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Stereotactic radiosurgery versus surgical resection for spinal hemangioblastoma: A systematic review

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ABSTRACT

Spinal cord hemangioblastomas are benign vascular tumors arising sporadically in approximately 70–80% of cases. They can also be manifestations of von Hippel-Lindau (VHL) disease, as these patients will often have multiple spinal hemangioblastomas. Historically, surgical management of symptomatic intramedullary hemangioblastomas has been considered the treatment of choice. However, recently, stereotactic radiosurgery has been utilized as an adjuvant therapeutic modality, and some have suggested it may have utility as the primary treatment option for these tumors. Because of the rarity of spinal hemangioblastomas, management options, clinical outcomes, and prognostic factors have not yet been fully elucidated. The National Institutes of Health (PubMed) was queried to identify all studies describing treatment of spinal hemangioblastomas. Focus was narrowed to institutional retrospective reviews, and comparisons were drawn regarding outcomes of both stereotactic radiosurgery and surgical resection. Stereotactic radiosurgery achieves stable or reduced tumor size with relatively little adverse clinical outcome long-term. Meanwhile, surgical resection results in successful removal of the tumor with approximately 96% stable or improved long-term clinical effect. Cross-platform analysis has been challenging when comparing efficacy amongst treatment modalities for this rare tumor. For the institutional retrospective reviews that exist, researchers tend to collect and record data in a multitude of fashions, making direct comparisons problematic. As such, the authors propose use of a national registry to input data prospectively about spinal cord hemangioblastomas.

Contents

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1. Introduction

Spinal cord hemangioblastomas are benign vascular tumors accounting for 3% of central nervous system tumors, and comprising 2–6% of all tumors within the spinal cord [1–3,41]. Hemangioblastomas arise sporadically in approximately 70–80% of cases, however 20–30% of these lesions are manifestations of von Hippel-Lindau (VHL) disease, a heritable multisystem cancer syndrome [1,3–5,39,40,42]. Hemangioblastomas are the most common lesions associated with VHL, and patients with this disease will frequently have multiple spinal cord hemangioblastomas with risk of developing additional tumors throughout their lifetime [2,6].

Although histologically benign, intramedullary hemangioblastomas can result in significant neurological symptoms as a result of their size, location, and venous congestion with associated peritumoral edema [3,7,8]. More than 50% of these spinal cord tumors have accompanying syringomyelia, which may also contribute to neurological sequelae [9].

Surgical management of symptomatic intramedullary hemangioblastomas has been considered highly effective and is the treatment of choice [5,6,8,10–23]. However, surgical resection can carry high risk with potential for poor prognosis, especially in the upper cervical spinal cord [7]. More recently, stereotactic radiosurgery has served as a therapeutic adjuvant, optimizing tumor dose while sparing the spinal cord [24,25]. It provides a noninvasive alternative to surgery and has been increasingly utilized in primary management of central nervous system hemangioblastomas [26]. Conversely, radiation-induced myelopathy has been reported in patients undergoing stereotactic radiosurgery to the spine with potential permanent neurological detriment [27–31].

Because of the rarity of spinal hemangioblastomas, management tactics, clinical outcomes, and risk factors affecting prognosis have not yet been fully realized. The aim of the present study was to review the outcomes of both radiosurgery and surgical resection of spinal cord hemangioblastomas. By performing cross-platform analysis and comparing these two groups, our goal was to provide a means in which the medical community can determine the safest, most efficacious treatment options for spinal hemangioblastomas.

2. Methods

2.1. Data source

The National Institutes of Health (PubMed) was queried to identify all studies describing treatment of spinal hemangioblastomas. Initial search terms included, “spine,” “spinal,” “hemangioblastoma,” “benign,” “tumor,” “radiation,” “radiosurgery,” “surgery,” and “surgical.” Article reference lists were additionally utilized to identify other pertinent studies. Criteria for final selection included a demonstration of qualitative and quantitative data specifically regarding spinal hemangioblastoma treated either by stereotactic radiosurgery or surgical resection. Focus was narrowed to institutional retrospective reviews of their own data. When data combined all benign spinal tumors, including low grade astrocytomas and ependymomas, this information was excluded unless detailed data could be isolated specifically for hemangioblastoma. Case reports were excluded. Ultimately, our analysis consisted of 4 articles assessing radiosurgical treatment of spinal hemangioblastoma, and 10 articles assessing surgical resection of a total of 538 tumors. However, all radiosurgery manuscripts were from a single institution, and, known via personal correspondence, there was some overlap in patients.

3. Results

3.1. Stereotactic radiosurgery

Our literature review yielded 4 manuscripts in which authors performed retrospective institutional reviews of stereotactic radiosurgery specifically for spinal hemangioblastomas, (Table 1) [6,29,32,33]. However, all 4 reports were from a single institution, and some patient overlap exists (personal communication). No statistics can therefore be calculated for all patients and tumors presented. Rather, data has been calculated separately for each of the individual studies. Averages of each study were calculated, after which overall averages were determined. All treatment utilized the CyberKnife system (Accuray Incorporated, Sunnyvale, California, United States of America). Authors did not routinely include maximum and average tumor sizes nor whether the locations were intramedullary, extramedullary, or combined. When tumor location was reported, an average of 65% involved the cervical spine, 32% involved the thoracic spine, and 3% involved the lumbar spine. Treatment plans typically included an average dose of 21 Gy over 1–3 fractions. Of those reported, an average of 56% of tumors reduced in size, 42% remained stable, and 2% progressed at the time of follow up, which ranged from 1 to 3 years. While three studies reported no complications, the largest study (27 tumors treated) identified 3 (11%) complications [29]. One patient developed unilateral foot drop 5 months after radiosurgery, and two patients had “sensory deficits,” although further details were not reported. In a study involving 16 tumors treated, post-radiation edema was seen in two C7 lesions; however, no clinical radiation toxicity was appreciated [6].

3.2. Surgical resection

Our literature review yielded 10 studies in which authors performed retrospective institutional reviews of surgical resection specifically for spinal hemangioblastomas, totaling 538 tumors (Table 2) [1–3,5,7,9,16,34–36]. However, given the variability in data presented in each study, there exists a different number of tumors or patients for each variable assessed. As such, this resulted in differing denominators for each data point reviewed.

3.3. Tumor characteristics and extent of resection

When tumor location was reported, 50% (227/457) were in the cervical spine, 42% (192/457) were in the thoracic spine, and 8% (38/457) were in the lumbar spine. Most authors did not clearly identify if the tumor was intramedullary, extramedullary, or a combination of the two. When specified, involvement was intramedullary in 33% (119/361) cases, extramedullary in 30.7% (104/339) cases, and combined in 40% (137/339) cases. Of note, one study did not specify whether non-intramedullary tumors were extramedullary or combined. This study was excluded when comparing extramedullary and combined values. This resulted in varied denominators and subsequent percentages.

Total resections were achieved in 92% (493/538) of tumors. Subtotal resections were often attributed to intraoperative bleeding, while other causes were loss of somatosensory evoked