory changes and pain.<sup>[5,21]</sup> However, Postacchini and Massobrio reported that there was no statistical difference between asymptomatic patients and those with coccydynia in terms of the numbers of coccygeal segments or the incidence of fusions between the segments.<sup>[22]</sup> Ankylosing spondylitis is often characterized by hypomobility in the spine, in contrast with hypermobility of the sacrococcygeal joint usually seen in patients with coccydynia. Maigne et al.<sup>[10]</sup> also found that the cases with bursitis are associated with a rigid coccyx that maybe explained by the absence of flexion, tip of the coccyx during sitting may lead to exert pressure and friction on the subcutaneous tissues. In this study, we also found that BASFI was the most effective factor affecting the presence of coccydynia. Coccydynia in patients with ax-SpA may be associated with hypomobility rather than enthesitis.

## Conclusion

In conclusion, our study supports the increased prevalence of coccydynia in patients with ax-SpA. There are some limitations that we did not use MRI or X-ray besides the clinical assessment tools. Second, we had a relatively small sample size. Patients were also generally older and the biologic agent use rate was low. Further studies are required with dynamic X-ray or MRI, in larger groups to understand the etiology in those with rheumatologic diseases.

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