# Cicatricial Fibromatosis Causing Cervical Myelopathy Due to Rapid Growth after Removal of Meningioma: A Case Report

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Cicatricial fibromatosis is often associated with inflammatory cell infiltration[1] and neovascularization[2], but it rarely occurs in the soft tissues of the neck[3]. This is the first report indicating that cicatricial fibromatosis caused cervical myelopathy after resection for spinal meningioma.

A 72-year-old female presented with numbness and pain in the left upper limb, and a physical examination indicated no obvious muscle weakness or increased deep tendon reflex. A computed tomography scan indicated an area of calcification in the left dorsal canal (Fig. 1A). Further, enhanced MRI indicated a homogeneous contrast effect in the mass lesion (Fig. 1B) that was diagnosed as spinal meningioma. We performed a Simpson grade II tumor resection.

Approximately 3 months after primary surgery, the patient's gait dysfunction and upper extremity numbness were found to gradually deteriorate. Enhanced MRI revealed an occupying lesion with a contrast effect in the posterior area of the spinal cord (Fig. 2A, 2B), which was initially identified as a recurrence of spinal meningioma. Pathological findings following biopsy indicated fibrous tissue and chronic inflammation. We believed that the dynamic factor led to the formation of inflammatory tissue. Therefore, we opted for a second surgery for posterior fixation. After a month, the contrasted extradural tissue was resected and cervical decompression and C2-6 posterior fusion was performed (Fig. 3A). Following the second surgery, a pathological diagnosis of benign cicatricial fibromatosis of the cervical vertebrae was confirmed (Fig. 4A-C).

The surgical outcome remained good until 3 years after the second surgery, at which point MRI revealed the disappearance of the contrasted extradural tissue (Fig. 2C, 2D). The surgical scar developed into a keloid and is being treated by a dermatologist 3 years after surgery (Fig. 3B).

Samples detected continuous growth of collagen fibers in the vertebral ligaments, which contained a flavum ligament or posterior longitudinal ligament and spindle-shaped cells and indicated neovascularization. A perivascular inflammatory cell infiltrate was composed mainly of lymphocytes (Fig. 4A). Spindle cells were partly α-smooth muscle actin (αSMA) positive (Fig. 4B) and fibroblasts and myoblasts were desmin negative as determined by immunostaining.

Because the mass lesion occurred in the same operative lesion, we suspected either cicatricial fibromatosis (hypertrophic scarring) or desmoid-type fibromatosis. However, it can sometimes be difficult to distinguish desmoid fibromatosis from reactive processes, such as scarring or other benign fibroblastic neoplasms[4], as was our challenge. In the histological analysis, both cicatricial fibromatosis (hypertrophic scarring) and desmoid-type fibromatosis were accompanied by an abundance of collagen fibers. β-catenin staining was performed to demonstrated differences between the two, since positive nuclear staining in desmoid-type fibromatosis is very common[4,5]. In our case, the nucleus was not stained (Fig. 4C). Furthermore, for the formation of cicatricial fibromatosis, T cells infiltrating scar tissues are capable of producing TNF-β, which has been reported to increase the growth of infant foreskin fibroblasts and could, therefore, also contribute to the abnormal growth of scar tissue[1]. In addition, reports have confirmed the presence of αSMA-positive myofibroblasts in keloid tissue. Moreover, because genetic factors are reported to be associated with keloid genesis[6,7], the patient may have been constitutionally

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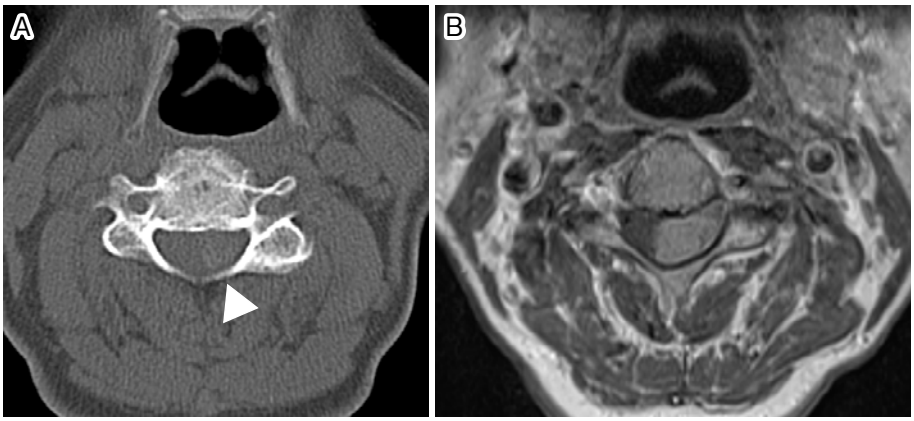


**Figure 1.** A: Axial computed tomography image. The arrowhead points to the calcification area. B: Preoperative axial enhanced T1 magnetic resonance image.

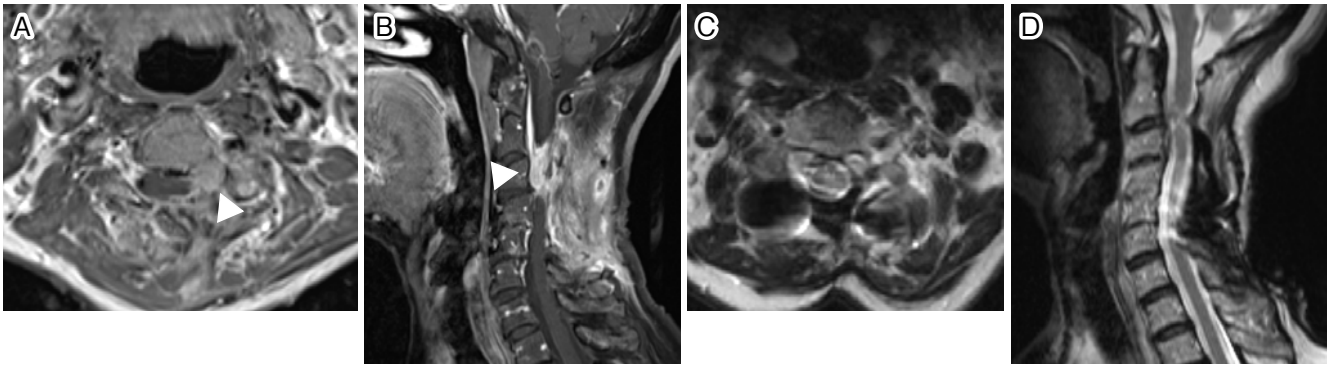


**Figure 2.** A, B: 3-month postoperative axial (A) and sagittal (B) enhanced T1 magnetic resonance images. C, D: Axial (C) and sagittal (D) T2 magnetic resonance images 3 years after the second surgery.

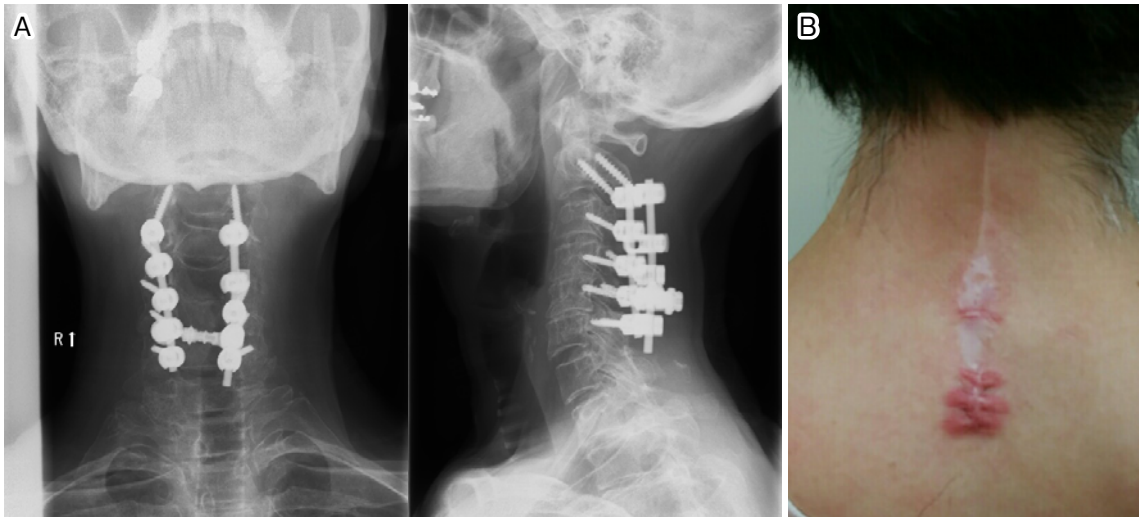


**Figure 3.** A: Anterior–posterior and lateral radiograph after the second surgery. B: Clinical photography indicates keloids in the surgical scar.

predisposed to abnormal fibrosis, as can be inferred from keloids in the surgical scar.

Throughout her life, the patient had spent much time performing repetitive forward bending at home, which would have caused much mechanical stress in the neck. Repetitive mechanical stress has been demonstrated to promote collagen synthesis and deposition, resulting in hypertrophic scarring[8,9]. In addition, neurogenic inflammation due to mechanical stress has been reported to be a potential cause of keloids and hypertrophic scars[10]. We hypothesized that the patient's repetitive neck motions had caused a bunch of inflammation and rapidly growing scarring.

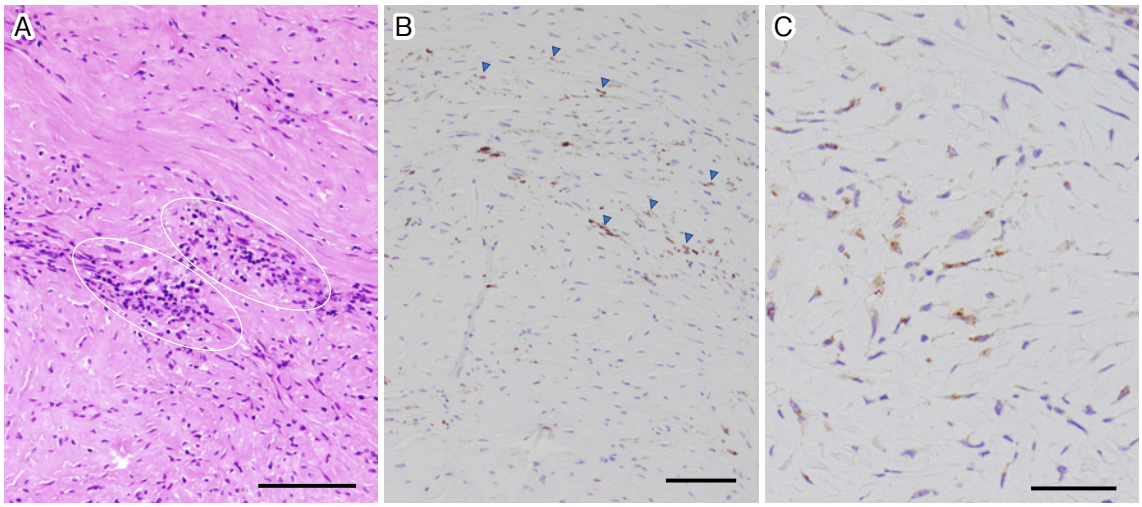


**Figure 4.** A: Hematoxylin and eosin staining of the tumor indicated lymphocyte-dominated inflammatory cell infiltration surrounding the blood vessels inside white circles. B, C: Immunohistochemistry showed α-smooth muscle actin-positive myofibroblast (depicted by blue arrowheads) (B), and the myofibroblast nucleus was not stained by β-catenin (C). Bars are 100 μm (A, B) and 50 μm (C).

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**Ethical Approval:** Approval code: 2005-0354. This study was approved by the committee on ethics on human research of Nagoya University Graduate School of Medicine.

**Informed Consent:** Informed consent was obtained from a participant in this study.

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