CASE REPORT

Cutaneous Metaplastic Synovial Cyst of the Cheek Generated by Repetitive Minor Trauma

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Cutaneous metaplastic synovial cyst (CMSC) refers to a cyst lined by metaplastic synovial tissue including villous structures. It is thought to be a tissue reaction to local trauma, and most cases have a history of preceding surgery or trauma. Clinically, most of the lesion is a tender intradermal nodule that is associated with scar tissue. However, dermatologists have difficulty in diagnosing CMSC because CMSC is reported only rarely and its clinical manifestation can be confused with implantational epidermal cyst, suture granuloma or other cutaneous cysts. Thus, a histopathological exam is mandatory for diagnosis of CMSC. Herein we report the case of an 18-year-old man who developed CMSC on the left cheek on the posterior ramus of the mandible. He had a history of repetitive digital manipulation of the lesion before the onset of CMSC. **(Ann Dermatol 23(S2) S235～S238, 2011)**

-Keywords-
Cyst, Trauma

INTRODUCTION

Cutaneous metaplastic synovial cyst (CMSC) is a rarely described cystic tumor that is thought to be a tissue response to local trauma[1-7]. Clinically, the lesion is a tender intradermal nodule that is frequently related to scar tissue, and histologically, the cystic cavity has a lining resembling hyperplastic synovium. CMSC may be frequently misdiagnosed because it is rarely reported and unknown to many dermatologists. Here we report a case of solitary CMSC on the left cheek on the posterior ramus of the mandible in an 18-year-old Korean man following repetitive manipulation of the lesion before onset of CMSC. To our knowledge, this is the first published case of CMSC in the Korean dermatological literature.

CASE REPORT

An 18-year-old Korean man was referred for evaluation of a tender nodule on his left cheek on the posterior ramus of the mandible (Fig. 1A). He had no remarkable past medical history except receiving topical treatment for facial acne. Six months before this visit, he had noticed a papular lesion that was suspected to be acne on his left cheek. He had stabbed the lesion with a needle and had habitually manipulated the lesion. Subsequently, a nodular lesion developed on the previously manipulated site, gradually enlarged, and became painful 1 month prior to his presentation at our clinic. Two days prior, the nodular lesion was punctured at a local clinic and a serosanguinous discharge drained from the punctured orifice. Clinically, there was no erythema or heatness around the lesion indicating local infection, and punctured orifice are observed in the center of the nodule (Fig. 1A). An ultrasonographic exam revealed a 1.31×0.62 cm sized hypoechoic oval structure with posterior enhancement (Fig. 1B). Clinical impressions included epidermal cyst, furuncle and acne. A further excision was performed for the nodular lesion on the left cheek.

A histopathological exam from the nodular lesion

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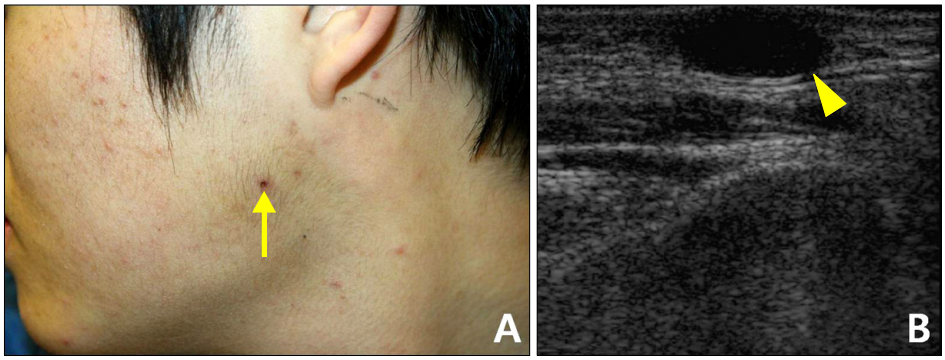

Fig. 1. (A) A localized, single, skin-colored nodule with a puncture opening on the left posterior ramus of the mandible (arrow). (B) A hypoechoic oval structure with posterior enhancement measuring 1.31×0.62 cm (arrowhead).

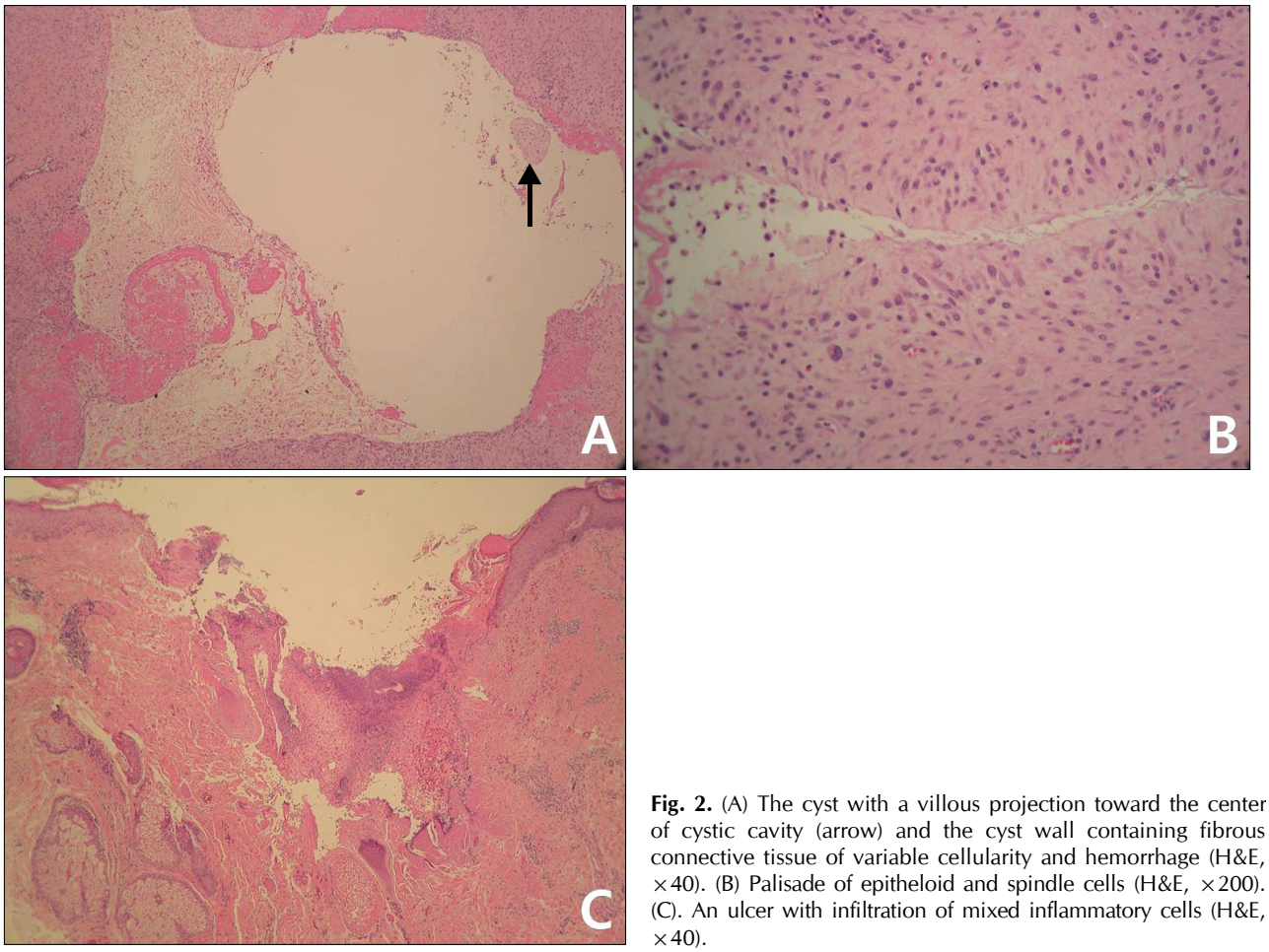

Fig. 2. (A) The cyst with a villous projection toward the center of cystic cavity (arrow) and the cyst wall containing fibrous connective tissue of variable cellularity and hemorrhage (H&E, ×40). (B) Palisade of epitheloid and spindle cells (H&E, ×200). (C). An ulcer with infiltration of mixed inflammatory cells (H&E, ×40).

revealed a cyst containing a villous projection and fibrin, and was lined by a membrane mimicking synovium with various degree of cellularity (Fig. 2A). The lining was composed of a mixture of epitheloid and spindle cells forming a cellular palisade (Fig. 2B). An ulcer was thought to be connected with the cyst (Fig. 2C). On the basis of characteristic histologic findings, the nodular lesion was diagnosed as cutaneous metaplastic synovial cyst. The entire lesion was excised during the biopsy. There was neither recurrence nor complications during the 3 months of follow-up after the excision.

DISCUSSION

In 1953, Selye[8] showed that cutaneous cysts resembling hyperplastic synovium can develop in the connective tissue of rats following injection of air. Subsequently, a synovium-like membrane was described after orthopedic

surgery[9,10]. Moreover, in some cases, the orthopedic surgeon intentionally created a structure resembling the site of a future tendon graft for enhancing lubrication of the grafted tendon[11].

On the other hand, CMSC, which was first described by Gonzalez et al.[5] in 1987, is rare and unfamiliar to most dermatologists. Although the etiology is not proven, a history of preceding surgery or trauma was present in most cases[1-7], and some cases occurred in patients with basal cell carcinoma[12], Ehler-Danlos syndrome[13] and rheumatoid arthritis[14]. The duration between the time of trauma and the onset time of CMSC ranges from a few weeks to a few years. There appears to be no site predilection or sex predominance; it can arise at any age. Cutaneous manifestations included a solitary tender erythematous nodule related to a scar in most cases. However, multiple or skin-colored lesions have also been reported. CMSC should be differentiated from implantational epidermal cyst, suture granuloma and other cutaneous cysts. Among these, implantational epidermal cyst, which arises after local trauma or surgery, can be very similar to CMSC in its clinical manifestation. However, implantational epidermal cyst is a true cyst which has a true epithelial lining, whereas, CMSC is not a true cyst and lacks a true epithelial lining. Histologically, CMSC is characterized by a cyst lined by a membrane resembling hyperplastic synovium. The lining may be hypo- or hypercellular and contain a mixture of epithelioid, fibroblastic, mononuclear inflammatory and multinucleated giant cells. The cysts have variable degrees of villous projections toward the center of the cavity. The surrounding dermis or subcutaneous tissue may show reactive change such as fibrosis. The epidermis may have a fistula tract connected to the cyst. Immunohistochemical studies indicate that the lining is strongly positive for vimentin but negative for keratin, S-100 and CEA[2,7,14]. However, an immunohistochemical study is not mandatory for the diagnosis of CMSC because it CMSC can be diagnosed using histopathologic findings.

Although the embryological origin of synoviocytes of a normal synovium is controversial, it is hypothesized that epithelioid cells are derived from histiocytes of bone marrow, whereas fibroblasts are from local mesenchymal cells[15]. Furthermore, a normal synovium is embryologically made from mechanical disruption of connective tissues which occurs when the embryo starts to move in vivo[16]. Hence, it is possible that the synovium-like membrane of a CMSC is made from disruption of connective tissue by way of air or oil injection, hyaluronic acid filler injection, orthopedic prosthesis, breast implant, surgery and repetitive trauma such as constant pressure[1-7]. There is no reported case of CMSC in association with herniation of the synovium nor with implantation of epidermal elements.

In this case, there was no history of preceding surgery, accidental trauma or injection of a foreign body at the site of the CMSC. However, the patient said that he had repetitively stabbed and squeezed the existing erythematous papular lesion thought to be acne before the onset of CMSC. Consequently, repetitive manipulation is the most suspected etiology for the development of CMSC in this case.

In conclusion, CMSC is rarely reported but can arise after surgery or trauma. Thus, CMSC should be suspected when a cystic lesion develops on the site of a previous surgery or trauma.

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