Clinical Neurology and Neurosurgery 142 (2016) 93–97

Contents lists available at ScienceDirect

Clinical Neurology and Neurosurgery

journal homepage: www.elsevier.com/locate/clineuro

Ossified spinal meningiomas: Clinical and surgical features

Concetta Alafaci[a,*], Giovanni Grasso[b], Francesca Granata[c], Francesco M. Salpietro[a], Francesco Tomasello[a]

[a] Department of Neurosurgery, University of Messina, Italy
[b] Neurosurgical Clinic, Department of Experimental Biomedicine and Clinical Neurosciences, University of Palermo, School of Medicine, Palermo, Italy
[c] Department of Neuroradiology, University of Messina, Italy

ARTICLE INFO

Article history:
Received 21 October 2015
Received in revised form 8 January 2016
Accepted 20 January 2016
Available online 22 January 2016

Keywords:
Ossified spinal tumor
Metaplastic meningioma
Gross total resection

ABSTRACT

Object: Meningiomas constitute 25% of primary spinal tumors and predominantly involve the thoracic spinal cord. Although calcifications are commonly seen in intracranial meningiomas, gross calcifications are observed in only 1–5% of all spinal meningiomas. We report the clinical findings, surgical strategy and histological features of 9 patients with ossified spinal meningiomas (OSMs).
Patients and methods: Clinical and surgical features of 9 patients with ossified spinal meningiomas were retrospectively reviewed.
Results: There were 8 women and 1 man with a mean age of 59 years. In 7 patients, the lesions were localized in the thoracic segment of the spine while in 2 patients in the lower cervical segment. All patients presented with weakness of the lower limbs and hypoesthesia below the site level of the lesion. Only 2 patients presented with urinary incontinence. Gross-total resection of the tumor was achieved in 6 patients while in 3 a subtotal removal of the meningioma was obtained. In all patients the postoperative course was uneventful. Six patients presented with a significant neurological improvement while in 3 patients a mild improvement was observed. Microscopically, all tumors showed typical histological pattern of ossified meningioma.
Conclusions: OSMs are amenable to surgery if the complete removal can be achieved. Because of their hard-rock consistency complete resection can be challenging. In difficult cases, subtotal removal can be advised and follow-up imaging is mandatory. Overall, the risk of long-term recurrence of the lesions is low, and a good clinical outcome after total or subtotal removal can be expected.

1. Introduction

Meningiomas constitute 25% of primary spinal tumors and predominantly involve the thoracic spinal cord [1]. Ossified spinal meningiomas (OSMs) are a specific subtype of meningiomas since the ossification is secondary to the metaplasia of the arachnoid cells [2]. Classic calcifications often found in meningioma are common features of psammomatous features which leads to progressive accumulation of hydroxyapatite crystals. OSM is characterized by the presence of a variable number of bone spicules within the tumor itself with a clear phenotype of metaplastic meningioma [2]. OSMs are mostly intradural extramedullary in location, and the diagnosis of the tumors is usually not difficult based on radiological findings. To our knowledge, only 13 cases of ossified spinal meningiomas have been described since the publication of the earliest report in 1928 [2–12]. In our search we excluded reports where not clear differentiation between ossification and calcification was provided.

Surgical removal may present difficulties since adhesion of the tumor to the surrounding tissues complicates its dissection and may affect the surgical outcome.

This study presents the clinical and surgical results from the largest surgical series of 9 patients with OSM.

2. Patients and methods

2.1. Patient population

Between the years 1998 and 2010, OSMs were pathologically diagnosed in 9 patients. Patients were included in the study based on the postoperative pathological diagnosis of ossified meningioma. According to the WHO classification, ossified meningiomas are histologically classified as a subtype of metaplastic meningioma, which is characterized by the presence of mesenchymal

* Corresponding author at: Department of Neurosurgery, University of Messina, Via Consolare Valeria, Messina 98125, Italy. Fax: +39 090 693714.
E-mail address: calafaci@unime.it (C. Alafaci).

http://dx.doi.org/10.1016/j.clineuro.2016.01.026
0303-8467/

components [13]. Data related to the clinical presentation, radiological imaging, treatment, and follow-up were collected and properly reviewed. Magnetic resonance imaging (MRI) with contrast enhancement was performed as standard neuroradiological investigation before and after treatment. All patients underwent surgical resection with intraoperative neurophysiological monitoring of somatosensory and motor evoked potentials.

2.2. Outcome assessment

Neurological function was assessed before surgery, at discharge, postoperatively at 6 and 12 months following surgery and every year thereafter. Modified Japanese Orthopedic Association (JOA) scores were applied to assess neurological function [14]. Differences were evaluated by the Mann–Whitney U test. $p < 0.05$ was considered as statistically significant.

3. Results

3.1. Patient characteristics

There were 9 patients with histologically proven OSMs included in this study. Table 1 depicts the characteristics of OSMs reviewed in our series.

Eight patients (89%) were women and 1 patient was a man (11%). The mean age at the time of the operation was 59 years, ranging between 40 and 86 years. The mean duration of preoperative symptoms was 14.0 ± 22.0 months (range 1–76 months). Neurological findings were related to the tumor localization. The preoperative presentations included motor deficits associated with sensory disturbance in all the cases (100%). Only 2 patients (22%) presented with sphincter dysfunction. The mean preoperative JOA score was 11.4 ± 2.2.

3.2. Tumor features

In 7 patients (78%) the tumor was localized in the thoracic segment while in 2 patients (22%) in the lower cervical segment. OSM was ventral to the spinal cord in 2 cases (22.2%), in ventro-lateral position in 2 cases (22.2%), pure lateral in 1 case (11.1%) and dorsal in 4 cases (44.4%).

Radiological diagnosis indicated the presence of intradural extramedullary spinal cord tumors. On MRI most of the lesions were hypointense to spinal cord on T1- and T2-weighted sequences with partial enhancement following gadolinium administration.

Upon the diagnosis, all the patients underwent surgical treatment. In all the cases, a posterior approach was performed and almost two levels laminectomy were carried out. After opening the dura, the lesions appeared yellowish or red-discolored in color, rock-hard in consistency and firmly adherent to the surrounding dura. An arachnoid layer separating the lesion from the spinal cord was present in 6 cases. In 3 cases the lesion was strictly adherent to the nearby spinal cord with poor tumor–arachnoid interfaces. In these cases the tumor was rather difficult to resect without unacceptable surgical complications. Thus, gross-total resection (GTR) of the tumor was achieved in only 6 cases (66.6%) with a well-demarcated dissection plane (Simpson 2), and subtotal resection (STR) was achieved in 3 cases (33%) (Simpson 3). For preventing recurrence, the tumor dural attachment was coagulated. An illustrative case is shown in Fig. 1. Histological examination showed in 7 cases osseous component in association with psammoma bodies and dystrophic calcifications. In 2 cases, in which no psammoma bodies or calcifications were found, foci with immature bone trabeculae, mineralized chondroid matrix, and osteoclasts were observed suggesting a pattern of enchondral ossification. Mitotic figures were rarely found, and the Ki-67 labeling index in all the lesions was less than 1%.

3.3. Outcome

The postoperative course was uneventful in all the cases. Immediately after surgery, 6 patients presented with a significant improvement of both paraparesis and hypoesthesia, while in 3 patients receiving a subtotal removal, there was a mild improvement of the motor deficits. The mean follow-up period was 36.8 ± 22.3 months (range 24–72 months). The mean postoperative JOA score was 15.1 ± 1.2 at last follow-up. The mean final JOA score for all patients was significantly improved over the preoperative JOA score ($p < 0.05$).

Postoperative MRI did not show tumor recurrence in the 6 GTR cases after an average follow-up period of 60 months. At the same period follow-up, in the 3 STR cases, no regrowth in the residual tumors was observed.

4. Discussion

The incidence of intradural tumors in the spine ranges from 3 to 10 per 100000 persons per year, and intradural extramedullary tumors account for 2/3 of all intraspinal neoplasms [15]. Among these, spinal meningiomas account for approximately 25–46% of all spinal cord tumors [1]. Spinal meningioma grows from the arachnoid cells that differentiate from neural crest cells. Although spinal meningiomas are not uncommon, totally calcified spinal meningioma is very rare in spinal location [1]. To date, only 13 cases of OSMs have been described with the first case reported in 1928 [2–12] (Table 2). Unlike the commonly encountered meningioma that develops into a variable interface in the small space confined between the arachnoid and the intermediate leptomeningeal space, the OSM presents with extensive matrix and tends to infiltrate the surrounding structures. It exhibits a growth pattern limited by an incompletely developed intermediate leptomeningeal layer. Since the intraspinal spaces are much smaller than intracranial spaces, symptoms usually appear rapidly compared with intracranial tumors.

The surgical series presented in this study included 9 patients with histologically proven ossified spinal meningioma. This is the largest series reported being almost single cases separately described in the current literature. In our series there were 8 female and 1 male patients with a mean age of 59 years. OSMs affected mainly women than men with a peak incidence during the fifth to sixth decade of life. OSMs are most frequently found in the thoracic spine [2–12]. Also in our series in 7 patients (78%) the tumor was localized in the thoracic segment while in 2 patients (22%) in the lower cervical segment. OSM was anterior to the spinal cord in 2 cases (22.2%), in ventro-lateral position in 2 cases (22.2%), pure lateral in 1 case (11.1%) and dorsal in 4 cases.

Similarly to the other intradural extramedullary tumors, the clinical features of OSMs include motor, sensory, or sphincter dysfunctions, which appear differently according to the tumor location and entity of tumor compression. In our series, the mean duration of preoperative symptoms was 14.0 ± 22.0 months (range 1–76 months) thus reflecting the slow pattern of tumor growth. The preoperative presentations included motor deficits associated with sensory disturbance in all the cases (100%). Only 2 patients (22%) presented with sphincter dysfunction. The mean preoperative JOA score was 11.4 ± 2.2.

Ossified meningiomas are a rare variant of meningiomas characterized by the presence of a variable number of bone spicules. As we found in our series, they are often characterized by foci with immature bone trabeculae, mineralized chondroid matrix,