Immunohistochemical analyses to determine pathogenesis of tenosynovitis with psammomatous calcification in the wrist: A case report

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Abstract. Tenosynovitis with psammomatous calcification is an extremely rare clinicopathological condition, which is characterized histopathologically by the presence of numerous psammomatous calcifications surrounded by a granulomatous reaction comprising a mixture of histiocytes and fibroblasts. The pathogenesis of this disease remains unclear, although an association with repetitive tendinous injury has been proposed. The present study describes the details of a case in an elderly Japanese female, and, to the best of our knowledge, the first known immunohistochemical analysis of the mechanism underlying psammomatous calcification formation. A 66-year-old Japanese woman presented with pain in the right wrist. The lesion was surgically resected. Histopathological examination revealed a well-circumscribed lesion composed of psammomatous calcification. The calcification was surrounded by histiocytes, and a few multinucleated giant cells and fibroblastic spindle cells. Immunohistochemical study revealed that these histiocytes were positive for cluster of differentiation 163, and the histiocytes and spindle cells surrounding the psammomatous calcification expressed bone morphogenetic protein-1 (BMP-1). Tenosynovitis with psammomatous calcification is hypothesized to be a distinctive subtype of idiopathic calcifying tenosynovitis involving an unusual reactive or degenerative process. BMP-1 has been demonstrated to be involved in the regulation of hard tissue mineralization, and its expression has been suggested to be associated with psammoma formation in papillary thyroid cancer. To the best of our knowledge, the case report within the present study suggested for the first time that BMP-1 expression was associated with development of psammomatous calcification in this condition.

Introduction

Tenosynovitis with psammomatous calcification is an extremely rare clinicopathological condition. Since it was first described by Gravanis and Gaffney in 1983 (1), only a few additional cases have been examined in the English language literature (2-5). This variant of calcifying tenosynovitis or calcific tendonitis is characterized histopathologically by the presence of numerous psammomatous calcifications surrounded by a granulomatous reaction comprising a mixture of histiocytes and fibroblasts (2,5). Recently, Michal et al (6) investigated a large case series of this disease, and confirmed that the disease exhibited a tendency to affect the fingers or toes of young to middle-aged women, and appeared to be associated with trauma and/or repetitive activity. They concluded that tenosynovitis with psammomatous calcification is a distinctive trauma-associated subtype of idiopathic calcifying tenosynovitis.

However, the pathogenesis of tenosynovitis with psammomatous calcification remains unclear. Although an association of this disease with repetitive tendinous injury has been described previously (2,6), other studies have described cases without a history of trauma (3,4). Bone morphogenetic protein (BMP)-1, also known as procollagen C-peptidase, is a multifunctional protein regulating of hard tissue mineralization (7). BMP-1 expression has been suggested to be associated with ectopic ossification (8) and psammoma formation in papillary thyroid cancer (9). The present study described a case report of tenosynovitis with psammomatous calcification that occurred in the wrist of an elderly female, and the immunohistochemical analysis of the mechanism of psammomatous calcification formation, particularly association with BMP-1 expression.

Case report

A 66-year-old Japanese female presented with pain in the right wrist. She had a history of De Quervain's disease and infliximab use for ulcerative colitis, but no history of trauma to the...
Discussion

In the present report, a case of tenosynovitis with psammomatous calcification was described. In addition, to the best of our knowledge, this was the first time immunohistochemical analysis was used to identify the potential mechanism of psammoma formation. Table I summarizes the clinicopathological features of the case in the present study, and those of previously described cases. As demonstrated, this disease affects patients with a median age of 44 years (14-83 years), with a female predominance (male:female ratio, 4:30), particularly in young to middle-aged women. Of the patients examined previously and in the present study, 12 of 25 had a history of trauma or repetitive activity. The majority of cases occurred in the hand, in particular in the finger, or the foot, and the most common complaint was a painful mass. A previous study involving the largest case series revealed these aforementioned clinicopathological features of tenosynovitis with psammomatous calcification (6), which is believed to be a distinct clinicopathological condition involving an unusual reactive or degenerative process in the chronically traumatized tendon and peritendinous tissue (2.6). However, the underlying molecular mechanism of development of this disease remains unclear.

BMP-1, also known as procollagen C-peptidase may convert a variety of precursor proteins, including procollagen and dentin matrix protein, into active forms, resulting in their involvement in cell adhesion and the regulation of hard tissue mineralization (7). Therefore, the present study focused on the association between psammomatous calcification of this lesion and BMP-1 expression. From the data in the present study, the expression of BMP-1 in histiocytes and spindle cells surrounding the psammomatous calcification was clearly observed. This suggests that the expression of BMP-1 may be associated with the development of psammomatous calcification.

In conclusion, the present study described a typical case of tenosynovitis with psammomatous calcification and reviewed its clinicopathological characteristics. The results suggested that the expression of BMP-1 in the histiocytes and spindle cells surrounding the psammomatous calcification may be associated with development of this condition.

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Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contributions

CM and MI were responsible for the conception and design of the study. CM, MI, YH, TT and KT were involved in the acquisition and analysis of the data. CM and MI drafted the manuscript. The final version of the manuscript was read and approved by all authors.

Ethics approval and consent to participate

This study was conducted in accordance with the Declaration of Helsinki, and written consent was obtained from the patient.
Patient consent for publication

Written informed consent for publication was obtained from the patient.

Competing interests

The authors declare that they have no competing interests.

References