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Minimally invasive surgery for spinal cerebrospinal fluid–venous fistula ligation: patient series

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BACKGROUND Cerebrospinal fluid–venous fistulas (CVFs) may cause cerebrospinal fluid leaks resulting in spontaneous intracranial hypotension (SIH). Surgical treatment of CVFs aims to eliminate abnormal fistulous connections between the subarachnoid space and the epidural venous plexus at the level of the nerve root sleeve. The authors propose a percutaneous minimally invasive technique for surgical ligation of CVF as an alternative to the traditional open approach using a tubular retractor system.

OBSERVATIONS Minimally invasive surgical (MIS) ligation of spinal CVF was performed in 5 patients for 6 CVFs. The definite disconnection of the CVF was achieved in all patients by clipping and additional silk tie ligation of the fistula. None of the patients experienced surgical complications or required transition to an open procedure. One patient underwent 2 MIS procedures for 2 separate CVFs. Postoperative clinical follow-up and cranial magnetic resonance imaging confirmed resolution of symptoms and radiographic SIH stigmata.

LESSONS MIS ligation of CVFs is safe and efficient. It represents an elegant and less invasive procedure, reducing the risk of wound infections and time to recovery. However, preparedness for open ligation is warranted within the same surgical setting in cases of complications and difficult accessibility.

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KEYWORDS CSF–venous fistula; minimally invasive; surgery; ligation; spontaneous intracranial hypotension

Spontaneous intracranial hypotension (SIH) has an incidence of 5 per 100,000 individuals per year and is often caused by a spinal cerebrospinal fluid (CSF) leak.[1–3] Spinal CSF leaks originate from dural weakness and meningeal diverticula at the surrounding nerve root sleeves, so-called cerebrospinal fluid–venous fistulas (CVFs), or from dural tears resulting from disc herniations/osteophytes (Fig. 1).[4–6] CVFs are mainly located in the thoracic region and defined as abnormal connections between the spinal subarachnoid space and adjacent paraspinal veins that allow unregulated egress of CSF into the venous system.[7] Epidural blood patching (EBP) is regarded as the mainstay therapy for SIH caused by CSF leaks, with a success rate of 70%.[8] However, it was shown to be less effective for SIH caused by CVF.[9,10] Thus, for patients with CVF in whom EBP fails, surgical ligation of the CVF represents an important therapeutic alternative.[11–13] Ligation of the fistula under direct visualization confirms the successful elimination of the CVF and is usually performed as an open surgical approach with a high success rate.[12,14] We herein describe our experience and observations with a percutaneous, minimally invasive approach for surgical ligation of CVFs using a conventional tubular retractor system.

ABBREVIATIONS CSF = cerebrospinal fluid; CT = computed tomography; CVF = cerebrospinal fluid–venous fistula; DSM = digital subtraction myelography; EBP = epidural blood patching; FU = follow-up; IOM = intraoperative neurophysiological monitoring; MIS = minimally invasive surgical; MRI = magnetic resonance imaging; SIH = spontaneous intracranial hypotension.

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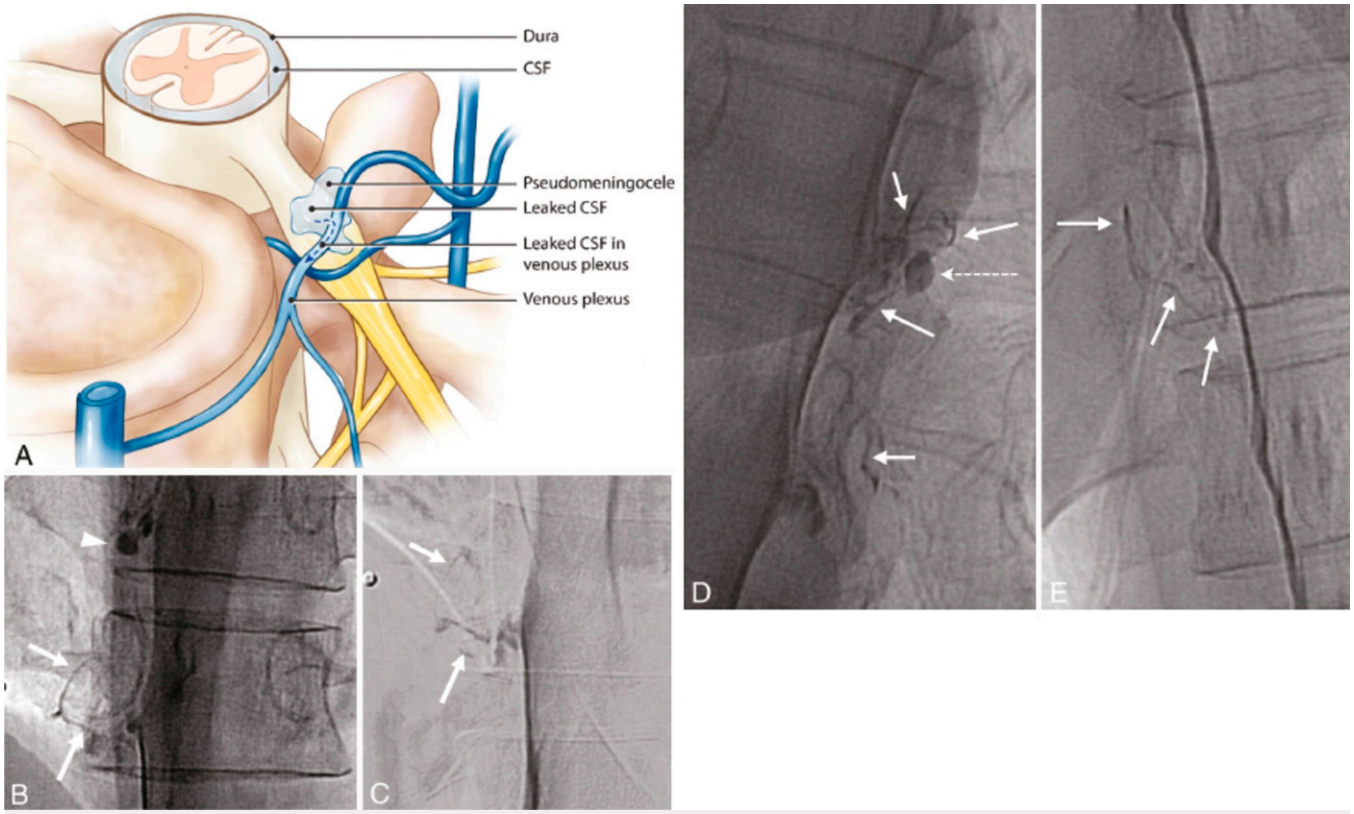

FIG. 1. Type 3 CSF leak (SLEC-N). **A**: Schematic depiction of a CSF-to-venous fistula arising from a dural tear along the nerve root sleeve beyond the epidural compartment (*see text*). Nonsubtracted (**B**) and magnified, subtracted (**C**) images from separate left-side-down DSM runs in a patient negative for SLEC with SIH. A small vascular structure, in keeping with a tortuous vein of a CVF, can be seen coursing away from the root sleeve (*arrows*). An incidental normal diverticulum is also noted at the level above (*arrowhead*). **D and E**: Nonsubtracted images of decubitus DSMs of 2 other similarly presenting patients negative for SLEC demonstrating CVFs. Globular collections of contrast (*dashed arrow*) are commonly seen near the expected zone of origin of the vein, possibly representing a focal extravasation (pseudomeningocele) of contrast or a diverticulum from which the vein appears to arise. The figure was adapted from Farb et al.[6] and used with permission from the authors and copyright permission from the *American Journal of Neuroradiology.*

Study Description

We performed a retrospective chart review, including patients who were treated for CVFs between October 1, 2018, and March 31, 2021, in our institution. The study subjects were identified using an electronic database of all patients with SIH, and demographic, clinical, and surgical data were extracted. The study was approved by the research ethics board at the University Health Network and conducted in accordance with their ethics guidelines and those of the University of Toronto. Informed consent was obtained from all patients at the time point of their surgery.

Patient Selection

Patients were selected for further diagnostic work-up when they presented with typical clinical and neurological signs of SIH. The diagnostic imaging work-up included radiography of the spine, magnetic resonance imaging (MRI) of the brain and spine, and digital subtraction myelography (DSM) followed by computed tomography (CT). DSM was performed with the patient in different positions (prone and bilateral decubitus, as described by Farb et al.[6] and Piechowiak et al.[15]) until a set of 3 negative myelograms was completed or a definitive CVF was localized. Surgical intervention was recommended for symptomatic patients who did not respond to conservative measures, including 1 to multiple blood patches and/or endovascular embolization, and who showed proof of persisting CVF in a dynamic subtraction myelogram. CVFs were localized in a preoperative angiogram by counting the segments and cross-linkage of different planes in the DSM.

Surgical Technique

All procedures were done with the patient under general anesthesia. Patients were positioned prone on a Jackson table with continuous intraoperative neurophysiological monitoring (IOM) of motor evoked potentials and somatosensory evoked potentials. Spinal localization was performed before incision using 3 sequential needles and a lateral plain radiograph. After a paramedian skin incision was made directly over the level of the CVF, subsequent percutaneous dilatation and deepening of the surgical access was performed using increasing minimally invasive surgical (MIS) tubes. The final 18-mm MIS tube was directly inserted over the site of the CVF. Final localization was repeated using fluoroscopy (Fig. 2A). At this time point, the operating microscope was brought into the field, followed by anatomical dissection of the lateral aspect of the lamina. A minilaminectomy was then performed using a 1.7-mm diamond matchstick, removing the bone far enough laterally, including a

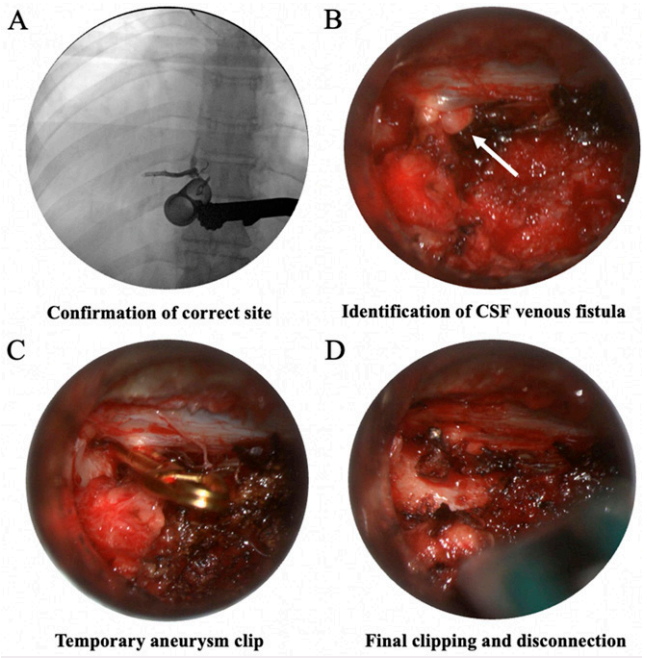

FIG. 2. Representative intraoperative imaging with fluoroscopic localization confirmation of the preembolized CVF at T9 left (**A**) and its microscopic visualization after surgical exposure (**B**). After a temporary clip was placed (**C**) and stable IOM signals were obtained, the final clipping and disconnection of the CVF were performed (**D**).

foraminotomy to allow careful exploration of the dura with the exiting nerve root. Careful removal of the yellow ligament and dissection at the lateral side of the dural sac was performed using a combination of electrocautery and Kerrison rongeurs. In case of venous engorgement occluding visualization of the fistula, these veins were coagulated and dissected carefully. The exposed lateral dura, the exiting nerve root, and the CVF were visualized (Fig. 2B). Subsequently, nerve hooks were placed around the nerve root complex. Once located, the fistula was clipped using a temporary aneurysm clip followed by IOM to confirm intact function of the nonexiting nerve roots (Fig. 2C). Once unremarkable, permanent clipping of the CVF was performed, followed by a 2-0 silk suture tie reinforcement when needed (Fig. 2D). This was reinforced with fibrin glue (DuraSeal, Integra LifeSciences) and autologous blood. The Valsalva maneuver was performed to confirm the complete closure of the fistula. Subsequently, removal of the MIS tube, irrigation with bacitracin-soaked lactated Ringer solution, and sequential layer-by-layer closure of the incision were performed. Self-resolving stitches were used for intracutaneous skin closure. No Hemovac drain was applied.

Patient Follow-Up

Patients were mobilized on the first postoperative day, and their outcome was assessed at day 1, at discharge, at 6 weeks, and 3 and 6 months after surgery. The assessment included their neurological status, presence, and quality of headaches and their ability to return to work. Our protocol included follow-up (FU) cranial MRI 3 months after the surgery for verification that radiographic stigmata of SIH had resolved.

Patient Characteristics and Surgical Results

A total of 5 patients with 6 CVFs were identified and treated surgically, including 3 females and 2 males. The mean age at surgery was 57.5 years (range 44–62 years). The duration between onset of symptoms and surgery was between 1.5 and 3 years. All patients complained of long-standing orthostatic headache, were diagnosed with a CVF via DSM, and had undergone 1 or 2 EBPs. One patient underwent additional endovascular embolization of the CVF. Upon persisting or recurrent symptoms after the attempted conservative measures, patients were consulted for surgery. Complications occurred in none of the patients. However, 2 patients showed a more complex diagnostic or clinical course.

A 62-year-old female presented with long-standing orthostatic headaches and was suspected of having SIH. Given the difficult visualization of the CSF leak in her, she required 5 CT myelogram procedures until a definitive CSF leak could be identified. For this particular patient, no abnormality was seen on the DSM; however, CT performed immediately after the myelogram in the left decubitus position showed evidence of a CVF from a diverticulum beneath the left T9 pedicle (Fig. 3). Subsequently, she underwent multiple EBPs without lasting benefit, followed by an endovascular embolization of the CVF, which led to transient relief of her symptoms for 1 month (Fig. 2). She presented again 2 months later with ongoing headache, cognitive changes, and recent difficulty swallowing, so we proceeded with surgery as described above.

A 59-year-old male was diagnosed with a CVF at T9 right, which was successfully ligated via an MIS procedure. At the time point of diagnosis, there were doubts about the exact level, because the DSM would identify a dominant CVF at T9 right but also show questionable abnormalities at the level above, at T8 right. After a symptom-free interval of 2 months, he presented with a new flair-up of headache and underwent another DSM, and he was consecutively diagnosed with a second, now more prominent CVF at T8 right, which was presumably masked by the primary, initially dominant CVF at T9 right. A second MIS procedure for ligation of this CVF was successfully performed 3 months after the initial surgery.

All patients were discharged on postoperative day 2 and seen 6 weeks later for FU in our clinic. None of the patients experienced

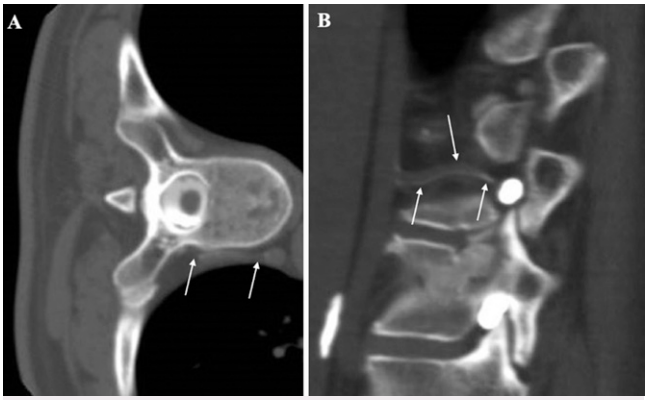

FIG. 3. CT performed immediately after myelography in the left decubitus position showing evidence of fistula with opacification of a small paravertebral vein running anteriorly to the hemiazygos vein from a diverticulum beneath the left T9 pedicle (*arrows*). Post-CT myelogram in left decubitus position with axial 3-mm image (**A**) and reformatted sagittal image (**B**).

wound complications. The mean FU for the cohort was 21.8 months (range 8–31 months). Complete resolution or improvement of symptoms was confirmed in all patients at their last FU. Four of 5 patients were back to work at 3 months after the index surgery. The patient who required a second MIS procedure reached symptom freedom and normal working status 7 months after the index procedure. All patients had characteristics of SIH in their preoperative brain MRI and demonstrated improvement at their last postoperative FU, confirming the definite ligation of the CVF (Fig. 4). An overview of the patients' demographics, clinical data, and FU is provided in Table 1.

Discussion

Observations

This study describes, for the first time, to our knowledge, a minimally invasive approach for ligation of CVF and the surgical results of 5 patients diagnosed with 6 CVFs resulting in SIH. All patients underwent MIS ligation for CVF following the same technical approach and experienced resolution or improvement of their symptoms at their last FU. Perioperative complications occurred in none of the patients. In our patients, we observed a good technical feasibility and ligation success rate of 100%, indicating that this MIS approach represents a valid alternative to open procedures. Moreover, we could observe a significantly reduced invasiveness and overall short recovery periods, similar to the benefits of other spine-related MIS procedures. None of the patients developed wound complications, and they could be discharged as early as postoperative day 2. From a technical perspective, we appreciated this targeted approach, allowing reduction of postsurgical dead space in the disrupted musculature of the spine as seen more commonly after open procedures.

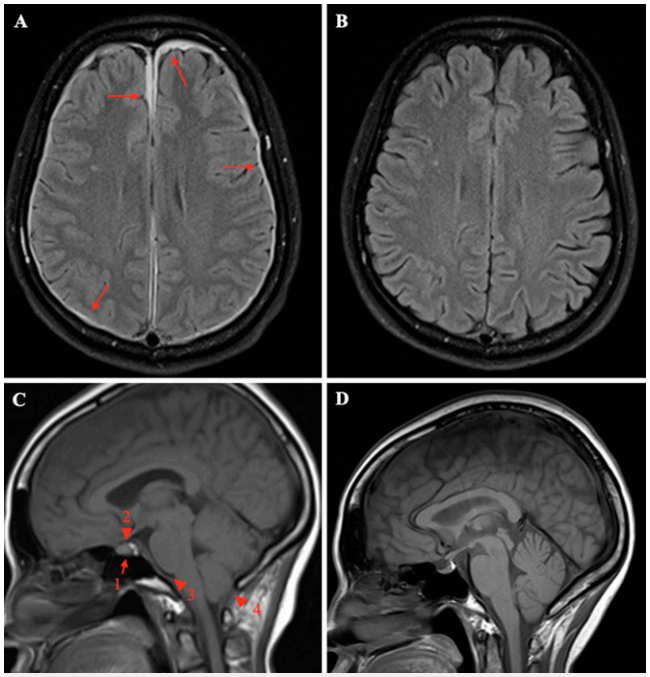

FIG. 4. Pre- and postoperative MRI including axial T1 sequences postcontrast showing pachymeningeal enhancement resulting from SIH (**A**) and its resolution postoperatively (**B**). Sagittal T1 sequences visualizing multiple stigmata of SIH (**C**), such as pituitary hyperemia (*1*), effaced suprasellar cistern (pathologic <4 mm) (*2*), effacement of the prepontine cistern (pathologic <5 mm) (*3*), and low-lying cerebellar tonsils (*4*). Improvement of these signs is seen in the postoperative FU MRI 3 months after surgery (**D**).

Beyond this surgical technique, there are standard treatment options for CVF, including EBP, endovascular embolization, and open surgery.[4,9,14,16] Given the novelty of CVF and the lack of randomized controlled trials, the treatment of choice relies on the physician's preference and expertise or the center's available modalities. As observed in our cohort, some patients do not respond to interventional measures such as EBP or endovascular embolization and remain symptomatic. For these patients, surgical ligation represents the most valuable option. Very few studies reported their outcomes after surgical ligation of CVF. Wang et al.[9] performed open CVF ligation in 20 patients with consecutive reduction of symptoms in all patients and in absence of surgical complications. Another study, by Majeed et al.,[14] demonstrated similarly positive results in a series of 6 patients who underwent open surgical ligation of CVF via clipping. All patients showed resolution of their symptoms by applying the same clipping technique as described in our study. The MIS technique, however, has not been reported so far and represents a new technical variant for this particular indication, which may offer equivalent beneficial results with reduced surgical impact. One patient in our series underwent 2 MIS procedures within 3 months and tolerated them well, underlining the benefit of the reduced invasiveness, the ability to focus on single levels, and the lower risk profile than open procedures. Only 1 study, by Corniola et al.,[17] reported a minimally invasive posterolateral, translaminar, facet-sparing approach for repair of a ventral CSF leak. However, this approach is not comparable to ours, but it again confirms the possibility of MIS procedures for intractable SIH caused by CSF leaks. Taken together, the patient numbers with surgical ligation of CVF remain low and do not allow outcome evaluation beyond their so far descriptively reported efficiency. Further studies with higher patient numbers are required to evaluate the detailed surgical outcome of surgical CVF ligation as well as different surgical techniques.

Another important factor for successful treatment of CVF, independent of the surgical technique, is the preoperative imaging approach. We observed in our cohort that DSM and/or dynamic CT myelography are the diagnostic modalities of choice for defining the location of the CVF. Unlike previously recognized cases of SIH caused by dural tears, CVFs do not necessarily cause pooling of fluid in the epidural space and therefore are more difficult to detect.[7] In recent years, several specialized imaging techniques have been developed, including dynamic DSM, which assesses the epidural space with the patient in different positions. CVFs were detected in 74% via DSM with patients in the decubitus position.[18] Consecutively, decubitus positioning has been adopted as a standard technique in DSM for CVF diagnosis.[7] Nevertheless, CVFs remain difficult to identify and require significant expertise, which is limited to very few centers. In our study, dynamic DSM was performed in all patients and led to successful detection of CVF after a variable number of attempts in 4 of 5 patients, underlining the complexity of definite CVF diagnosis and emphasizing the need for experienced interventional neuroradiologists. The combination or succession of different modalities, such as a post-DSM CT scan, as performed in the above-described female patient, can be helpful. The literature describes the majority of CVFs being located at the thoracic level, which was confirmed in our cohort with all CVFs localized in the thoracic spine.[13,19–21]

TABLE 1. Patient characteristics and clinical information

Case No.	Age at SX (yrs)	Sex	Clinical Presentation	Localization	Previous EBPs (no.)	Previous Embolization (no.)	FU (mos)	Complications	Outcome
1	44	F	Orthostatic HA >2 yrs	T9 left	1	0	25	None	Resolved
2	59	F	Orthostatic HA >3 yrs	T2 left	2	0	31	None	Resolved
3	59	M	Orthostatic HA >1 yr	T9 right	1	0	23	None	Recurrent orthostatic HA
	60	M	Second CVF w/ SIH	T8 right	1	0	18	None	Resolved
4	61	M	Bilateral subdural hematoma	T3 left	2	0	26	None	Resolved
5	62	F	Orthostatic HA >1.5 yrs	T9 left	2	1	8	None	Improved

HA = headaches; SX = surgery.

Summary of patient characteristics, including clinical presentation, interventions and outcome results. Patient 3 underwent 2 MIS procedures for 2 CVFs.

Lessons

We have learned from our case series that MIS ligation of CVFs can be performed safely and successfully. The reduced invasiveness is in favor of the patient's recovery, wound complications, and overall surgical outcome. However, this conclusion is impacted by the limited number of cases. The study includes 5 patients with 6 treated CVFs and represents the experience of 1 neurosurgeon in 1 center, which limits the scientific evidence of the proposed technique as well as the potential to identify complications other than unsuccessful CVF ligation. Also, the reported number of cases is currently too small to allow any comparison between MIS and open techniques. However, given the common application of MIS tubular retractors for many other spinal pathologies, we think that this technique will be an easy adjustment for many surgeons in this particular indication, that it can be applied more frequently in the future, and that it may allow bigger case studies.

Another lesson we took from our case series is the importance of definite and precise CVF localization via preoperative imaging. Without the substantial expertise of the investigating neuroradiologists, their perseverance, and the technical amendments during their interventions, as provided in our cohort, a definite diagnosis would have been missed. This emphasizes the complexity of CVFs and CSF leaks as a pathological condition, and it explains the high number of patients remaining undiagnosed. However, given that CVFs were only first described in 2014 by Schievink et al.,[11] thus representing a recent diagnosis, further studies will increase the general awareness of and expertise in this condition and fill the knowledge gaps about outcome prediction, surgery-related risks, and optimization of variable surgical techniques.

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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: Massicotte, Farb. Acquisition of data: Massicotte, Lohkamp, Marathe, Farb. Analysis and interpretation of data: Massicotte, Lohkamp, Marathe, Farb. Drafting the article: Massicotte, Lohkamp, Marathe, Farb. Critically revising the article: Massicotte, Nicholson, Farb. Reviewed submitted version of manuscript: Massicotte, Nicholson, Farb. Approved the final version of the manuscript on behalf of all authors: Massicotte. Statistical analysis: Massicotte. Administrative/technical/material support: Massicotte, Lohkamp, Marathe. Study supervision: Massicotte.

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