INTRODUCTION

Masticatory muscle myositis (MMM) is the most common inflammatory myopathy among canines and was extensively investigated. It is restricted to masticatory muscles and limb muscles are typically spared. It is also associated with an autoimmune process involving the formation of auto-antibodies against type 2M muscle fibers in the masticatory muscles, the cause of which is unknown, although some authors proposed the theory of molecular mimicry (Caley et al., 2004; Tresamol et al., 2012; Quiroz-Rothé et al., 2002; Evans et al., 2004; Jennifer et al., 2003).

In humans, MMM has not been extensively documented thus far and is rare. Here, we presented a case of MMM in a 28-year-old woman and focused on histopathologic findings and differential diagnosis.

Case report

A 28-year-old woman experienced left facial swelling and pain, particularly during mouth opening, for approximately 2 months. She had no history of trauma, dental problems, or other systemic disorders. No limb weakness, pain, or rash was ever noticed by the patient. She initially received conservative treatment including painkillers; however, the symptoms persisted. Laboratory tests revealed that white blood cell, hemoglobin, platelets and electrolyte counts were within normal limits.

Computed tomography (CT) (Figure 1) scans of the head and neck revealed an irregular heterogeneously enhanced lesion with a maximum diameter of approximately 3 cm in the left masseter muscle. Clinically, hemorrhage, a degenerative change in muscle and a vascular neoplasm were first considered and the mass was less likely to be malignant due to the location. Sonography-guided aspiration cytology of the lesion revealed the presence of only inflammatory cells and finally, tumor excision was performed.

The gross tumor consisted of two friable fragments measuring up to 1.0 × 1.0 × 0.5 cm in size, soft to elastic in consistency and tan in color. Microscopically, the tumor was composed of skeletal muscle tissue with degenerative and focal atrophic changes characterized by fused and
small round muscle fibers. Figure 2 shows the Endomysial or perifascicular infiltration with lymphocytes. Masson trichrome staining demonstrated the muscular characteristic of the degenerative mass. Both Congo red staining and amyloid immunostaining were negative for amyloid accumulation. The histopathologic findings and histochemical and immune-histochemical staining were consistent with MMM rather than a true neoplasm.

RESULTS AND DISCUSSION

We presented a case of unilateral MMM in a young woman without related factors such as trauma, infection and autoimmune disease and no other muscle or organ involvement was found. Differential diagnoses including a group of inflammatory myopathies, myositis ossificans and eosinophilic myositis/perimyositis was considered.

Dermatomyositis (DM) affects children and adults from young to middle age and is predominant in female patients and such patients often present preceding skin rashes before developing muscle weakness. Microscopically, characteristic perifascicular atrophy with mild or absent inflammatory cell infiltrates was observed. Polymyositis (PM) affects adults and is predominant in women which results in symmetrical proximal muscle weakness. No specific microscopic features were observed for PM. Inclusion body myositis/myopathy (IBM) often affects adults in the sixth decade of life and is predominant in men and results in asymmetrical limb muscle weakness.

Microscopically, typical rimmed vacuoles within muscle fibers and endomysial infiltrates with mononuclear cells between normal-appearing muscle fibers are observed in IBM. Eosinophilic inclusion bodies can be identified and confirmed using histochemical staining, such as amyloid immunostaining. Muscular dystrophy is less suspected because of the absence of limb weakness and predominance in men (Dalakas, 1991, 2011, 2010; Janice et al., 2013; Askanas et al., 2009, 1992).

Myositis ossificans can involve the masseter muscles and this condition is often attributed to trauma. Imaging and histopathological studies can be used to identify osseous lesions (Piombino et al., 2013; Muralidhar et al., 2014). Idiopathic eosinophilic myositis is a rare disease characterized by hypereosinophilia in peripheral blood and/or in the bone marrow and eosinophilic infiltration of the skeletal muscle accompanied by parasitic infections or other systemic disorders (Krahn et al., 2006).

Based on a review of the literature, our case did not fulfill any criteria of the aforementioned myopathies due to the unique location, absence of limb weakness and non-specific histopathologic findings. Therefore, we diagnosed the patient with MMM, which is rare in humans and is easily misinterpreted as a neoplasm. We cannot completely exclude that MMM may be an initial manifestation of one of the aforementioned myopathies and we planned to continue follow-up of this patient.

Conclusion

MMM is rare and has not been extensively documented in humans and easily misinterpreted as a neoplasm. We presented the case of MMM in a young woman; however, the collection of additional cases and further investigation are necessary for understanding of the disease.
REFERENCES


