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Conservative Management of Low Back Pain Related to an Unresectable Aggressive Sacral Hemangioma: A Case Report

Authors' Contribution:

Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Patient: Female, 56-year-old
Final Diagnosis: Aggressive sacral hemangioma
Symptoms: Constipation • low back pain • lower extremity weakness
Medication: —
Clinical Procedure: Exercise • manual therapy
Specialty: Neurosurgery • Rehabilitation

Objective: Rare disease
Background: Aggressive sacral hemangiomas are extremely rare benign tumors that can extend into the spinal canal, causing neurologic symptoms. Research on these tumors is limited given their rarity, and they have no agreed-upon treatment strategy. We report the first case of an aggressive sacral hemangioma responding well to conservative care, including manual therapies.

Case Report: A 56-year-old Asian woman presented upon referral to a chiropractic office in Hong Kong with a 6-month exacerbation of chronic low back and lower extremity pain and weakness, requiring the use of a cane to walk. She had a known history of an aggressive sacral hemangioma with neural compromise and had been deemed ineligible for surgery and tumor ablation due to the large hemangioma size and risk of complications. Examination and imaging findings were consistent with compensatory myofascial pain, postural changes, sacral plexopathy, and lower cauda equina involvement. After coordinating with the neurosurgical team, the chiropractor carefully treated the patient with manual therapies, including thoracic spinal manipulation and instrument-assisted soft tissue manipulation, and mobility and strengthening exercises. She responded well, with symptom reduction lasting through 1-year follow-up, and no longer required the use of a cane.

Conclusions: This case illustrates the success of conservative chiropractic care for a patient with an aggressive sacral hemangioma not amenable to other treatments. As further research is needed on this topic, providers considering using manual therapies on patients with an aggressive sacral hemangioma should do so with caution and in collaboration with the patient's medical or surgical team.

Keywords: Cauda Equina • Chiropractic • Hemangioma • Low Back Pain • Manipulation, Chiropractic • Neoplasms • Sacrum

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Background

Aggressive vertebral hemangiomas (VH), also called Enneking Stage 3 VH [1], are extremely rare benign tumors, accounting for only about 1% of cases of VH [2]. Those affecting the sacrum and extending into the spinal canal, such as reported in the current case, are even less common. Aggressive VH are characterized by bone expansion, erosion through the cortex, hematoma, or fracture, and can encroach upon the spinal canal [1,2]. Accordingly, aggressive VH are more likely to cause neurologic symptoms than the more prevalent non-aggressive VH [2].

To the best of our knowledge, and according to a Google Scholar search on April 16, 2022, only 7 cases of aggressive VH of the sacrum have been reported previously [1,3-7], only 5 of which reported extension into the spinal canal, as in the current case [3-7]. Due to the limited evidence and unique features of these lesions, there has not been a consistently agreed-upon or successful treatment strategy [3-7].

Surgical procedures commonly used for VH are either challenging, not possible, or counterproductive in patients with aggressive sacral VH. Concerns regarding potential surgery in these patients include bleeding due to tumor vascularity [5], nerve compression in cases of canal compromise [5], and failure of en bloc resection [7]. One case reported the sacral VH recurred with a larger size following resection [4].

Ablative procedures are also challenging for aggressive sacral VH. Radiotherapy generally has a low success rate for aggressive VH [8]. Sacral VH present an additional difficulty as these tumors are difficult to access [7]. While 1 case reported success with radiotherapy [3], another reported a failure of this treatment [7]. Radiofrequency ablation was likewise only reported to be successful in 1 previous case [7].

Considering the limited research and treatment options regarding aggressive sacral VH, we present a patient with symptoms related to this tumor who was successfully managed via a carefully applied conservative chiropractic approach.

Case Report

Patient Information

A 56-year-old Asian woman presented to a chiropractic clinic with a 6-month exacerbation of low back pain with sharp radiating pain extending into her right buttock and lateral thigh. The patient also described lower extremity weakness which required her to use a cane to ambulate. Even with ambulatory assistance, she could only walk about 50 meters before

needing to stop due to severe bilateral buttock and thigh pain. Her pain severity on the numeric rating scale was 8 out of 10. The patient denied having any loss of bladder control or saddle anesthesia but did endorse constipation. The patient was a non-smoker and worked as an insurance agent. Her family history was negative for neoplastic conditions. Her World Health Organization Quality-of-Life scale (WHOQOL-100) score was recorded as 62%.

Prior to this exacerbation, she had a 10-year history of mild low back pain and was diagnosed 16 months prior with an aggressive sacral VH by positron emission tomography/magnetic resonance imaging (PET/MRI), contrast MRI, and computed tomography-guided biopsy (Figure 1). Her neurosurgical team decided to avoid attempting any operation or procedure given the large VH size, low probability of success, and risk of complications. Previously, the patient tried rest, exercise, oral nonsteroidal anti-inflammatory drugs (etoricoxib and celecoxib), and underwent a right sacroiliac joint steroid injection, all without relief. As options were limited for the patient, her neurosurgical team referred her for chiropractic care. The patient had no previous history of manual therapy treatments.

Review of her most recent MRI from October 2019 revealed a large contrast-enhancing mass extending from the S1 to S3 sacral segments (Figure 2). It was measured in centimeters (cm) at 5.6×9.8×6.6 cm (anterior-posterior×transverse×sagittal) and compressed and completely obliterated the sacral spinal canal from S2 to S3, causing compression of the lower cauda equina, and enclosed the S1 and S2 sacral foramina bilaterally. The aggressive VH had slightly increased in size in comparison to her previous positron emission tomography (PET) MRI from November 2018, in which it was 4.6×8.7×4.6 cm. No significant neuroforaminal or canal stenosis or other VH were noted in the lumbar spine visible in the field of view (L2-L5).

Clinical Findings

In March 2020 the patient presented to a chiropractor in Hong Kong for evaluation. On examination, the patient had a flattened back posture and antalgic lean to the left side. On palpation the patient had tenderness bilaterally in the paraspinal areas from L1 through L5. Spinal palpation also revealed motion restriction and tenderness at the T3-4, T7-T8, and T12-L1 levels. Hypertonicity was noted at the iliopsoas, rectus femoris, and erector spinae bilaterally. Her active lumbar spine range of motion was restricted and uncomfortable at 10° extension (normal is 20-35°). Straight leg raising did not exacerbate symptoms, but the femoral nerve stretch test reproduced her symptoms when performed on each side. Motor evaluation revealed 4/5 strength (Medical Research Council Scale) of right knee extension with pain.

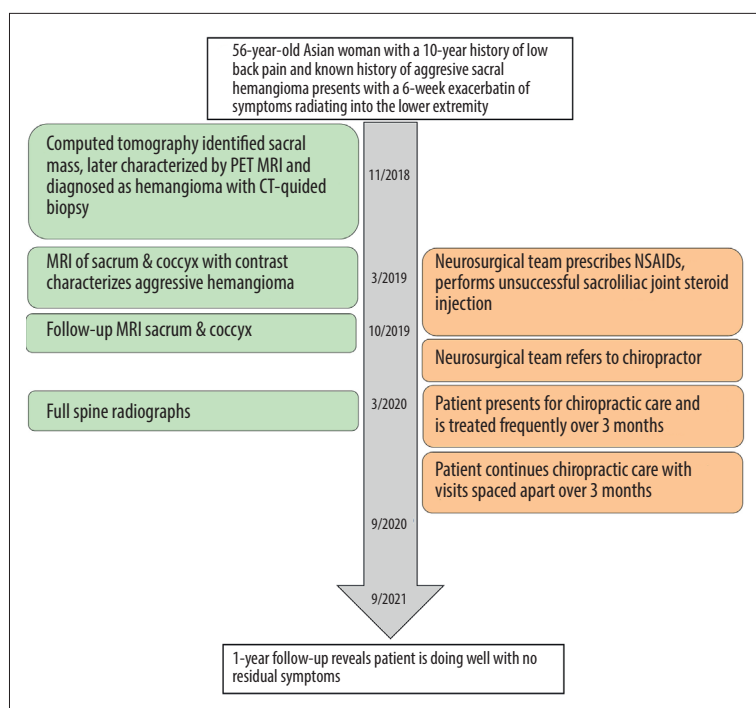


Figure 1. Timeline of care. Left – imaging studies. Right – clinical decision making and interventions. Abbreviations: Computed tomography (CT), magnetic resonance imaging (MRI), positron emission tomography (PET).

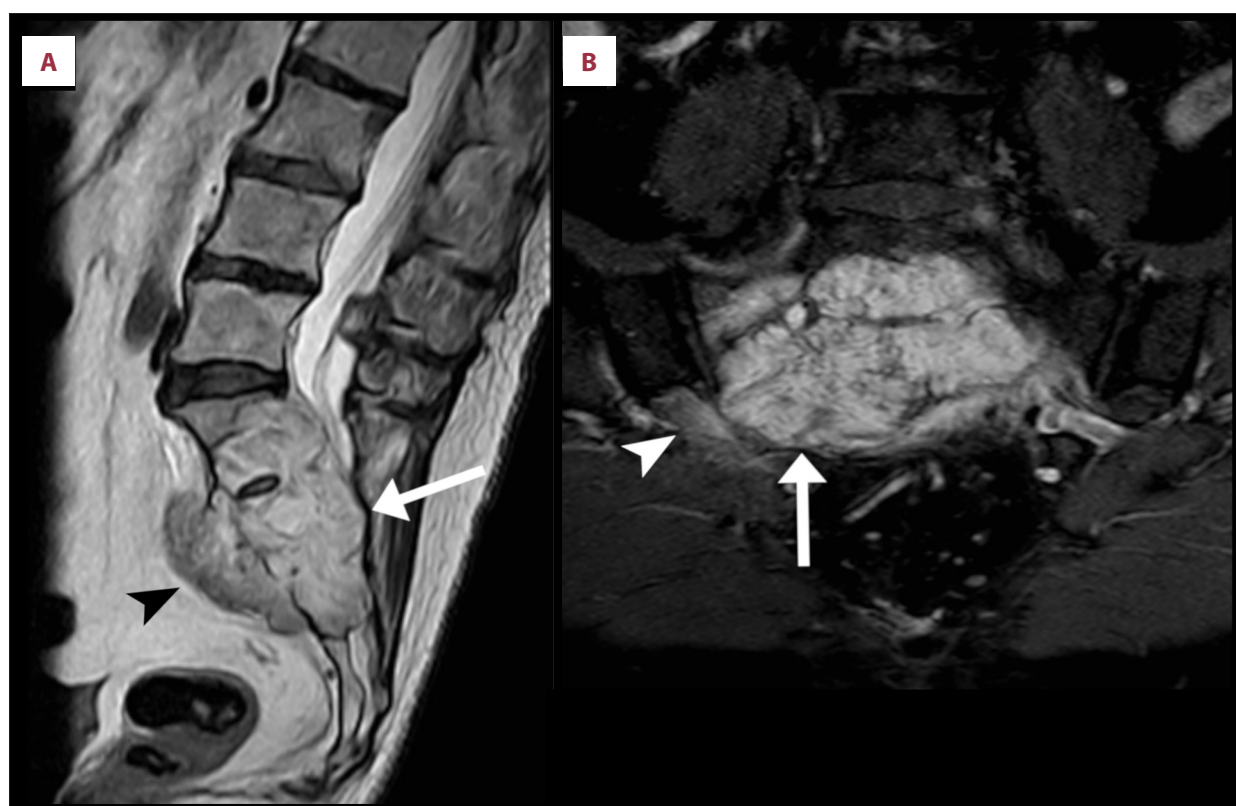


Figure 2. Sacral MRI (2019). The sagittal T2-weighted view (A) reveals a large, lobulated mass (arrow) which is heterogeneously hyperintense and obliterates the sacral spinal canal. The mass has a presacral component (arrowhead). The coronal T1-weighted, post-gadolinium view oriented to the anterior surface of the sacrum (B) reveals that the mass is contrast-enhancing and its presacral extension is more prominent on the right side (arrow). The mass also extends to the iliac side of the right sacroiliac joint (arrowhead).



Figure 3. Full-spine radiographs. The anteroposterior view (**A**) shows an abnormal coronal balance with the patient's spine deviating to the left of the central sacral vertebral line (the vertical line bisecting the first sacral segment and perpendicular to the iliac crests). The aggressive sacral hemangioma (*) is visible spanning the sacrum, more prominently on the patient's right side. There is no appreciable scoliotic curvature. The lateral view (**B**) shows the lumbar lordosis angle (θ) of 7° calculated using a 4-line L1-L5 Cobb method (normal is $20-45^\circ$). Thoracic hypokyphosis (flattening) is also evident, while the patient's C7 plumb line intersected the sacrum, indicating a neutral sagittal balance (measurements not shown). Mild spondylosis is apparent in the lower thoracic and lumbar spine.

Full-spine radiographic examination was conducted due to postural signs of potential scoliosis, history of failure of other forms of care, history of aggressive sacral VH potentially leading to spinal deformity, and absence of previous full-spine imaging. An overall assessment of spinal balance was valuable in this case to guide chiropractic treatment, which was more comprehensive rather than focused on the site of the VH. Radiographs revealed degenerative changes in the thoracic and lumbar regions, pelvic imbalance, abnormally decreased thoracic and lumbar curves, and coronal imbalance (**Figure 3**). The sacral VH was also visible, yet no other VH were evident in the remainder of the spine.

The chiropractor formulated a differential diagnosis including myofascial pain syndrome, which was thought to be secondary to postural changes, pain referral, and guarding related to the aggressive sacral VH. Due to the extension intolerance, femoral nerve stretch pain (possible L4 sign), and lateral thigh pain (possible L5 sign), a diagnosis of lumbosacral plexopathy was also considered. This could result from compression of the lumbosacral trunk (which derives from the L4 and L5 spinal

nerves) by the presacral extension of the sacral VH, which abutted the overlying plexus. Compression of the lower cauda equina was also considered as contributing to symptoms.

The patient consented to a trial of conservative chiropractic treatment including thoracic spinal manipulation using a high-velocity, low-amplitude force, and manual contact at T3-4 and T7-T8 to improve spinal mobility (**Figure 4**). Lumbar and pelvic manipulation was avoided after discussion with the patient's spine surgeons, to avoid iatrogenic fracture of the VH. The patient was treated daily for the first week, during which time her pain severity reduced to 3 out of 10. The initial high visit frequency was recommended given the patient's severe pain, weakness, limited mobility, and an initial positive but transient response to care. Visits were gradually spaced apart as she obtained longer-lasting relief with treatments.

Subsequently, treatment sessions were provided twice per week for 3 months, then reduced to once per week for an additional 3 months. After the 1st week of care, therapy sessions also included instrument-assisted soft tissue mobilization



Figure 4. Demonstration of thoracic spine high-velocity, low-amplitude manipulation. An impulse is delivered by the chiropractor to the mid-back along areas of restriction and tenderness identified by palpation, with the vector being oriented posterior to anterior and slightly superior. In the current case this technique was avoided in the vicinity of the sacrum as a precaution.



Figure 5. Demonstration of instrument-assisted soft tissue mobilization. A massage tool (Strig, Korea) is gently stroked across the skin surface with a thin layer of emollient. This produces petechiae (redness), which is benign and self-resolving.

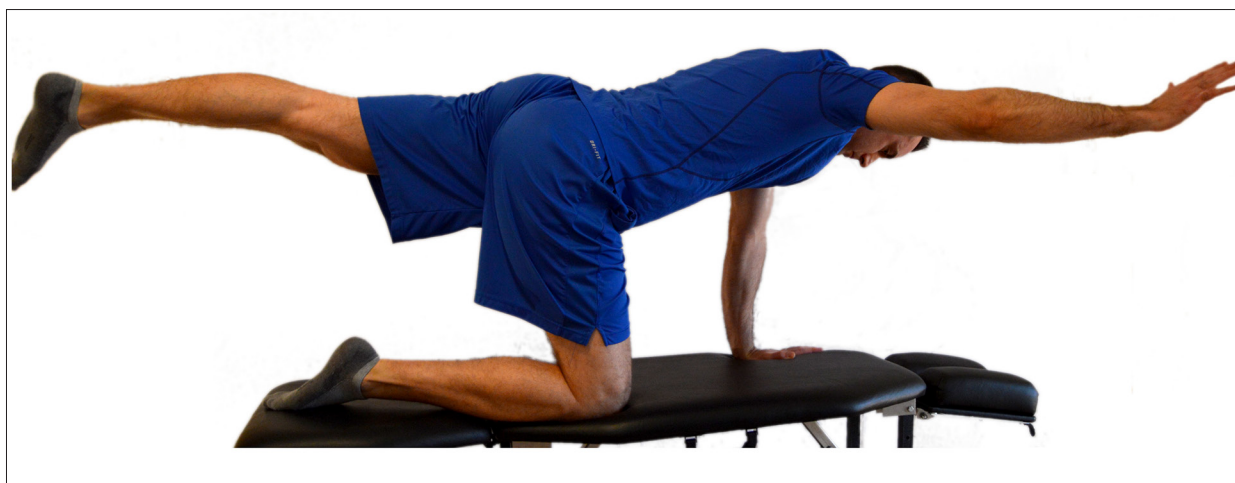


Figure 6. Demonstration of the “bird-dog” exercise. The patient lifts one leg and one arm on the opposite sides of the body, then alternates to lifting the opposite limbs in a repeating manner. Image reproduced with permission from Robert J. Trager, from *Sciatica: Foundations of diagnosis and conservative treatment* [9].

(Massage instrument, Strig, Korea; **Figure 5**) to alleviate hypertonic lumbar muscles. The chiropractor also taught the patient low-impact rehabilitative strengthening and stretching techniques performed in the quadruped position (ie, on the hands and knees). This included the cat-cow exercise, which involves alternating between spinal flexion and extension to improve spinal mobility, and bird-dog exercise, which aims to strengthen and stabilize the low back (**Figure 6**) [9]. During

the final 3 months of treatment, the patient gradually reduced then stopped taking her pain medication.

By the end of the 6-month period of treatment, the patient was able to resume normal daily activities such as grocery shopping without experiencing any residual symptoms. At her a 1-year follow-up visit, the patient reported being fully symptom-free for the previous 6 months and no longer used a cane

to ambulate. Her 1-year follow-up WHOQOL-100 score was recorded as 98%. The patient was not under the care of other providers for her aggressive VH during the course of chiropractic care. Written informed consent was obtained from the patient to publish her case report and any accompanying images.

Discussion

This case is the first report of a patient with a symptomatic aggressive sacral VH that responded well to non-invasive conservative treatment. Among previous cases of aggressive sacral VH, the outcome was either not reported [1,5,6], resulted in a failed surgical intervention and VH recurrence [4], or displayed mixed results with an ablative procedure [3,7]. In the present patient, the risks of invasive interventions were high, and the patient was referred for chiropractic care.

Although spinal manipulative therapy is supported by high-level evidence as a treatment for chronic low back pain [10], it has seldom been described in the context of patients with neoplasms [11]. Given potentially complex patient presentations such as the current case, spinal manipulation should be applied by a licensed, trained provider such as a chiropractor to maximize safety. To avoid pathologic fracture, spinal manipulation should not be applied in the region of bone weakened by neoplasm [12].

Spinal manipulation can provide relief by providing a mechanoreceptive stimulus that inhibits pain, relaxes hypertonic muscle, and influences posture [13]. In addition, recent evidence suggests spinal manipulation can produce clinical benefits at regions of the spine away from the area of application [14]. These mechanisms could explain the low back and lower extremity pain relief afforded by thoracic spinal manipulation in the present case. Instrument-assisted soft tissue mobilization was used in this patient to further inhibit pain [15], while rehabilitative exercises were used to improve strength, gait, and posture.

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Limitations

As a case report, these results may not be broadly generalizable. Neither digital rectal examination nor intrinsic foot muscle strength were evaluated; thus, further signs of sacrococcygeal nerve impairment may not have been identified. Electrodiagnostic testing was not conducted thus the exact neuroanatomical source(s) of symptoms were less clear. The patient could have improved with other forms of conservative care such as acupuncture or physical therapy. An ablative procedure could have been helpful but was not attempted. Follow-up spine imaging was not conducted and was deemed unnecessary given patient improvement. Although the outcome with care was positive, the patient remains at risk for pathologic VH fracture and eventually could require surgery on an emergency basis.

Conclusions

As a result of the rarity of aggressive sacral VH and challenges related to surgical and ablative procedures, there is no consensus regarding appropriate treatment, and patients are managed on a case-by-case basis. The present case report highlights a patient with aggressive, symptomatic sacral VH that was not amenable to invasive procedures but responded positively to a conservative course of chiropractic care. Conservative care such as chiropractic could be considered for use in patients with aggressive sacral VH, particularly when other approaches are not feasible. However, as further research on this subject is needed, such manual therapy approaches should be used with caution and in collaboration with the patient's surgical team.

Department and Institution Where Work Was Done

New Chiropractic and Physiotherapy Centre, EC Healthcare, Hong Kong.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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