INTRODUCTION

Many patients present with multiple symptoms, and some are medically unexplainable. These can occur after provision of medical care such as vaccinations and can be mistaken as "side effects." We here present a case of bodily distress syndrome, or bodily distress syndrome (BDS), after receiving tetanus toxoid injections and discuss the issues involving diagnosis and treatment of the illness.

CASE REPORT

A 48-year-old Japanese woman was referred to our clinic for various symptoms and the fear of side effects of vaccines. She had a dog bite on her face 2 months prior to the visit to us. At a hospital she visited, she received tetanus toxoid twice. Shortly after the second injection, she started to develop chill, back pain, and neck and shoulders stiffness. She also suffered polydipsia, shortness of breath, and chest discomfort, and she visited the hospital again. There she was reportedly told by an emergency physician that she had "abnormal electrocardiogram that could be fatal." She also developed pins-and-needles sensations on four limbs. The pain radiated from shoulders to her elbows and from lateral aspects of her hips to her knees. Because of her back pain, she feared zoster and visited a dermatologist, but she was told she did not have this. She also suffered insomnia, which she ascribed to pain and anxiety, but she never woke up because of pain after she fell asleep. She also developed headache, abnormal genital bleeding, and frequent watery diarrhea. She visited a gynecologist, and her hormonal workup was normal. She visited a neurologist for her limb pain and was prescribed vitamin B 12 tablet without specific explanation. She also visited a psychosomatic medicine specialist for her insomnia and was prescribed gabapentin and alprazolam. She also visited a rheumatologist due to fear of Sjogren syndrome because of her thirst, but the laboratory tests did not suggest any kind of collagen vascular diseases. The rheumatologist suspected potential side effects of tetanus toxoid, and she was referred to our clinic.

On physical examination, she appeared anxious and was in agony. Her vital signs were normal. Head and neck examinations were normal including thyroid palpation. Her eyes and mouth appeared appropriately wet. Her trapezius muscles were stiff, and her symptoms improved by massaging them. She did not have tender points suggestive of fibromyalgia, nor any other part of her body.
She also did not have any rash, lymphadenopathy, or arthritis. The rest of her physical examination, including thorough neurological examination, was entirely normal.

She received all recommended immunization during her childhood. We called the emergency physician for the details of her cardiac workup. It turned out that her electrocardiogram merely showed J-point elevations in the ST segments, and they were wrongly interpreted as “possible Brugada pattern.”

Because of the broadness of her symptoms’ categories, which were medically unexplainable, painful sensation on all four limbs but only proximally led us to consider that she was suffering from a functional syndrome secondary to tetanus toxoid injection. With symptoms such as palpitation, dry mouth, diarrhea, pains in arms and legs, and disability she suffered from these symptoms, while excluding other relevant differential diagnoses, fulfilled the diagnostic criteria of bodily distress syndrome. We told her what we considered, for which she expressed her delight with relief. By further history taking, she disclosed that she received another tetanus toxoid 2 years ago when she had a dog bite. She thought she received too many tetanus toxoids, and she wondered whether these could have been toxic to her. The comment by the rheumatologist and referral to us, the infectious diseases department, and “abnormal ECG” augmented her fear.

We prescribed amitriptyline 10 mg daily, Kakkonto, and Keishikaryukotsuboreito 1 packet 3 times a day. Her symptoms disappeared shortly, except for occasional back and left upper arm pain, and her medications were continued.

3 | DISCUSSION

Bodily distress syndrome (BDS) was recently introduced as a diagnostic category, and it is considered as one of functional disorders. BDS is characterized by multiple unexplainable symptoms (Table 1). Many terms have been used to describe symptoms without diagnosis, such as medically unexplained symptoms (MUS), functional symptoms, functional somatic syndrome, central sensitivity syndromes, or somatoform disorders. Among these functional disorders, the presence of multiple system symptoms, often pertaining to multiple organ systems, constitutes BDS.

We consider that the current patient suffered from BDS, triggered by her tetanus toxoid injection, and exacerbated by her fear of side effects. Tetanus toxoid is considered generally safe but associated with local symptoms, such as pain and swelling at injection site, or generalized symptoms, such as fever. However, these symptoms are mostly self-limited and typically last only for several days. Tetanus toxoid could be associated with other inflammatory disorders such as rheumatoid arthritis or systemic lupus erythematosus, but lack of physical findings of joints and skin and lack of positive serology tests indicate that the patient did not suffer these complications.

Treatment of functional disorders such as BDS is not well-established, but various treatments such as psychological therapies and pharmacological therapies were proposed. We tried amitriptyline, Kakkonto, and Keishikaryukotsuboreito with apparent success. Amitriptyline has been used for various pain syndromes including fibromyalgia. Kakkonto can be used for stiffness shoulders. Keishikaryukotsuboreito has been used for anxiety disorders, functional palpitation, dizziness, and various other symptoms associated with anxiety. However, early diagnosis and reassurance also could have positive impact on her symptoms since she appeared to have felt better immediately after providing our assessment. On the contrary, wrongly implying potential side effects of vaccines without sound clinical reasoning or diagnostic confirmation could have negative effects on treatment of BDS.

Many suffer from illness after vaccinations, and some of them could be identified as side effects of vaccines. On the other hand, several hypotheses of vaccine-related illness, such as autism by measles-mumps-rubella vaccine, have been denied by large scale comparative studies. Human papilloma virus vaccines were also alleged to be associated with multisymptoms such as pain and insomnia in Japan, but recent comparative study failed to find association between symptoms and vaccinations.

We are not to allege, based on our single case, that all symptoms after any vaccinations are functional. But this single case demonstrates that some symptoms after vaccinations could be functional, reasoned by falsifiability of Karl Popper, and ignoring this possibility could rather worsen the suffering of the recipient of vaccines. Literatures on functional disorders after vaccinations are scarce, although we see such cases not that infrequently. It could partly

| TABLE 1 | Diagnostic criteria for bodily distress syndrome or bodily distress syndrome (BDS) |
|---|---|
| 1) ≥3 symptoms from at least one of the following groups: |
| • Cardiopulmonary/autonomic arousal: |
| Palpitations/heart pounding, precordial discomfort, breathlessness without exertion, hyperventilation, hot or cold sweats, dry mouth |
| • Gastrointestinal arousal: |
| Abdominal pains, frequent loose bowel movements, feeling bloated/full of gas/distended, regurgitations, diarrhea, nausea, burning sensation in chest or epigastrium |
| • Musculoskeletal tension: |
| Pains in arms or legs, muscular aches or pains, pains in the joints, feelings of paresis or localized weakness, backache, pain moving from one place to another, unpleasant numbness, or tingling sensations |
| • General symptoms: |
| Concentration difficulties, impairment of memory, excessive fatigue, headache, dizziness. |
| 2) The patient has been disabled by the symptoms (ie, daily living is affected) |
| 3) Relevant differential diagnoses have been ruled out |
| • Severity: |
| Single-organ BDS (mild-moderate) involves one or two of the symptom groups |
| Multi-organ BDS (severe) involves three or four of the symptom groups |
be due to the unawareness of the illness entities among physicians or refusal to consent them to be reported by the patients or their families.

In conclusion, we report a case of BDS after receiving tetanus toxoid injection. Our case illustrates the importance of early accurate diagnosis and reassurance to the patient, to help avoiding lengthy continuation of functional disorder, which can be very painful to the patients.

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CONFLICT OF INTEREST

The authors have stated explicitly that there are no conflicts of interest in connection with this article.

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