JOURNAL OF NEUROSURGERY:
Case Lessons

J Neurosurg Case Lessons 4(4): CASE22184, 2022
DOI: 10.3171/CASE22184

Spinal intraosseous arteriovenous fistulas with perimedullary drainage associated with vertebral compression fracture: illustrative case

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BACKGROUND Although osseous involvement is occasionally observed in spinal epidural arteriovenous fistulas (AVFs) or seen as a part of diseases of spinal arteriovenous metameric syndrome, purely intraosseous spinal AVFs are extremely rare. Their clinical and imaging characteristic features are not well known. The authors present a case of purely intraosseous AVFs associated with compression fracture.

OBSERVATIONS A 76-year-old man presented with back pain and progressive myelopathy. Computed tomography showed compression fracture of the T12 vertebral body and dilatation of perimedullary veins. Spinal angiography revealed an intraosseous AVF at the T12 spine level, which was fed by multiple feeders of ventral somatic branches and drained into the paravertebral and perimedullary veins. The intraosseous AVF was completely occluded by the combined techniques of transarterial and transvenous embolization with glue and a coil. The symptoms disappeared within 1 month after embolization.

LESSONS Although extremely rare, spinal intraosseous AVFs can develop after compression fracture and cause congestive myelopathy. Combined transarterial and transvenous embolization is useful for the specific case of spinal intraosseous AVFs with both paravertebral and perimedullary drainage.

https://thejns.org/doi/abs/10.3171/CASE22184

KEYWORDS spinal arteriovenous fistula; spinal arteriovenous malformation; compression fracture; spinal artery; spinal vein; embolization

According to recent developments in imaging technology and the increasing recognition of extradural spinal arteriovenous fistulas (AVFs), the number of patients diagnosed with extradural AVFs has been increasing.[1–4] Among the extradural spinal AVFs, epidural AVFs are the most common pathology and are located in the epidural space, mostly in the ventral epidural space.[2] Osseous involvement is occasionally seen in spinal extradural AVFs, most of which are caused by epidural AVFs.[5–7] Bone erosion by the epidural venous pouch or varix secondary to AVFs is a common feature of osseous involvement. Enlarged feeders of the ventral somatic branch and AVFs themselves can cause bone erosion at the posterior surface of the vertebral body. Spinal arteriovenous metameric syndrome, also called Cobb syndrome, can also involve the vertebra.[8] However, pure intraosseous AVFs are extremely rare.

Here, we demonstrate a case of intraosseous AVF with paravertebral and perimedullary venous drainage associated with compression fracture, which presented with congestive myelopathy. The intraosseous AVF was successfully treated by combined transarterial and transvenous embolization. The pathophysiology and techniques of endovascular treatment are discussed with a literature review.

Illustrative Case

A 76-year-old man presented with a 2-month history of back pain and a 1-month history of progressive numbness of both legs. Neurological examination showed weakness (grade 3/5) and hypesthesia of the bilateral lower extremities. Deep tendon reflexes were slightly increased. Urinary retention and fecal incontinence were also noted. T2-weighted magnetic resonance imaging (MRI) of the thoracic and lumbar spine

ABBREVIATIONS AVF = arteriovenous fistula; CT = computed tomography; MRI = magnetic resonance imaging; NBCA = *n*-butyl cyanoacrylate.

INCLUDE WHEN CITING Published July 25, 2022; DOI: 10.3171/CASE22184.
SUBMITTED April 22, 2022. **ACCEPTED** May 19, 2022.

revealed an old compression fracture of the T12 vertebral body and edema of the lower spinal cord and conus medullaris with dilated and multiple flow voids on the surface of the cord (Fig. 1A), suggesting spinal dural or epidural AVF. A dilated flow void at the dorsal surface of the vertebral body was also seen. Spinal angiography demonstrated AVF fed by multiple ventral somatic branches from the right subcostal artery (Fig. 1B–D). Multiplanar reformatted images of rotational angiography of the right subcostal artery clearly showed the AVF located in the vertebral body, which was drained via the basivertebral vein to the ventral epidural vein, forming a varix (Fig. 1E). The shunted blood from the ventral epidural vein drained laterally to the right subcostal vein, and it also drained retrogradely via the radiculomedullary vein to the perimedullary veins. Extraspinal venous drainage was continued via the longitudinal intercostal anastomotic vein between the right subcostal vein and right T10 posterior intercostal veins into the azygos vein (Fig. 1D). Because the intraosseous AVF was fed by multiple feeders with multiple fistulas and drains via a long epidural vein into the extraspinal and intradural veins, there was a potential risk of incomplete occlusion of the AVF with persistent perimedullary drainage by transarterial embolization alone. Therefore, combined transvenous and transarterial embolization were performed with the patient under local anesthesia (Fig. 2). For the arterial approach, a 5-Fr guiding sheath with a shepherd hook shape (Axcel guide, Medikit) was introduced from the right femoral artery into the right T12 subcostal artery. A microballoon catheter (SHORYU, Kaneka) and a 1.3-Fr microcatheter (Defrictor, Hirata) were introduced through the 5-Fr guiding sheath into the subcostal artery. The 1.3-Fr microcatheter was subsequently introduced into a feeding artery of the ventral somatic branch distally close to the fistulous point as much as possible (Fig. 2A). The microballoon catheter was positioned crossing over the origin of the feeding artery (Fig. 2C). Then, a 4-Fr guiding sheath (Fubuki, Asahi Intecc) was advanced into the azygos vein with a right cubital venous approach. A 1.6-Fr/2.6-Fr coaxial microcatheter system (Carnelian MARVEL S,

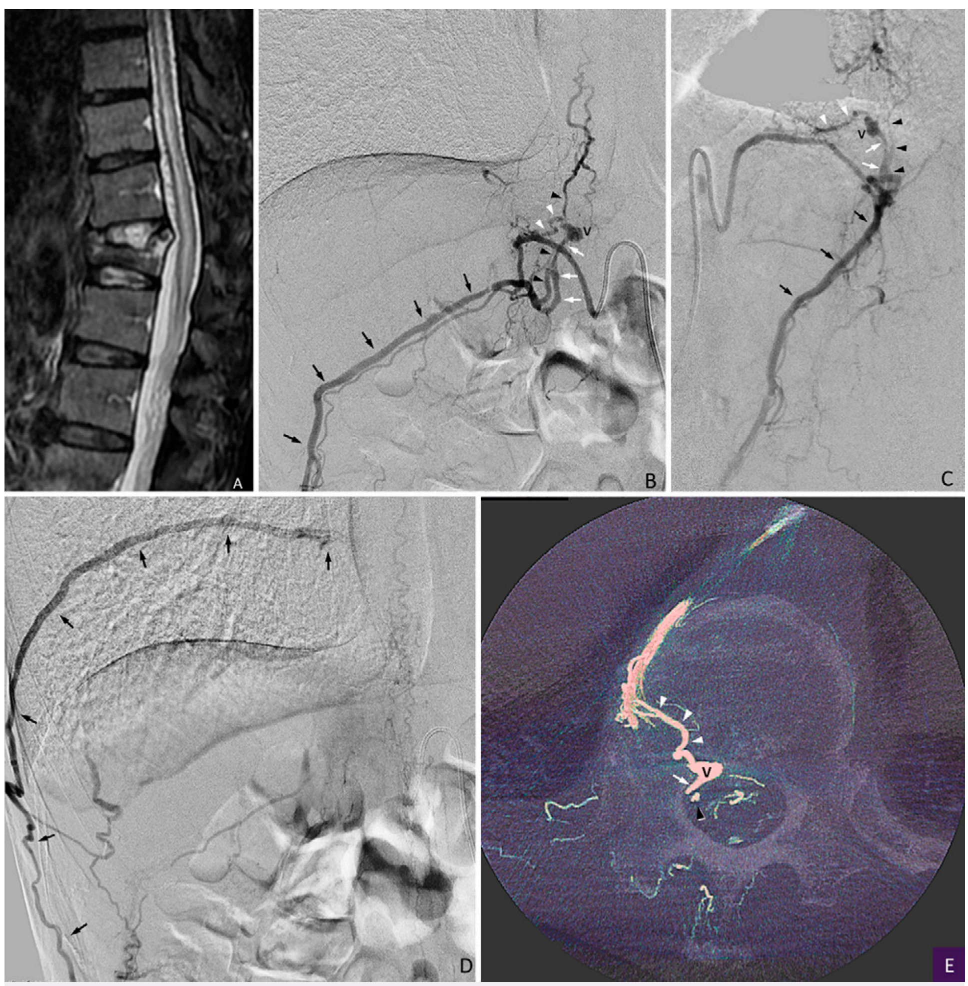

FIG. 1. Sagittal fat-suppressed T2-weighted image (**A**) shows the high signal intensity of the spinal cord and conus medullaris with multiple small flow voids surrounding the spinal cord. A compression fracture of the T12 vertebral body is also noted. Frontal views (**B and D**), lateral view (**C**), and axial reformatted image (**E**) of selective angiography of the right subcostal artery at early (**B and C**) and late (**D**) arterial phases show the AVFs fed by multiple ventral somatic branches and draining into the basivertebral vein (*white arrowheads*) and epidural vein (*white arrows*) with a varix (*V*) and then into the paravertebral veins (*black arrows*) (the subcostal vein, intersegmental anastomotic vein, T10 intercostal vein, and the azygos vein) and retrogradely into the perimedullary veins via the radiculomedullary vein (*black arrowheads*).

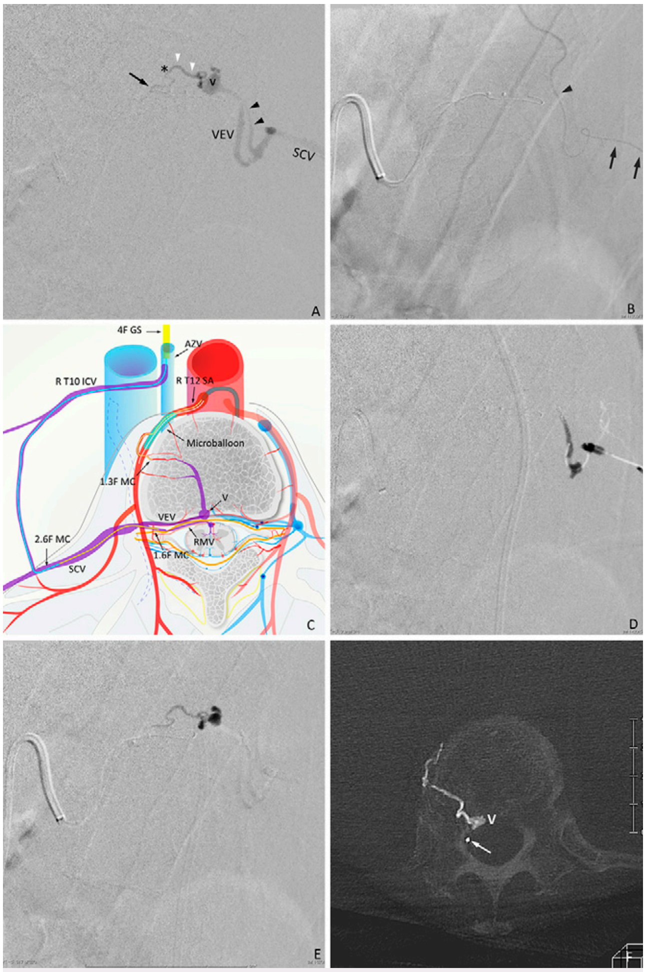

FIG. 2. A: Lateral view of selective angiography of the feeder of a ventral somatic branch shows an AVF (*asterisk*) draining into the basivertebral vein (*white arrowheads*), ventral epidural vein (*VEV*), subcostal vein (*SCV*), and radiculomedullary vein (*black arrowheads*). The *arrow* indicates the tip of a 1.3-Fr microcatheter. **B:** Lateral view of selective venography shows retrograde intradural drainage via the radiculomedullary vein. The tip of a 1.6-Fr microcatheter (*arrowhead*) is introduced transvenously into the outlet to the radiculomedullary vein via the subcostal vein (*arrows*). **C:** Schematic of the angioarchitecture and embolization techniques of the presented case. The AVFs were fed by multiple ventral somatic branches of the right subcostal artery (R T12 SA) and drained into the basivertebral vein and ventral epidural vein (*VEV*), forming a varix (*V*) and then into the subcostal vein (*SCV*) and the radiculomedullary vein (*RMV*). A 1.3-Fr microcatheter (*1.3F MC*) was advanced into a feeder of the ventral somatic branch of the right subcostal artery, and a microballoon catheter was placed in the stem of the subcostal artery. A 4-Fr guiding sheath (*4F GS*) was placed in the azygos vein (*AZV*) with a right cubital venous approach, a 1.6-Fr (*1.6F MC*)/2.6-Fr (*2.6F MC*) coaxial microcatheter system was introduced into the right subcostal vein (*SCV*) via the right T10 intercostal vein and a longitudinal anastomotic vein, and a 1.6-Fr microcatheter was further advanced into the extradural portion of the radiculomedullary vein (*RMV*). **D:** Lateral view of angiography during injection of 33% NBCA-lipiodol mixture via the 1.6-Fr microcatheter in the extradural portion of the radiculomedullary vein. The glue cast fills the radiculomedullary vein and the epidural vein while partially filling the paravertebral vein.

FIG. 2. (*continued*) →

Tokai) was advanced through the 4-Fr guiding sheath to the right T10 intercostal vein, intersegmental longitudinal anastomotic vein, and right subcostal vein. The 1.6-Fr microcatheter was further advanced through the epidural vein into the outlet to the radiculomedullary vein (Fig. 2B and C). A small detachable coil (2 mm × 2 cm; AXIUM, Medtronic) was placed in the transdural portion of the radiculomedullary vein, and a 33% *n*-butyl cyanoacrylate (NBCA)-lipiodol mixture was subsequently injected via the same microcatheter to obliterate both the paravertebral and intradural venous drainage (Fig. 2D). After transvenous embolization, a 20% NBCA-lipiodol mixture was injected from a feeder via the 1.3-Fr microcatheter under temporal occlusion of the stem of the right subcostal artery with the microballoon catheter, and the glue mixture filled into the fistula, the basivertebral vein, and the varix (Fig. 2E and F). Immediately after embolization, angiography showed disappearance of the intraosseous AVF (Fig. 3). The patient's symptoms improved rapidly. He had fully regained his motor function within 4 weeks, and urinary retention and fecal incontinence were completely resolved. Hypesthesia of the bilateral lower extremities was also completely resolved, and MRI showed the disappearance of abnormal signal intensity of the spinal cord and enlarged perimedullary veins at the 3-month follow-up. Follow-up spinal angiography 1 year after embolization showed no recurrent/residual AVF.

Literature Review

The PubMed and Embase databases and Google Scholar were searched in December 2021 for English-language publications of case series and case reports of spinal intraosseous AVFs using the following terms in various combinations: "arteriovenous fistulas," "arteriovenous malformation," "arteriovenous shunt," "spinal," "spine," "extradural," "vertebral," "osseous," and "intraosseous." After reviewing the retracted cases, the following cases were excluded: epidural AVFs or paraspinal AVFs involving the vertebra and spinal metameric syndrome. The 8 remaining patients with purely intraosseous AVFs in 8 published papers were further reviewed in detail.[9–16] Table 1 shows the characteristics of the 8 previously published patients and our patient. There were 5 males and 4 females, with ages ranging from 8 to 93 years (mean 64.1 years). The AVFs involved the thoracic spine in 4 cases and the lumbar spine in 5 cases. In all but 1 case, the spinal levels involved were between the T12 and L4 levels. The symptoms were back pain in 6 patients, myelopathy in 6 patients, radiculopathy in 1 patient, and no symptoms in 1 patient. Five patients had a history of trauma, and 6 patients, including those 5 patients and our patient, showed a compression or transverse fracture of the vertebral body at the level corresponding to the AVF. The locations of the AVFs were the ventral element (vertebral body) in those 6 patients and the dorsal elements (lamina with or without the transverse process) in the other 3 patients. The AVFs were fed by the unilateral segmental artery in 5 patients, bilateral segmental arteries at the affected level in 1 patient, and multiple bilateral multisegmental arteries in 1 patient. In the remaining 2 patients, feeding arteries

FIG. 2. E: Lateral view of angiography during injection of 20% NBCA-lipiodol mixture via the 1.3-Fr microcatheter in the feeder. The glue cast fills beyond the fistula into the basivertebral vein and the epidural varix. **F:** Axial reformatted image of rotational angiography without contrast injection clearly shows the glue cast in the feeder (a ventral somatic branch), the basivertebral vein, and the epidural varix (*V*). The *arrow* indicates the radiculomedullary vein embolized transvenously with glue and coil.

FIG. 3. Frontal view of selective angiography of the right subcostal artery immediately after embolization shows disappearance of the AVFs.

were not described. Draining veins were not described in 3 cases. In 4 cases, the AVFs drained into the epidural veins, then into the perimedullary vein in 1, into the paravertebral veins in 1, and into both the perimedullary and paravertebral veins in 3 cases. In the other case, the AVFs directly drained into the paravertebral veins. All 6 cases of AVFs located in the vertebral body were treated by endovascular treatments.[9–13] In contrast, the other 3 cases involving the dorsal elements were treated by surgical resection.[14–16] Among the 6 cases treated by endovascular techniques, 3 cases were treated by transarterial embolization alone using NBCA, Onyx, and/or coils. One case was treated by an aortic stent graft.[13] The other 2 cases were treated by combined techniques of transarterial embolization with direct puncture embolization or transvenous embolization with NBCA. Complete resolution of AVFs was obtained in all 6 cases treated by endovascular techniques and in 2 of 3 cases treated with surgery. No complications were noted, and symptoms were resolved or markedly improved after treatment in all patients.

Discussion

Among the 9 published cases of spinal intraosseous AVFs, there are 2 types of AVFs involving the ventral and dorsal elements of the vertebra. There are some different characteristics in pathophysiology between the ventral and dorsal types. All 6 patients with ventral-type AVFs had a compression or transverse fracture at the same vertebra, and 4 of them had a history of trauma. No patients with dorsal-type AVFs showed a vertebral fracture or history of trauma.[14–16] It is well known that trauma can cause some types of AVFs. Compression fracture can cause tearing of the vascular wall in the vertebral body, resulting in AVF formation. In addition, both trauma and fracture can cause venous occlusion and/or venous thrombus and inflammation in and around the vertebral body, all of which activate angiogenesis and contribute to AVFs. Therefore, intraosseous AVFs of the vertebral body are acquired lesions that may be caused mainly by compression/transverse fractures. However, the dorsal type is not related to trauma or fracture, and it may be an intraosseous vascular malformation.

Regarding drainage pattern and symptoms, perimedullary drainage causing congestive myelopathy was found in 4 of 6 cases of ventral-type AVFs but in no cases of dorsal-type AVFs. One patient with a dorsal-type AVF showed myelopathy due to compression by an enlarged vein. The internal vertebral (epidural) venous plexus lies within the spinal canal between the dura mater and the vertebra, which receives blood from the vertebral body via the basivertebral veins and from the spinal cord via the radiculomedullary veins or bridging veins and connects to the external vertebral plexus through the intervertebral foramen. Venous blood from the vertebral body is mainly drained by the basivertebral vein into the ventral epidural plexus. A small area of the anterior part of the vertebral body is drained directly into the anterior external vertebral plexus via small tributaries. Blood from the posterior elements of the vertebra is drained mainly into the posterior external vertebral plexus via small tributaries and less dominantly into the dorsal epidural plexus. Ventral-type AVFs are generally fed by the ventral somatic branches and drain through the basivertebral vein into the ventral epidural venous plexus. Shunt blood from the ventral epidural plexus drains into the paravertebral vein (external vertebral plexus). Retrograde intradural venous drainage and subsequent congestive myelopathy will occur when paravertebral drainage is disturbed by thrombosis or steno-occlusive changes in the paravertebral drainage routes. Although there is no detailed description of drainage routes in the three cases of dorsal-type AVFs, the dorsal type may drain directly into the posterior external vertebral plexus because the dorsal epidural plexus develops much less than the ventral one. Furthermore, the arch is dominantly drained directly into the posterior external vertebral plexus. Therefore, most cases of dorsal-type AVF may not present with congestive myelopathy. A large epidural varix on the drainage route can cause compressive myelopathy or radiculopathy.

Among the 6 cases of intraosseous AVF treated by endovascular treatment, various techniques were used. Two cases with paravertebral venous drainage were successfully treated by occlusion of the feeding artery with coils or aortic stent grafts.[12,13] Although the angioarchitecture of the 2 cases was not clearly described, the 2 cases consisted of a large and direct AVF with a single feeder and single drainage vein. In the other 4 cases with perimedullary venous drainage, the AVFs consisted of multiple feeders and a single drainage vein of the basivertebral vein. All 4 cases were successfully treated by using liquid embolic materials, such as Onyx or NBCA, that filled the venous sides. For the treatment of cases with congestive myelopathy, it is most important to occlude the retrograde intradural drainage. However, it is often difficult to obtain sufficient filling of the liquid embolic materials injected from the feeding arteries far away into the intradural drainage vein, and incomplete filling may result in residual or recurrent AVFs. Onyx, a nonadhesive embolic material, allows a more durable injection of a larger amount of material delivered in a single session. Ou et al. noted the utility of Onyx for a case of intraosseous spinal AVFs.[10] Transvenous embolization is another approach for embolization of the drainage vein of intraosseous AVFs, as described in our patient. Intradural drainage is easily occluded at first when a microcatheter can be navigated transvenously into the affected epidural vein. Recent advances in microcatheter/microguidewire technology allow us to advance a microcatheter distally through the tortuous transvenous access route more easily and safely. When sufficient embolization by transarterial or transvenous approaches is difficult or fails, direct puncture embolization is an alternative approach.[11]

TABLE 1. Summary of published cases of intraosseous spinal arteriovenous fistulas

Authors & Year	Age (yrs), Sex	Symptoms	Trauma*	Vertebral Fx	Level	Locations	Drainage	Feeders	Techniques	Tx Results	Outcomes
Present case	74, M	Low-back pain, progressive myelopathy	No	Compression	T12	Ventral	ParaVV, PMV	Rt T12	TVE TAE (NBCA)	CR	GR
Jin et al., 2012[9]	68, F	Progressive myelopathy	Yes	Compression	L1	Ventral	ParaVV, PMV	Bilat L1 Bilat L2 Rt T12	TVE (failed) TAE (NBCA)	CR	GR
Ou et al., 2015[10]	57, M	Low-back pain, progressive myelopathy	Yes	Compression	T12	Ventral	PMV	Lt T12	TAE (ONYX)	CR	GR
Gjertsen et al., 2010[11]	67, M	Progressive myelopathy, back pain, radicular pain	Yes	Compression	T12	Ventral	ParaVV, PMV	Rt T12	TAE (coil), Direct embo (NBCA)	CR	GR
Imajo et al., 2015[12]	74, F	Low-back pain	Yes	Transverse	L4	Ventral	ParaVV	Rt L4	TAE (coil)	CR	GR
Iwakura et al., 2012[13]	93, F	No symptoms	Yes	Compression	L3	Ventral	ParaVV	L3	Stent graft	CR	GR
Molina et al., 1997[14]	8, M	Back pain	No	No	L4	Dorsal	NA	NA	Resection	CO	GR
Louis et al., 2011[15]	59, F	Myelopathy (compression)	No	No	T5	Dorsal	NA	Rt T5	Resection	PR	GR
Antoniades et al., 2020[16]	75, M	Back pain	No	No	L1	Dorsal	NA	NA	Resection	CO	GR

Bilat = bilateral; CO = complete obliteration; CR = complete resolution; embo = embolization; Fx = fracture; GR = good recovery; NA = not available; ParaVV = paravertebral vein; PMV = perimedullary vein; PR = partial resolution; TAE = transarterial embolization; TVE = transvenous embolization; Tx = treatment.

* History of trauma.

Observations

In this report, we describe a rare case of spinal intraosseous AVFs along with a literature review. Intraosseous AVFs located at the ventral element of the vertebra were frequently associated with compression fracture and often caused congestive myelopathy. Endovascular treatments could successfully obliterate the AVFs with improving clinical symptoms.

Lessons

Although extremely rare, spinal intraosseous AVFs can develop after compression fracture and cause congestive myelopathy. Combined transarterial and transvenous embolization is useful for the specific case of spinal intraosseous AVFs with both paravertebral and perimedullary drainage.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Kiyosue, Baba. Acquisition of data: Kiyosue, Baba, Ide, Onishi, Kubo. Analysis and interpretation of data: Kiyosue, Baba. Drafting the article: Baba, Tokuyama. Critically revising the article: Kiyosue, Baba. Reviewed submitted version of manuscript: Kiyosue. Approved the final version of the manuscript on behalf of all authors: Kiyosue. Study supervision: Kiyosue.

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