Trismus originating from rare fungal myositis in pterygoid muscles: A case report

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Abstract

BACKGROUND

Trismus is a common problem with various causes. Any abnormal conditions of relevant anatomic structures that disturb the free movement of the jaw might provoke trismus. Trismus has a detrimental effect on the quality of life. The outcome of this abnormality is critically dependent on timely diagnosis and treatment, and it is difficult to identify the true origin in some cases. We present a rare case of trismus due to fungal myositis in the pterygoid muscle, excluding any other possible pathogenesis.

CASE SUMMARY

The patient presented with a 2-mo history of restricted mouth opening. Computed tomography showed obvious enlargement of the left pterygoid muscles. Furthermore, the patient had trismus without obvious predisposing causes. The primary diagnosis was pterygoid myosarcoma. Consequently, lesionectomy of the left pterygoid muscle was performed. Intraoperative frozen biopsy implied the possibility of an uncommon infection. Postoperative pathologic examination confirmed myositis and necrosis in the pterygoid muscle. Fungi were detected in both muscle tissue and surrounding necrotic tissue. The patient recovered well with antifungal therapy and mouth opening exercises. The rarity of fungal myositis may be responsible for the misdiagnosis. Although the origin of pathogenic fungi is still unknown, we believe that both hematogenous spread and local invasion could be the most likely sources. To the best of our knowledge, this...
is the first case in the literature that reported fungal myositis in pterygoid muscles as the only reason that results in trismus.

CONCLUSION
Surgeons should remain vigilant to the possibility of trismus originating from fungal myositis.

Key Words: Trismus; Fungal myositis; Infection; Immunodeficiency; Pterygoid muscle; Case report

Core Tip: Trismus has a detrimental effect on the quality of life. Early diagnosis and treatment have the potential to minimize the consequences of this condition. However, it is not always easy to identify the true origin in some cases. We report the first case in the literature that fungal myositis in pterygoid muscles is the only reason for trismus. We initially misdiagnosed this case of fungal origin because of its rarity. Surgeons should consider the possibility of fungal myositis in trismus diagnosis.

INTRODUCTION
Trismus refers to a severely restricted mouth opening of different etiologies. In most cases, trismus is the result of sustained contraction of the masticatory muscles. This abnormality has a negative impact on the quality of life. Early diagnosis and treatment have the potential to minimize the consequences of this condition. However, there are some special cases with rare origins that are difficult to identify.

Herein, we report the first case of trismus originating from fungal myositis in the pterygoid muscle.

CASE PRESENTATION

Chief complaints
A 58-year-old Chinese female presented with a 2-mo history of restricted mouth opening that gradually aggravated with pain 2 wk before admission. The patient had difficulty speaking and chewing.

History of present illness
The patient had trismus without obvious predisposing causes. The symptoms started 2 mo prior, which had gradually exacerbated. The patient did not receive any treatment before her first clinic visit.

History of past illness
The patient was healthy. She denied any history of immunodeficiency throughout the illness.

Personal and family history
No personal or family history of fungal myositis or trismus exists.

Physical examination
On admission, her mouth opening was less than 10 mm. Her face was symmetrical. She felt pain when palpated in the left preauricular region. Her vital signs were within
normal range.

**Laboratory examinations**
Pathoglycemia (fiber Bragg grating = 10.75 mmol/L) and hyperglycosuria (UGLU = 56 mmol/L) were implicated in routine blood tests. Her white blood cell count was normal. Ketone body was detected in her urine (U-Ket = 15 mmol/L).

**Imaging examinations**
Computed tomography (CT) revealed obvious enlargement of the left pterygoid muscles. The boundary between the lateral and medial pterygoid muscles was obscure (Figure 1). Patchy enhancement was observed in muscles after intravenous injection of a contrast agent (Figure 2). Bone destruction and thickened mucous membrane on the maxillary sinus back wall, which was very close to the pterygoid muscle, were also seen on CT.

**Primary diagnosis and initial treatment**
The primary diagnosis of left pterygoid myosarcoma and diabetes was established. Glucose-lowering medications were immediately administered to the patient. Consequently, lesionectomy of the left pterygoid muscles was scheduled under good glucose control. However, the repeated intraoperative frozen biopsy did not validate the original diagnosis and implied the possibility of an uncommon infection.

**FINAL DIAGNOSIS**
Routine postoperative pathologic examination confirmed myositis and necrosis in the pterygoid muscle. Fungi were detected in the pathological sections of both muscle tissue and surrounding necrotic tissue (Figure 3 and 4). In addition, aspergillosis was diagnosed based on morphological analysis. Therefore, the final diagnosis was amended to fungal myositis in the pterygoid muscle with necrosis.

**TREATMENT**
The patient was prescribed antifungal therapy and mouth opening training. Considering that the original lesion had been resected thoroughly, treatment with oral voriconazole (loading dose, 300 mg bid; maintenance dose, 200 mg bid) was instituted. Voriconazole was discontinued after 8 wk of therapy. No adverse reactions were detected during the treatment. The patient was recommended to perform mouth opening exercises 1 wk postoperatively using a T-shaped mouth opener.

**OUTCOME AND FOLLOW-UP**
The interincisor distance of the patient increased to 30 mm at 15 d postoperatively. After 6 mo of follow-up, her mouth opening was stable at 36 mm. She did not complain of pain or trismus.

**DISCUSSION**
The word trismus was originally used only in tetanus as a prolonged masticatory muscle spasm[1]. However, the term is currently used to indicate varying degrees of restricted mouth opening regardless of the etiology. In most of the studies we reviewed, the authors only set criteria for trismus but did not explain why they defined it in that way. In addition, no study has provided justification for its criteria. Some authors defined trismus as a mouth opening less than an appointed number, while others defined it according to a more gradual scale[2-4]. However, most authors agree that a mouth opening of 35 mm or less should be regarded as a trismus.

Trismus has a negative impact on quality of life. It may impair basic oral functions such as chewing, swallowing, and speech. It also detrimentally affects oral hygiene and tumor surveillance[5-7]. Early diagnosis and treatment have the potential to minimize the consequences of trismus[8].
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Figure 1 Obvious enlargement of the left side pterygoid muscles appear on computed tomography scanning. The boundary of the lateral and medial pterygoid muscles is obscure. Bone destruction and thickened mucous membrane on the maxilla sinus back wall appear as well (arrow).

Figure 2 Patchy enhancement is observed in the pterygoid muscles after injection of the contrast agent (arrow).

The identification of the true causes of trismus is complicated. Traditionally, they can be divided into intra-articular or extra-articular, which are often difficult to distinguish. Many causes are related to abnormalities in the masticatory muscles. Malignant diseases in the head and neck area can provoke trismus by infiltration and irritation of the muscles adjacent to the mandibular locomotor structure. Treatment of the malignancy, including surgical resection and radiotherapy, can also lead to trismus by producing muscular fibrosis and muscle contraction[5,9]. Some authors emphasize the impairment of the pterygoid muscles for the development of trismus[10,11]. Although it is widely recognized that pterygoid myositis can give rise to trismus[12], a fungus-originated case is still unexpected.

In general, healthy muscles are resistant to infection[13]. Muscle infection is uncommon, and fungal myositis is even rarer. It is well known that fungal infections are almost totally opportunistic. Fungi turn into pathogens only under the right circumstances[14]. Although fungal myositis is occasionally reported in immunocompetent individuals, most cases involve immunocompromised patients[15-18].
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Figure 3 Pathological section shows diffuse fungi among the muscle cells. The section also shows focal necrosis with inflammatory cell infiltration (periodic acid-Schiff stain, × 400 magnification).

Figure 4 A fluorescence staining section of the surrounding necrotic tissue shows fungal hyphae (fungal fluorescence stain, × 400 magnification).

Advisory Committee on Immunization Practices has identified many possible risk factors for immunodeficient patients. The most common conditions for fungal infection include diabetes, especially cases with ketoacidosis and hematological malignancies with neutropenia. Immune deficiency in acquired immunodeficiency syndrome still plays a controversial role in mycosis generation. Some authors have not considered it a risk factor[19].

The potential routes of fungal myositis are an invasion of the musculature via contiguous sites[20], hematogenous dissemination, and trauma with direct seeding of spores. The use of needles or intravenous catheters as iatrogenic factors has also been reported[21]. In our case, routine blood examination revealed abnormalities in connection with immunocompromise. As fungal myositis is usually recognized as a complication of systemic mycosis[22], blood dissemination may be a reasonable approach in our case.

To our knowledge, there has been a steady increase in the incidence of fungal sinusitis in immunodeficient patients. Currently, fungal sinusitis is divided into two dominant types: Invasive and noninvasive. As its name implies, invasive fungal sinusitis can invade and destroy neighboring tissues.

The diagnosis of invasive fungal sinusitis (IFS) remains difficult. As seen in bacterial or viral sinusitis, early radiologic findings of IFS are nonspecific[16,23]. Sometimes, bone destruction can be seen in the progressive stage. Moreover, acute IFS can disseminate to adjacent structures. However, definitive diagnosis and identification of the species can only be made by fungal culture. In this study, bone destruction of the maxillary sinus back wall was observed. In addition, abnormal mucosal lesions were
also observed. Therefore, we could not exclude local invasion as one possible route, although there was no pathological evidence of fungal infection in the maxillary sinus. However, routine histological analysis verified that fungi, most likely *Aspergillus*, were located in the muscle and necrotic tissue.

For a definite diagnosis of fungal myositis, a timely treatment protocol must be implemented. However, due to its rarity and limited clinical experience, no consensus has been reached regarding the best means of treating it [22,24]. Therefore, the treatment is regularly combined and consists of aggressive surgical debridement and administration of antifungal agents. A distinctive finding during debridement is that the affected tissue did not bleed, presumably because of tissue infarction. Furthermore, therapies to reverse underlying risk factors are recommended for immunocompromised patients, for example, hypodermic injection of granulocyte colony-stimulating factor to restore neutrophil counts.

In addition to these etiological treatments specific to fungal infections, there are some conventional symptomatic treatments for trismus. According to many reports, conservative treatment is effective. Exercise is believed to be an indispensable mainstay for different etiogenic trismus. Tongue spatulas, TheraBite Jaw Motion Rehabilitation System™, and Dynasplint Trismus System have presented promising results in clinical use [5,25]. Other conservative treatments, such as thermal therapy, electrotherapy, and botulinum toxin injection, are also optional, but their potential effects are uncertain [26-28]. Finally, it cannot be denied that quality of life deficits originate from trismus results even in social inhibition and depression. Based on this reality, trismus should be treated in an integral way, including measures to sustain patients’ mental health. Simultaneously, we should always remember that prevention, rather than treatment, is the most important objective.

Regardless of fungal myositis or trismus, the prognosis largely depends on early diagnosis and timely treatment. Many factors are associated with the prognosis. Basic immune status, mental status, uncontrolled diabetes, and mouth opening exercises are considered the most important prognostic factors.

**CONCLUSION**

The rarity of fungal myositis may be responsible for the misdiagnosis of this case. Although a definitive pathological diagnosis was obtained, the origin of the pathogenic fungi remains unconfirmed. Since the patient was also diagnosed with diabetes, which could erode her immunity, we believed that opportunistic fungal infection was possible. Under these circumstances, both a hematogenous spread and local invasion could be their true origins. Therefore, surgeons should remain vigilant in infection was possible. Under these circumstances, both a hematogenous spread and local invasion could be their true origins. Therefore, surgeons should remain vigilant in prevention, rather than treatment, being the most important objective.

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