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Letter to Editor

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Sporadic inclusion body myositis (sIBM) in a patient with a history of diffuse large B-cell lymphoma

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Sporadic inclusion body myositis (sIBM) is an insidious myopathy that affects patients late in life. Patients present with the slowly progressive disabling weakness that mostly includes quadriceps, finger flexor, and ankle dorsiflexion muscles. sIBM drives unrecognized early in the disease course, requiring increased awareness by clinicians.[1]

Though malignancy is not known comorbidity of sIBM, there have been several case reports, describing the development of typical features of sIBM after the diagnosis of malignancy, especially chronic lymphocytic leukemia.[2] We report the comorbidity of diffuse large B-cell lymphoma in a patient with sIBM for the first time.

A 69-year-old woman was referred to our outpatient clinic with difficulty in rising from chair and climbing stairs. She was a known case of diffuse large B-cell lymphoma [CD 45+, CD 20+, Ki67 (80%), CD30-, CD15-, CD3-] at 64 years of age, and was treated with cyclophosphamide, doxorubicin hydrochloride, vincristine sulfate, and prednisone chemotherapy regimen. Since the initiation of treatment, she developed a slowly progressive proximal lower limb weakness.

At the time of examination, her Medical Research Council (MRC) strength (right/left) was as follows: shoulder abduction 5/5, wrist flexion and extension 5/5, finger flexion 4/5, finger extension 5/5, hip flexion 4/4, knee extension 4+/4+, and knee flexion 4+/4+. Laboratory studies revealed creatine kinase (CK) of 834 U/l (normal value of < 195). Electromyography and nerve conduction study showed a myopathic process with positive sharp wave and fibrillation potentials predominantly in the quadriceps, tibialis anterior, and finger long flexors muscles.

The patient was referred for left vastus

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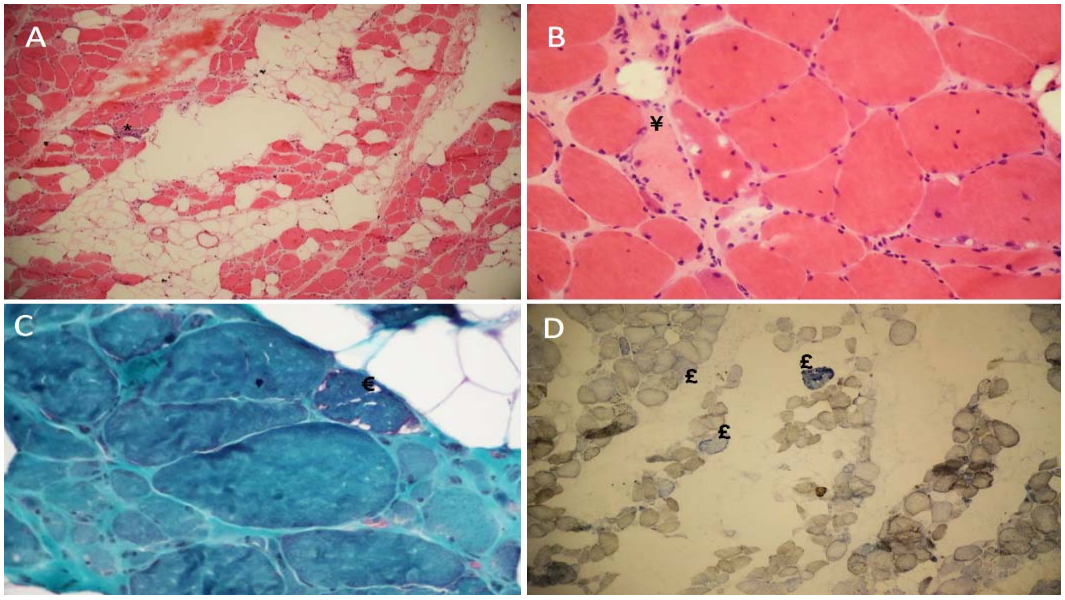

Figure 1. A: Severe atrophic changes with some necrotic fibers, focal endomysial chronic inflammatory cell infiltration (*), and extensive adipose tissue replacement (hematoxyline and eosin × 40), B: Atrophic round fibers, internalization of nuclei, necrosis (¥) and some tiny cytoplasmic vacuoles (hematoxyline and eosin × 400), C: Red-rimmed vacuoles (€) (modified Gomori Trichrome × 400), D: Presence of some cox-negative fibers (£) (Cytochrome C oxidase/succinate dehydrogenase ×100)

lateralis muscle biopsy. The muscle biopsy revealed severe atrophic changes with round atrophic and hypertrophic fibers with some necrotic and degenerative/regenerative fibers.

Myophagocytosis was seen with partial invasion associated with few foci of endomysial chronic inflammation. Endomysial connective tissue was focally severely increased with marked adipose tissue replacement in hematoxylin and eosin (H&E) staining (Figure 1, A and B). Modified Gomori Trichrome staining revealed some fibers containing red-rimmed vacuoles as well as some ragged-red-fibers (Figure 1, C). Cox + SDH reactions reveal some cox-negative fibers (Figure 1, D). The diagnosis of inclusion body myositis was then confirmed.

Sporadic inclusion body myositis (sIBM), although a rare disease, is the most common acquired muscle disorder in individuals over the age of 50. The clinical picture consists of a proximal myopathy. Some degree of asymmetry and atrophy is described in some cases. There is a predilection for the involvement of quadriceps femoris and deep flexors of hand. There is no established treatment for this disease, and immunotherapy has shown little effect.[1]

Other than clinical and pathologic features and lack of response to immunotherapy sIBM has another distinct feature. Associated malignancy is well established in other types of inflammatory myopathies, but this relationship is not as robust in sIBM. Although cancer is not a known comorbidity of sIBM, there have been several case reports, describing the evolution of typical features of sIBM after the diagnosis of malignancy, especially chronic lymphocytic leukemia. The issue was first introduced in a case report describing a patient with biopsy-proven sIBM and T-cell chronic lymphocytic leukemia (CLL).[2] In a cohort study on 62 patients with sIBM, four other cases with T-cell or B-cell CLL were also described.[3] Other case reports include endometrial carcinoma, carcinoma of the bladder, and hepatocellular carcinoma.[4] There are still many questions regarding the overall risk of cancer amongst the patients with sIBM.[4] A 2001 study suggested a small increased risk of malignancy [standardized incidence ratios (SIR) of 2.4, with 95% confidence interval (CI) of 1.2-4.9]; but this was not confirmed in a newer study.[5] There are still many questions regarding the pathologic mechanism of sIBM. The issue is still open, and more studies are needed for a definite answer.

The case reports, especially the ones that brought up the association between sIBM and CLL, can also point to a pathologic mechanism for

sIBM; an immunologic process involving T-cells. On the other hand, this association may suggest the need for malignancy surveillance in patients with sIBM.

In conclusion, we described the possible association of diffuse large B-cell lymphoma in a patient with sIBM. The disease emerged following the diagnosis of malignancy, and had typical clinical and pathological features of sIBM. Patients with diffuse large B-cell lymphoma have increased risk of autoimmune diseases including diffuse connective tissue disease. The concordance between this report and former cases of CLL and sIBM emphasizes the role of lymphoid cells (B-cell or T-cell) in the pathologic mechanism of this disease.

Conflict of Interests

The authors declare no conflict of interest in this study.

Acknowledgments

None.

References

1. Weihl CC. Sporadic inclusion body myositis and other rimmed vacuolar myopathies. Continuum (Minneap Minn) 2019; 25(6): 1586-98.
2. Arnardottir S, Ansved T, Nennesmo I, Borg K. Report of a patient with inclusion body myositis and CD8+ chronic lymphocytic leukaemia--post-mortem analysis of muscle and brain. Acta Neurol Scand 2001; 103(2): 131-5.
3. Beck EH, Amato AA, Greenberg SA. Inclusion body myositis and chronic lymphocytic leukemia: A case series. Neurology 2014; 83(1): 98-9.
4. Tiniakou E, Mammen AL. Idiopathic inflammatory myopathies and malignancy: A comprehensive review. Clin Rev Allergy Immunol 2017; 52(1): 20-33.
5. Buchbinder R, Forbes A, Hall S, Dennett X, Giles G. Incidence of malignant disease in biopsy-proven inflammatory myopathy. A population-based cohort study. Ann Intern Med 2001; 134(12): 1087-95.