Clinical Case Report

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Tetanus presenting primarily as periocular discomfort in a patient who underwent surgery for sleep apnea

A case report and brief literature review

Donghwi Park, MD[a], Jung Soo Kim, MD, PhD[b], Sung Jae Heo, MD[c,*]

Abstract

Rationale: Tetanus is caused by a neurotoxin (tetanospasmin) secreted by a spore forming gram-positive, anaerobic rod-shaped motile bacillus, *Clostridium tetani*. The most common symptoms of tetanus are trismus (100%), dysphagia (70.5%), dysarthria (35.2%), and neck stiffness (29.4%). Respiratory failure, laryngeal spasm, seizure, chest pain, nausea/vomiting, opisthotonus, back pain, and rigid abdominal wall can also be observed during progression of the disease. However, there has been no report of periocular discomfort as an initial manifestation after endoscopic sleep surgery in a patient with tetanus. Here, we report a patient who underwent endoscopic sleep surgery with a concurrent diagnosis of tetanus infection presenting with atypical periocular discomfort as the initial symptom.

Patient concerns: A 63-year-old man complaining of sleep apnea, snoring, and daytime sleepiness visited our department. He subsequently underwent sleep surgery (anterior pharyngoplasty with tonsillectomy, septoplasty, microdebrider-assisted inferior turbinoplasty, and an endoscopic sinus surgery) for the treatment of his newly diagnosed obstructive sleep apnea. After 3 weeks of surgery, he visited the outpatient clinic of our department with right side periocular discomfort.

Diagnoses: Four days after presenting with periocular discomfort, he was diagnosed with tetanus by presenting trismus, jaw pain, dysphagia, and ptosis at an emergency department of a different hospital.

Interventions: Tetanus immunoglobulin and antibiotics were administered.

Outcomes: His symptoms then resolved after a month without sequelae.

Lessons: Although periocular discomfort is atypical and is not uncommon after nasal and oral surgeries, care should be taken when patients present with periocular pain because it could be a rare initial symptom of tetanus.

Abbreviation: CT = computed tomography.

Keywords: periocular pain, sleep apnea, surgery, tetanospasmin, tetanus

1. Introduction

Tetanus is caused by a neurotoxin (tetanospasmin) secreted by *Clostridium tetani*, which is a spore forming gram-positive, anaerobic rod-shaped motile bacillus.[1] Contaminated open trauma is the most common cause of tetanus, while burns, animal bites, and puncture injury from rusted nails are important predisposing factors for tetanus infection.[2] The most common symptoms of tetanus are trismus (100%), dysphagia (70.5%), dysarthria (35.2%), and neck stiffness (29.4%).[3] Respiratory failure, laryngeal spasm, seizure, chest pain, nausea/vomiting, opisthotonus, back pain, and rigid abdominal wall can also be observed during progression of the disease.

Periocular discomfort is not a typical symptom of tetanus and can be observed in multiple other conditions, for example, after nasal surgery or endoscopic sinus surgery (periocular discomfort due to mucus retention caused by mucosal swelling and edema), and oral surgery (periocular discomfort due to electrolyte imbalance caused by poor oral intake). However, because periocular discomfort can be a rare initial symptom of tetanus, care should be taken when initiating treatment for patients with periocular pain. Moreover, there has been no report of periocular pain as an initial manifestation after sleep surgery in a

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[a] Department of Rehabilitation Medicine, Daegu Fatima Hospital, [b] Department of Otorhinolaryngology-Head and Neck Surgery, School of Medicine, Kyungpook National University, Kyungpook National University Hospital, [c] Department of Otorhinolaryngology-Head and Neck Surgery, School of Medicine, Kyungpook National University, Kyungpook National University Chilgok Hospital, Daegu, South Korea.

[*] Correspondence: Sung Jae Heo, Department of Otorhinolaryngology-Head and Neck Surgery, Kyungpook National University Chilgok Hospital, 807, Hoguk-ro, Buk-gu, Daegu 41404, Korea (e-mail: blueskyhsj@hanmail.net).

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patient with tetanus. Here, we report a patient who, after undergoing sleep surgery (nasal and oral surgery), presented with initial periocular discomfort and was later diagnosed with tetanus. Informed written consent was obtained from the patient for publication of this case report and accompanying images. This case was approved by ethics committee of our hospital (Institutional Review Board of Kyungpook National University Chilgok Hospital).

2. Case report

A 63-year-old man complaining of sleep apnea, snoring, and daytime sleepiness visited our department. Nasal endoscopy showed septal deviation, inferior turbinate hypertrophy, nasal polyp, tonsillar enlargement of Friedman grade 2, and redundant palatal tissue, and a big tongue of modified Mallampati grade 3 was observed on oral examination. Polysomnography confirmed the diagnosis of severe obstructive sleep apnea (apnea-hypopnea index of 41.9), and drug-induced sleep endoscopy showed complete obstruction of the velum and the lateral wall of the oropharynx. Since the patient wanted to undergo surgery, sleep surgery (anterior pharyngoplasty with tonsillectomy, septoplasty, microdebrider-assisted inferior turbinoplasty, and endoscopic sinus surgery) was performed for the treatment of obstructive sleep apnea.

He was discharged on the second day after surgery without any complaints. Surgical wound was unremarkable on examination in the outpatient clinic 1 week after operation. Discomfort and pain due to the surgery were alleviated after 3 weeks of surgical intervention. His surgical wound was still unremarkable on examination 3 weeks postoperatively (Fig. 1); then, the patient visited our outpatient clinic with right side periocular discomfort. His symptom was mild and no prominent abnormal findings were observed on physical examination. Suspecting sinus ostium obstruction, we prescribed antibiotics and steroid and encouraged a balanced diet to address the possibility of electrolyte imbalance caused by his surgery 3 weeks earlier. He underwent ophthalmologic examination, but no abnormal findings were observed, and there was no improvement in medication of antibiotics and steroid.

Four days after presenting with periocular discomfort, he developed trismus, jaw pain, dysphagia, and ptosis, and he presented at an emergency department in a different hospital. The patient had an unsure history of tetanus immunization and denied any previous injury or traumas, except for the sleep surgery he recently underwent. After the diagnosis of tetanus was confirmed, with a positive spatula sign, tetanus immunoglobulin and antibiotics were administered. His symptoms resolved after a month without sequelae.

3. Discussion

Tetanus is not an infection, but rather a toxemia with a characteristic nervous system tropism.[4]*C tetani*, the source of the tetanospasmin toxin, is found ubiquitously in the soil and in the excrement of some farm animals and humans. The spores of *C tetani* usually gain entry into human tissues through traumatic or inflammatory/infected wounds.[5] Subsequently, if they encounter an anaerobic environment (more common in contaminated open trauma), they germinate into the vegetative form of the organism, the toxin-producing bacilli.[4,6,7] Thus, the clinical form of a particular case—that is, either general or local tetanus—is determined based on various factors, including the age of patients, immunization status, and, importantly, site of entry.[8] Although tetanus is an uncommon disease in the developed world due to vaccination coverage, it is still a concern due to high mortality rates of up to 90%.[9,10] In South Korea, however, the number of reported cases of tetanus has increased gradually over the past 20 years (Table 1). In 2001, there were 8 cases reported in South Korea, which increased to about 30 in 2018 (Table 1).

Tetanospasmin is the toxin responsible for the clinical manifestations of tetanus.[5] It works through central interruption of glycinergic neurons of inhibitory interneurons and the inhibitory gamma-amino butyric acid (GABA)-ergic at the level of the spinal cord.[5] The goals of tetanus treatment are to stop the production of toxin at the site of infection by eliminating the

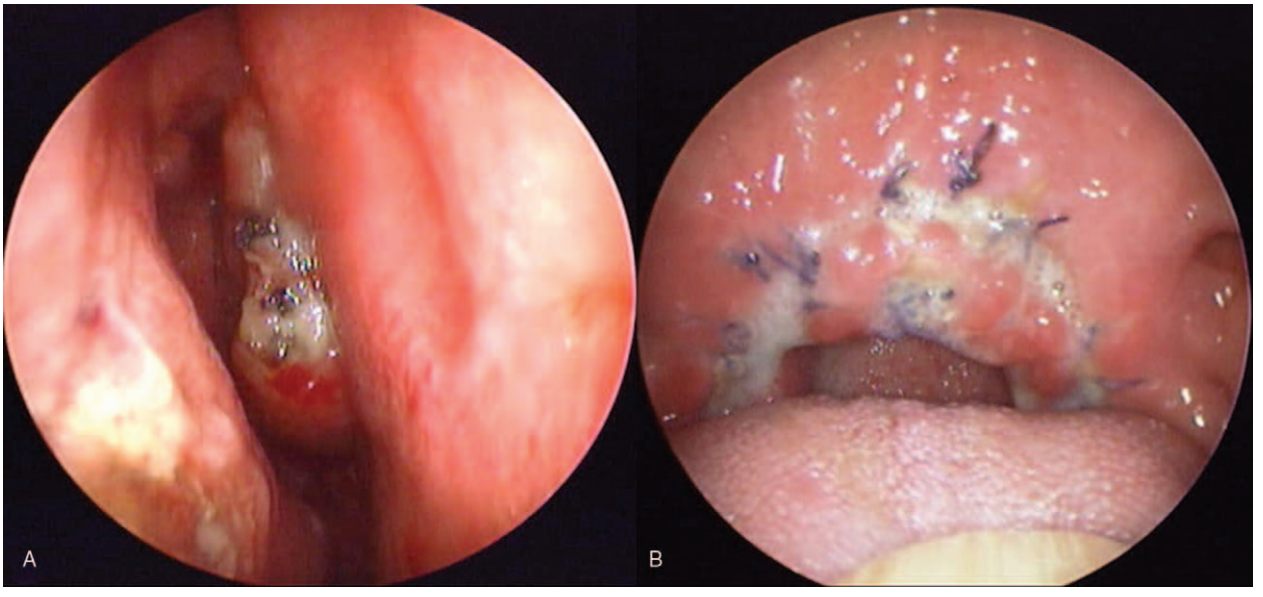

Figure 1. Endoscopic findings of nasal cavity (A) and oral cavity (B) on postoperative day 21.

Table 1

Incidence and mortality rate of tetanus in South Korea, 2001 to 2018.

Year	Number of cases	Death from tetanus
2001	8	0
2002	4	0
2003	8	0
2004	11	0
2005	11	0
2006	10	0
2007	8	0
2008	16	0
2009	17	0
2010	14	0
2011	19	2
2012	17	1
2013	22	1
2014	23	0
2015	22	0
2016	24	2
2017	34	0
2018	30	0

2001–2018 data are from the infectious disease surveillance systems of the Korea Centers for Disease Control and Prevention (KCDC). (http://www.cdc.go.kr/npt/biz/npp/list/bass/bassDissStatsMain.do).

Table 2

Incidence according to age in South Korea, 2001 to 2018.

	Tetanus incidence (%)
Total	298
0–9 years	2 (0.67%)
10–19 years	3 (1.00%)
20–29 years	10 (3.37%)
30–39 years	11 (3.69%)
40–49 years	25 (8.39%)
50–59 years	59 (19.80%)
60–69 years	82 (27.52%)
More than 70 years	106 (35.57%)

2001–2018 data are from the infectious disease surveillance systems of the Korea Centers for Disease Control and Prevention (KCDC). (http://www.cdc.go.kr/npt/biz/npp/list/bass/bassDissStatsMain.do).

organism through appropriate antibiotic use, for example, metronidazole; wound debridement and neutralization of tetanus toxin with human or equine tetanus immunoglobulin; and management of muscle spasms with diazepam and treatment of autonomic dysfunction and other complications that develop during the course of disease.[1]

In our report, the cause of tetanus development is obscure. Since surgical conditions are always sterile, postoperative tetanus rarely occurs, especially in the developed countries. For example, it develops at the rate of about 1 case in every 7 years in Canada and 1 case per year in the United States.[10] The incidence of postoperative tetanus is extremely rare, and most reported cases occurred after abdominal or gynecological surgery with the intestinal tract as the source of infection in both types of surgeries.[10] To date, no case of postoperative tetanus has been reported after sleep surgery. Therefore, nosocomial infection is unlikely in our case. In addition, epidemiologic evidence suggests contrary to possible nosocomial infection.

In addition, the patient's symptoms occurred a month after surgery, while the incubation period of tetanus is usually 3–21 days (mean 14 days).[11] Especially, postoperative tetanus develops quickly, usually within 24 hours.[10] Therefore, the possibility of *C tetani* infection resulting from the surgical intervention in our case is very low. Therefore, the cause of tetanus in this patient might be an unrecognized small injury even though the patient denied any trauma history. Since *C tetani* is widely distributed in the environment, tetanus can be acquired through injuries like a scratch, which is generally not recognized. Similar to our case, Ryu et al[12] and Yanagi et al[13] reported cases of tetanus that did not have any history of injury. Lee et al[14] reported that the most common causes of tetanus infection are open wound injury (48.2%), sting by foreign body (29.6%), unknown (11.1%), and cesarean operation (5.6%). Unknown origin of infection was the third most common cause of tetanus.

Kim et al[11] reported a case of tetanus that developed during adjuvant chemotherapy of breast cancer. Their explanation was that the decreased immunity caused by chemotherapy might have contributed to the development of tetanus. Therefore, our case might be related to an unrecognized injury in combination with the transient immunosuppressed state caused by oral surgery (which commonly induces insufficient feeding). In addition, epidemiologic evidence shows that, from 2001to 2018 in South Korea, the incidence of tetanus increased with age (Table 2). This reported increased incidence in the elderly supports the hypothesis that older people are more likely to acquire tetanus due to their inherent immunosuppressed state (Tables 1 and 2). This could also be a contributory factor to the case we have reported here.

The symptom of tetanus is characterized by painful muscle spasm, increased muscle tone (rigidity) and, in severe cases, cardiovascular instability caused by autonomic dysfunction. The presenting symptoms of tetanus are usually trismus, neck stiffness, convulsion, dysphagia, dysarthria, opisthotonos, and abdominal rigidity.[14] The patient in this case presented with discomfort in the periocular area as an initial symptom. The increased muscle tension commonly observed in tetanus might explain the patient's symptom of periocular discomfort. However, this symptom is atypical and is not uncommon after nasal surgery and oral surgery. Therefore, although this is the first report of a patient with tetanus presenting with periocular pain as an initial manifestation after sleep surgery, care should be taken when patients present with periocular pain after nasal and oral surgery because periocular discomfort can be a rare initial symptom of tetanus.

Author contributions

Conceptualization: Sung Jae Heo.
Data curation: Donghwi Park, Jung Soo Kim.
Formal analysis: Donghwi Park, Jung Soo Kim, Sung Jae Heo.
Methodology: Donghwi Park, Jung Soo Kim, Sung Jae Heo.
Writing – Original Draft: Donghwi Park.
Writing – Review & Editing: Sung Jae Heo.

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