Oral and Maxillofacial Surgery Cases 5 (2019) 100104

Contents lists available at ScienceDirect

Oral and Maxillofacial Surgery Cases

journal homepage: www.oralandmaxillofacialsurgerycases.com

A case of Sweet's syndrome secondary to removal of infected mandibular titanium mesh and plate

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ARTICLE INFO

Keywords:
Sweet's syndrome
Titanium plate
Titanium mesh
Infection
Plate removal
Erythema

ABSTRACT

A 67-year-old woman, who had a history of an open surgery for mandible and right knee joint fractures in a traffic accident 28 years ago, visited our hospital due to the swelling of lower jaw. Under the diagnosis of titanium mesh and plate infection, the mesh and the plate removal with debridement was performed. After operation, she realized mild erythemas on her trunk and limbs skin. On the fourth day after operation, she had a fever, increased neutrophils and the elevation of C-reactive protein (CRP). Initially, post-operative infection was suspected and an antibiotic was administered, while it was ineffective. Accordingly, the patient was referred to the Department of Internal Medicine, and subsequently to dermatology specialist for the further diagnosis of skin erythema and blood test abnormalities. Since the dermatology specialist suggested a possible clinical diagnosis as Sweet's syndrome, skin and mucosal biopsy was performed, which confirmed the diagnosis. As far as we could search, this is the first report of Sweet's syndrome followed by the surgery in oral and maxillofacial region. Since this case developed after surgery, it was difficult to distinguish it from postoperative infection or drug allergy.

1. Introduction

Sweet's syndrome (acute febrile neutrophilic dermatosis) was first reported by Sweet in 1964 and is characterized by 4 cardinal features: 1) fever, 2) a relative increase of neutrophils in the peripheral blood, 3) skin lesions such as tender erythematous plaques, nodules, vesicles, and pustules on the face and extremities, and 4) a dense dermal infiltrate with mature neutrophils seen histologically [1,2]. Since fever and neutrophilia are not consistently present, the diagnosis is based on the two constant features, i.e. typical eruption and characteristic histologic features [3].

Although the pathogenesis of Sweet's syndrome is not well understood, dysregulation of immune function or an immune-mediated hypersensitivity to an eliciting bacterial, viral, or tumor antigen are considered as possible triggers [3]. Although the occurrence of Sweet's syndrome is not very uncommon, the cases relating to oral maxillofacial diseases are extremely rare and, as far as we could

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https://doi.org/10.1016/j.omsc.2019.100104
Received 5 January 2019; Received in revised form 1 April 2019; Accepted 6 May 2019
Available online 10 May 2019

search, there is no previously reported case associating to the titanium plate or mesh infection or their removal in maxillofacial region [2]. (166 words).

2. Report of case

A 67 years-old woman visited Matsumoto Dental University Hospital with a chief complaint of the swelling of lower jaw. She had a history of the fracture of mandible and the right knee joint due to a traffic accident 28 years ago and had an open surgery to reconstruct the mandible with a titanium mesh, plate and wires at an emergency hospital. At the first visit to our hospital, her body temperature was 36.8 °C, and her lower jaw showed swelling with redness and a percutaneous fistula was observed, while bone resorption was not evident from panoramic radiograph (Fig. 1). A diagnosis of titanium mesh and plate infection was made and an operation of mesh and plate removal was planned. An incision was made at the skin under chin including the fistula. After elevation of skin-periosteal flap, the titanium mesh and plate were removed. The wires were also removed except for the parts varied inside the mandible. Subsequently, the granulation tissue around the titanium mesh was removed and the incision was closed.

From the evening on the day of surgery, mild erythema was noted at her right hand and the flexion side of right elbow. However, the erythema showed improvement next day, so that no specific treatment was applied. On the third day after the surgery, she complained joint pain on her back and waist, so that a poultice was prescribed. On the fourth day after the surgery, she developed fever (39 °C) and sore throat. Hematologic examination showed a slightly high white blood cell (WBC) count (8500/mm^3) with 87.6% neutrophils, and elevated CRP (6.54 mg/dl). Although the inflammation of the operated region was not evident, postoperative infection could not be excluded and the antibiotics were changed from CEPN-PI (300mg/day) to IPM (1g/day, div). However, after two days, the medication was still ineffective and the fever and sore throat were continued. At this time point, significant erythema recurred accompanying with oppressive pain on her upper and lower limbs on both sides, right side of back and right upper eyelid (Fig. 2a and b). No culture was performed before surgery. On the fifth day after surgery, the bacteriological examination was carried out for the exudate from the chin, but neither aerobic/anaerobic bacteria nor fungi was detected.

Since antibiotics were not effective and the development of erythema was suspected as an allergic reaction, we consulted the Department of Internal Medicine at 6 days after surgery. The primary diagnosis was drug allergy, so that the medication was changed to acetaminophen, CAZ (1g/day, div) and prednisolone (30 mg) was also prescribed. Further examination by a dermatology specialist was also recommended from the physician for skin erythema. Accordingly, we consulted dermatology specialist next day (seven days after surgery) for the skin erythema, and the diagnosis of Sweet's syndrome was suggested, which was confirmed by skin and oral mucosa biopsy. Histologically, edema beneath the epidermis and diffuse infiltration of neutrophils in the dermis were observed (Fig. 2c and d).

After the administration of prednisolone, the skin erythemas were significantly improved and fever, WBC count and CRP level returned to normal range. The prednisolone was then tapered by 5 mg every 5 days and was discontinued by 35 days. The patient discharged on the fifteenth postoperative day. Although the patient was free from symptoms after discharge, it was informed from a dermatologist that her skin erythemas were relapsed at about 4 months after discharge. (569 words).

3. Discussion

Although the pathogenesis and underlying-mechanism of Sweet's syndrome is unknown, Sweet considers the fundamental feature of this disease might be a hypersensitivity reaction to infection because there are cases related to various precede infectious disease [1]. Furthermore, the eruption is sterile and responds to steroid. In the second report, he described that the histology of this disease is similar to that in arthus type reaction, since vasculitis was not evident and the reaction might be stopped at the stage of polymorphonuclear leukocyte infiltration [4]. In the present case, the patient had a continuous contact with bacteria due to plate infection and it may exaggerated at the time of operation by the surgical manipulation to the infected tissues.

Delayed infection cases after surgical fracture treatment have been reported and the possible causes were considered as the presence of foreign body (resorbable plates and absorbable suture materials) and delayed bone union [5]. However, the onset of infection in those cases were about 13 months after surgery and 18 months at the longest, which were not comparable to 28 years in the present case [6]. Although the cause of infection in this case is unknown, long-term presence of the titanium mesh and plate might

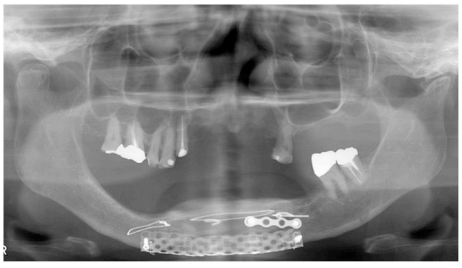

Fig. 1. Panoramic radiograph at first visit. Titanium mesh plate, titanium plate, and multiple wires were observed in the mid-mandible, but no obvious bone resorption was noted.

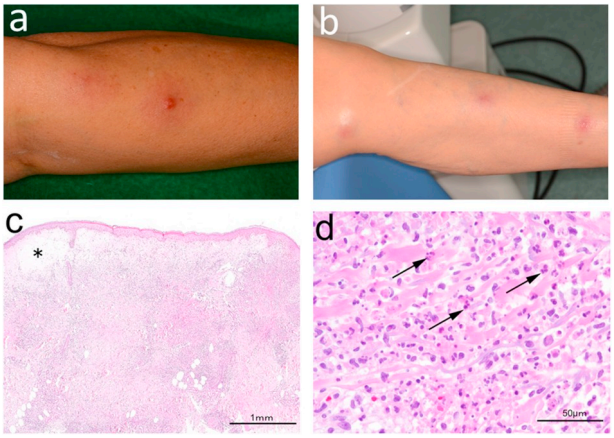

Fig. 2. Macroscopic and microscopic features of the skin erythemas. Erythema of the left forearm with pustular (a), and coin size painful erythemas of the left lower limb (b). Edema beneath the epidermis (*)and diffuse inflammatory cell infiltration in the dermis (HE staining, 20×) (c). Neutrophils infiltration in the connective tissue (Arrow) (HE staining, 400×) (d).

increase the risk of infection via hematogenous rout or through mucosal injury/ulceration.

There are some reports that Sweet's syndrome has developed after surgery such as tibial osteotomy [7]. However, the relationship between surgery and Sweet's syndrome is unclear and there are more cases accompanying with the preceding infections. Importantly, some of the cases were related to skin bacterial flora [8]. Since the possible pathogenesis of Sweet's syndrome relating to immunological reaction against bacterial components, it might be reasonable to consider that the exposure to bacterium around infected titanium mesh and plate may cause the reaction.

On the other hand, the occurrence of this disease associated with dental treatment or oral surgery has limited [2]. However, there are many reports that it is an initial symptom of stomatitis and pharyngitis, the dentist and the oral surgeon should also aware of this disease. Since this case developed after the operation, it was difficult to differentiate it from postoperative infection or drug allergy, and we were struggling with diagnosis and treatment. As described in the text, we could not initially rule out the potential infection, since the patient suffered from high fever and the abnormal laboratory values such as relatively high white blood cell count (8500/mm3) with 87.6% neutrophils and elevated CRP (6.54 mg/dl). However, the lack of response to antibiotic treatments as well as the presence of skin erythema lead us to the alternative diagnosis. The differential diagnoses of the case include Erythema multiforme and Behçet's disease. As for the treatment, a steroid and a potassium iodide are effective, and it should be noted that recurrence occur one third of the cases [9].

4. Conclusion

We reported a case of Sweet's syndrome, which occurred secondary to the titanium mesh and plate infection and their removal, which was a difficult case to diagnose.

Funding source

None.

Acknowledgement

This work was supported in part by a Grant-in-Aid for Scientific Research (JSPS KAKENHI Grant Numbers JP16H05546) and a grant from Matsumoto Dental University. The authors wish to thank Dr. Tomohiro Motoyama, Dr. Naomi Matsumura, Dr. Eri Uchikawa, for their contribution to the treatment of this case and Professor Masahiro Hasegawa for the pathological diagnosis.

References

[1] Sweet RD. An acute febrile neutrophilic dermatosis. Br J Dermatol 1964;76:349–56.
[2] Notani K, Kobayashi S, Kondoh K, Shindoh M, Ferguson MM, Fukuda H. A case of Sweet's syndrome (acute febrile neutrophilic dermatosis) with palatal ulceration. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2000;89:477–9.
[3] Vashisht P, Holmes MPH. Sweet syndrome. StatPearls [Internet]. Treasure Island (FL). StatPearls Publishing; 2018. 2017 Nov 28.
[4] Sweet RD. Further observations on acute febrile neutrophilic dermatosis. Br J Dermatol 1968;80:800–5.
[5] Snäll J, Apajalahti S, Suominen AL, Törnwall J, Thorén H. Influence of perioperative dexamethasone on delayed union in mandibular fractures: a clinical and radiological study. Med Oral Patol Oral Cir Bucal 2015 Sep 1;20(5):e621–6.
[6] Lee HR, Shin HK, Lee DL, Jung GY. Delayed-onset methicillin-resistant Staphylococcus aureus infection at 18 Months after absorbable plate fixation for zygomaticomaxillary complex fracture. Arch Craniofac Surg 2016 Dec;17(4):229–32. https://doi.org/10.7181/acfs.2016.17.4.229. Epub 2016 Dec 23.

[7] Minocha R, Sebaratnam DF, Choi JYJ. Sweet's syndrome following surgery: cutaneous trauma as a possible aetiological co-factor in neutrophilic dermatoses. Australas J Dermatol 2015;56:e74–6.
[8] Abramovits W1, Stevenson LC. Sweet's syndrome associated with Staphylococcus aureus. Int J Dermatol 2004;43:938–41.
[9] Pai S, Rytina E, Sterling J, Karas JA, Aliyu SH. Campylobacter gastroenteritis associated with Sweet's syndrome. J Med Microbiol 2012;61(Pt 10):1473–5.