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Unprecedented recurrence of carpal tunnel syndrome by metaplastic synovial cyst in the carpal tunnel

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ABSTRACT

Metaplastic synovial cyst (MSC) is a benign cystic lesion observed after surgical intervention and recurrent skin trauma. Because of its rarity, the etiology is not fully understood. The most emphasized etiologic factors are recurrent surgical procedures and cutaneous pathologies, which cause cutaneous fragility and abnormal wound formation. In the literature, MSC is exemplified as a mass that can be observed by the naked eye and palpated. All patients had a history of previous surgical procedures in the area. In the present case, we report a 48 -year-old woman with recurrent carpal tunnel syndrome due to a MSC. This report showed that MSC can be detected at deeper locations than the regions described in the literature. To our knowledge, this is the first report of MSC causing carpal tunnel syndrome recurrence. It is thought that previous operations are the most important etiologic factor in MSC occurrence.

Introduction

Metaplastic synovial cyst (MSC) is a cystic lesion consisting of a membrane covered with hyperplastic synovial villi histopathologically.[1] MSC also known as synovial metaplasia of the skin, was first described by Gonzalez and his colleagues in bone or soft tissues following surgical procedures or under experimental conditions.[1] MSC is a rare cyst variety and usually appears subcutaneously as a tender solitary nodule.[2] Although it is very rare in the literature, it occurs especially after recurrent trauma and repetitive surgery[1,3–8]; it is usually detected in the surgical field and is frequently be drawn with suture granuloma.[2] Other than cases involving surgical intervention, in Ehlers-Danlos syndrome, which causes cutaneous fragility and abnormal scar development, it is observed as a result of recurrent microtrauma.[4,9,10]

Herein, we report a case of deep located MSC causing carpal tunnel syndrome recurrence with multiple surgical procedures as the primary etiology.

Case description

A 48 -year-old woman was admitted to our polyclinic complaining of numbness, pain, tingling, and weakness in the thumb and thenar eminence of the right hand. The patient had undergone two surgeries for carpal tunnel syndrome (CTS) 5 years and 2 years ago. Tinnel test and scratch collapse test were found to be significant. No mass was detected by palpation therefore imaging was not needed. Electromyography (EMG) was performed. Based on a physical examination and electromyography (EMG) the patient was thought to have CTS recurrence, and surgery was thus planned. The operation was performed after approving informed consent form by the patient. During surgery we did not observe a thick scar tissue formation or adhesions which also may cause the recurrent carpal tunnel syndrome. Intraoperatively, after the transvers carpal ligament was opened to reach the median nerve via the incision used in previous surgery, a cystic structure was observed on the flexor tendon sheath (Fig. 1A). The cyst was smooth and 15 × 10 × 10 mm in size. The cyst was noted to have pushed the median nerve into the radial region (Fig. 1B). Gentle dissection showed no anatomical

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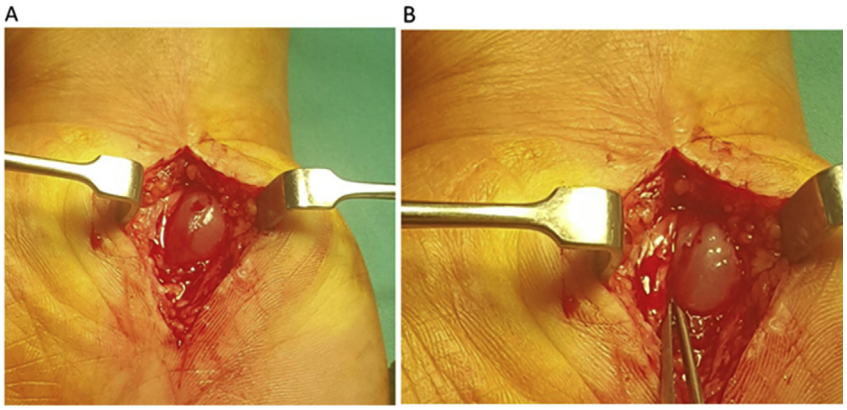

Fig. 1. A. Cystic structure observed in the carpal tunnel. **B**. The cyst is pushing the median nerve into the radial region.

connection between the median nerve and cyst. Cyst was thought to be ganglion cyst at first glance therefore puncture was performed, but aspirate could not be obtained. The cyst was separated fully and excised. Finally, median nerve release was completed. The cyst was sent to the pathology department for histopathological examination. Histopathologically, the villous structures were lined by a membrane of varying cellularity, which mimicked the synovium. Some villous structures were composed of hyalinized connective tissue, whereas others were highly cellular and lined with multiple layers of epithelial and fibroblast cells (Fig. 2A). Immunohistochemistry revealed that the cells lining the cyst were positive for vimentin (Fig. 2B), but negative for CD68 (Fig. 2C). Histopathological examination indicated that the cyst material as compatible with a diagnosis of MSC. The patient had no complaints 19 months postoperatively.

Discussion

MSC is a benign cystic lesion observed after surgical intervention and recurrent skin trauma.[1,3–8] MSC can occur in many parts of the body; however, in many cases, it is superficially located and visible by the naked eye.[2] There are few articles that reported deep location of MSC.[7,11] Our case is the first reported MSC in literature that is deeply located in carpal tunnel.

Metaplastic synovium-like tissue usually forms after trauma or surgery during maturation of multipotent cells at the connective tissue repair stage.[1] All reported patients had a history of previous surgical procedures in the area, and the lesions clinically resembled suture granulomas. The gap between the skin during tissue repair or internal suture application can stimulate the formation of synovial metaplasia. MSC may interfere usually epidermal cyst or suture/foreign body granuloma and it sometimes misdiagnosed as a mucous cyst by oral and maxillofacial surgeons but MSC can often interfere especially with the ganglion cyst. Ganglion cysts are synovial cysts that are filled with gelatinous mucoid material and commonly encountered in orthopedic clinical practice.

Recurrent carpal tunnel syndrome after previous surgery may be caused by inadequate release, scar tissue formation or occupying lesions which are located in carpal tunnel. In this case we did not make any radiological evaluation based on physical examination findings and EMG results but one should remember that the space occupying lesions may cause recurrent CTS and ultrasonography or magnetic resonance imaging might be necessary for diagnosis especially for recurrent suspected cases. When cystic lesion was detected, it was thought that it would be more appropriate to have a radiological evaluation. Our case was accepted as recurrent because he had an initial relief of symptoms after the index procedure, for a 6-month interval. The absence of preoperative radiological examination, although the symptoms indicate a pure carpal tunnel recurrence, can be stated as the weak side of the case.

Palmar masses can cause CTS as occupying lesions. Most of the palmar soft tissue masses are benign.[12] Ganglion cysts and giant cell tumors of the tendon sheath are the commonest lesions. Also, giant cell tumors of the tendon sheath, lipomas, Dupuytren's contractures, nerve sheath tumors, glomus tumors, hemangioma/vascular malformations can be accounted as solid masses.[13] The common characteristics of these masses are that they are palpable, have pain and usually have a slow growth pattern.[14] In our case, there was no palpable mass or pain. Therefore, we could not consider radiological evaluation with MRI or US.

Malignant soft tissue tumors of the hand are uncommon. The lesions most often encountered are undifferentiated pleomorphic sarcoma in the older population, synovial sarcoma, rhabdomyosarcoma, malignant nerve sheath tumors, liposarcomas and extraskeletal chondrosarcoma. The rapid growth pattern is one of the specific sign of these malign masses.

Ganglion cysts are the most common masses of the hand and also they may cause CTS recurrence as occupying lesions. Histopathologically ganglion cysts are mucin filled synovial cysts containing paucicellular connective tissue. Typical histopathological appearance is a mucin-filled synovial cell lined sac without a true epithelial lining.[15] MSC has been identified as a pseudocyst without an epithelial lining with a large papillary and villous projection observed along the center of the cavity inside the cyst, also Positive immunoreactivity of lining cells against vimentin, but not against cytokeratin, CEA, or S-100,3 together with the absence of expression of synovium marker CD68, supports the diagnosis. In our case immunohistochemistry study showed vimentin positive and CD68 negative.[11,16] With the help of all these features, it is possible to distinguish between MSC and ganglion cyst.

Trauma is the most common etiology for MSC. In the review of literature performed by Fukuyama declared that just 30 cases have been reported in the literature and 17 cases of MSC (59%) developed at the scar site of operations or trauma.[17] Our case is the 18th case in literature reported after trauma. In addition, it can be brought about by certain diseases, such as Ehlers-Danlos syndrome, which may cause deterioration of the connective tissue and lead to MSC formation.[4,5,10] Despite all this studies, because MSC is very rare, the causes have yet to be fully determined.

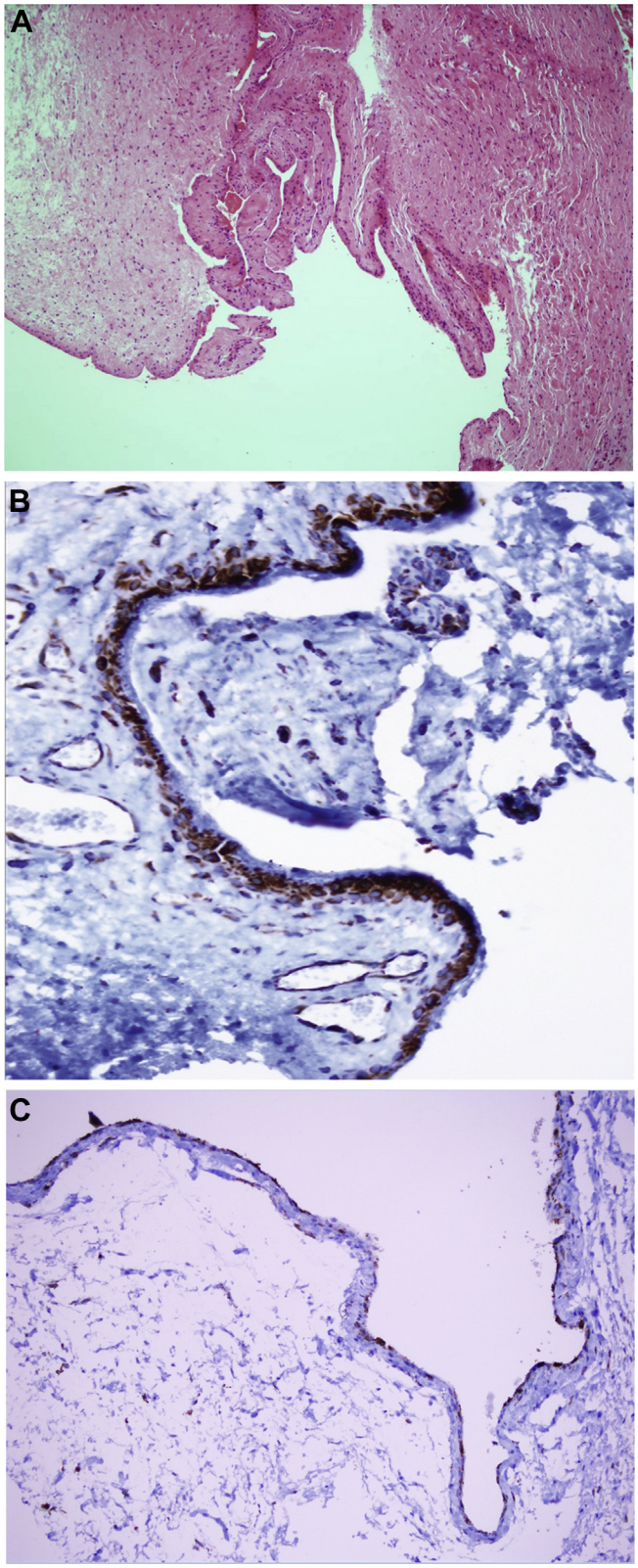

Fig. 2. A. The villous structures were lined by a membrane of varying cellularity, which mimicked the synovium. Some villous structures are composed of hyalinized connective tissue, whereas others were highly cellular and lined with multiple layers of epithelial and fibroblast cells Hematoxylin and Eosin staining (×100). **B.** The cells lining the cyst were positive for vimentin (×400). **C.** Immunohistochemistry negative for CD68 (×100).**C.** The cells lining the cyst were negative for CD68 (×200).

MSC has a low recurrence rate. Local recurrence has been reported in just oral and maxillofacial surgery but no recurrence has been detected after dermatologic surgery. This can be explained by the technical difficulties of surgical excision associated with anatomic location. Close follow-up may be recommended for patients with a risk of recurrence like repetitive surgery, trauma or underlying diseases that predispose to the development of MSC.[17] The recommended method of MSC treatment is excision.[18] In our case, MSC was believed to have caused CTS recurrence and excision of the cyst is curative surgical treatment.

Conclusion

MSC is a benign and rare cyst. It mostly occurs in superficial locations. Contrary to the literature described this study shows that the MSC may be determined at deeper locations and can lead to recurrent carpal tunnel syndrome. On the other hand In patients with recurrent CTS, space-occupying lesions and malignant tumors should be considered as etiologic factors and preop radiological evaluation should not be neglected. Otherwise, misdiagnosis and inadequate treatment may be inevitable.

Disclosure of interest

Authors declares that they have no competing interest.

References

1. Gonzalez JG, Ghiselli RW, Santa Cruz DJ. Synovial metaplasia of the skin. *Am J Surg Pathol*. 1987;11:343–350.
2. Kim BC, Choi WJ, Park EJ, et al. Cutaneous metaplastic synovial cyst of the first metatarsal head area. *Ann Dermatol*. 2011;23:S165–S168.
3. Bhawan J, Dayal Y, González-Serva A, Eisen R. Cutaneous metaplastic synovial cyst. *J Cutan Pathol*. 1990;17:22–26.
4. Nieto S, Buezo GF, Jones-Caballero M, Fraga J. Cutaneous metaplastic synovial cyst in an Ehlers-Danlos patient. *Am J Dermatopathol*. 1997;19:407–410.
5. Gómez Dorronsoro ML, Martinez-Peñuela JM, Ruiz de la Hermosa J. Metaplastic synovial cyst. *Am J Surg Pathol*. 1988;12:649–650.
6. Singh SR, Ma AS, Dixon A. Multiple cutaneous metaplastic synovial cysts. *J Am Acad Dermatol*. 1999;41:330–332.
7. Lin YC, Tsai TF. Cutaneous metaplastic synovial cyst: unusual presentation with "a bag of worms". *Dermatol Surg*. 2003;29:198–200.
8. Chakravarthy KM, Lavery KM, Barrett AW. Recurrent cutaneous metaplastic synovial cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2007;103:e42–e44.
9. Goiriz R, Ríos-Buceta L, Alonso-Pérez A, et al. Cutaneous metaplastic synovial cyst. *J Am Acad Dermatol*. 2005;53:180–181.
10. Guala A, Viglio S, Ottinetti A, et al. Cutaneous metaplastic synovial cyst in Ehlers-Danlos syndrome: report of a second case. *Am J Dermatopathol*. 2008;30:59–61.
11. Beham A, Fletcher CD, Feichtinger J, Zelger B, Schmid C, Humer U. Synovial metaplasia of the skin. *Virchows Arch A Pathol Anat Histopathol*. 1993;423:315–318.
12. Johnson J, Kilgore E, Newmeyer W. Tumorous lesions of the hand. *J Hand Surg Am*. 1985;10:284–286.
13. Capelastegui A, Astigarraga E, Fernandez-Canton G, et al. Masses and pseudomasses of the hand and wrist: MR findings in 134 cases. *Skeletal Radiol*. 1999;28:498–507.
14. Teh James. Ultrasound of soft tissue masses of the hand. *J Ultrasonograph*. 2012;12:381–401.
15. Gregush RE, Habusta SF. *Cyst, Ganglion*. StatPearls.Treasure Island (FL): StatPearls Publishing; 2017 Dec 5, 2018 Jan-. 2017 Dec 5.
16. Ramdial PK, Singh Y, Singh B. Metaplastic synovial cyst in male breast. *Ann Diagn Pathol*. 2005;9:219–222.
17. Fukuyama M, Sato Y, Hayakawa J, Ohyama M. Cutaneous metaplastic synovial cyst: case report and literature review from the dermatological point of view. *Keio J Med*. 2017;66:9–13.
18. Majdi M, Saffar H, Ghanadan A. Cutaneous metaplastic synovial cyst: a case report. *Iran J Pathol*. 2016;11:423–426.