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Review

Surgical limitations in convexity meningiomas en-plaque: Is radical resection necessary?

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ABSTRACT

Meningiomas-en-plaque (MEP) comprise 2.5% of all meningiomas. While they typically arise in the sphenoid wing, convexity MEP are comparatively rare and are often confused with meningeal sarcoidosis, osteoma, tuberculoma, or fibrous dysplasia, with very little information published in the literature. We conducted a literature review on PubMed of English-only articles using a keyword search. All studies that described reports of convexity MEP were reviewed for patient demographics, presenting symptoms, radiological reports, surgical management, recurrence rates, histopathological presentation, post-operative complications, and follow-up. This resulted in 12 papers comprising 22 cases of convexity MEP. Seventeen (77%) of the 22 patients were female with an average age of 53.2 years. Intitial presenting symptoms included headache in 12/20 (60%), hemiparesis in 5/20 (25%), and visual symptoms in 1/20 (5%). Of the 14 patients who underwent surgical resection, only four were reported as gross total resection. Twelve reports had associated pathology reports, with all 12 tumors graded as World Health Organization Grade I. Convexity MEP, while rare, present a challenge with regard to correct diagnosis and subsequent resection. The easier accessibility of these meningiomas predicts higher surgical success rates and incidence of total resection, though care must be taken to ensure gross total removal of tumor, dural attachments, and any overlying hyperostotic bone. Though hyperostosis is frequently observed with this variant of meningioma, it is neither exclusive nor wholly indicative of MEP. Due to its rarity in both clinical practice and the literature, further studies are warranted to identify modern imaging means to correctly diagnose this condition.

1. Introduction

Meningiomas, the most frequently diagnosed type of brain tumor, account for over one-third of primary brain and central nervous system tumors reported in the USA between 2007–2011 [1]. The morphological variant termed meningioma-en-plaque (MEP), first coined by Cushing in 1922 [2], is a rare but distinct tumor characterized by flat, carpet-like proliferation along the dura, in contrast to the more common meningiomas "en masse" that tend to occur as intracranial masses attached securely to the dura [3]. MEP, which comprises 2.5% of all meningiomas [4], typically arises in the spheno-orbital regions, whereas convexity MEP is comparatively rarer [3] and is observed with extensive hyperostosis of the skull (Fig. 1). [2,3] This hyperostosis was once thought to be characteristic of MEP, but it often bears a confusing similarity to the clinical presentations of other pathological conditions such as meningeal sarcoidosis, osteoma, tuberculoma, and fibrous dysplasia [5,6]. Because the current literature varies considerably in study design, depth of analysis, and methodology, a more robust and reliable criteria for diagnosis is needed.

The main treatment options for convexity MEP include surgical resection and radiotherapy. While the previous understanding over the past two decades was that total surgical resection of convexity MEP is much more feasible and successful than MEP of other regions due to the accessibility of the tumor, [3] this assumption has been poorly documented in the literature. Recently published literature is limited to rudimentary case studies with minimal detail of the surgical techniques and little to no follow-up on patient outcomes. Many of the published case reports describe scenarios in which convexity MEP was only diagnosed following surgery and histological confirmation. The lack of quality histological analysis in the literature, as well as sizable discrepancies in pathology grading scales, limits the current understanding of convexity MEP and poses a barrier to proper treatment planning, counseling, and surgical management. The etiology of the associated hyperostosis and the extent that the hyperostotic bone should be considered part of the neoplasm is widely debated, and to our knowledge there is no conclusive reasoning currently available.

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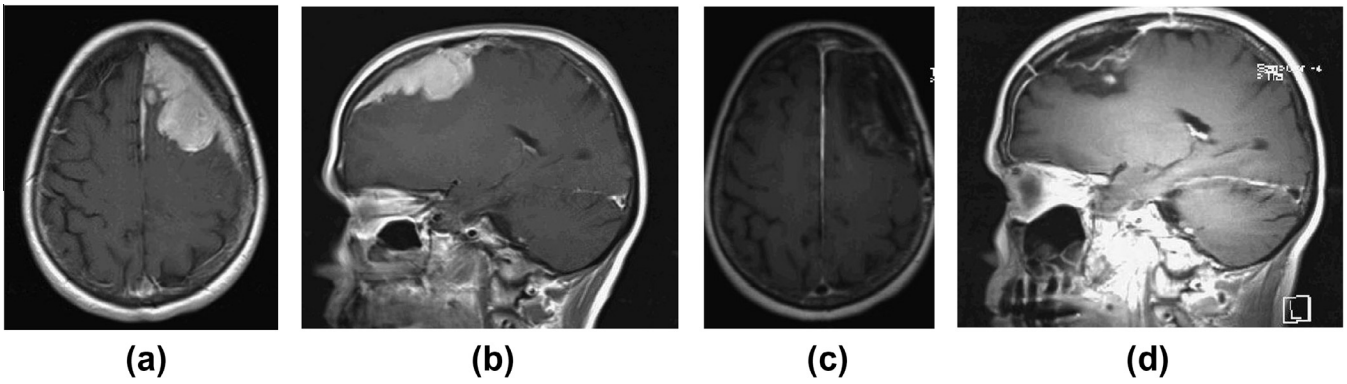

Fig. 1. Pre-operative (a) axial and (b) sagittal T1-weighted MRI with contrast and postoperative (c) axial and (d) sagittal T1-weighted MRI with contrast of a patient with a convexity meningioma en-plaque.

The likelihood of total surgical resection is therefore unclear, necessitating a summary of all results from the included case reports of convexity MEP and a detailed analysis of their differential diagnoses, methods, surgical techniques, and short- and long-term outcomes. Recent studies have proposed new mechanisms for the hyperostosis associated with convexity MEP and its associated implications for currently available treatment options.

This systematic review aims to summarize the body of current evidence and review the histopathological analysis and management of convexity MEP.

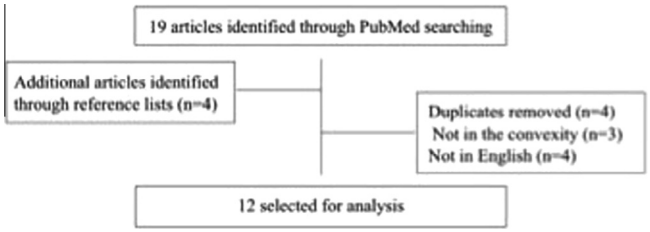

Fig. 2. Flow chart describing selection process for relevant studies.

2. Methods

2.1. Literature search

A literature search of the PubMed database was conducted for relevant clinical studies published since its inception. Only English publications were eligible for inclusion. The key words used were a combination of "convexity", "meningioma en plaque", "parasagittal", and "cranial vault". The date of the last search was 29 March 2015. The inclusion criteria were case reports or case series that described diagnosis, surgical techniques, and clinical outcomes following resection of convexity MEP, as well as studies describing grading score systems and outcome predictors. Reference lists of relevant articles were reviewed to identify additional studies and case reports.

2.2. Data collection and statistical analysis

All studies that described case reports of convexity MEP were reviewed. The case reports were reviewed for study characteristics (design, sample size, study period), patient demographics (age, sex, presenting symptoms, tumor location), and, when present, World Health Organization (WHO) tumor grade, surgical techniques, recurrence rates, complications, histopathological analysis, and follow-up treatment (for example, post-operative radiotherapy). CT scans and MRI, when available, were reviewed to confirm that each case report was indeed convexity MEP. In studies that described MEP of various regions, only pure convexity cases were included in the review. All studies were graded using the Oxford's Center for Evidence-Based Medicine (OCEBM) Levels of Evidence.

3. Results

Figure 2 demonstrates a flow chart outlining the selection process for relevant studies. The literature search retrieved 19 articles, 12 of which were eligible to be included in this review. Studies were conducted in the USA, India, France, Thailand, Turkey, Israel, and Japan. All were determined to be OCEBM Level 4 studies (case reports or series).

Twelve case reports or series were reviewed, resulting in 22 patients. Table 1 summarizes the extent of analysis for each of the 12 papers, as well as basic demographic information. Years of publication ranged from 1972 to 2013. Seventeen of the 22 patients were women. Patients ranged in age from 32–90 with a mean age of 53.2 years. Table 2 outlines each case of MEP described in the papers based on patient demographics, tumor size, location, pathology, and the extent of resection.

The scope and depth of each paper varied widely in their descriptions of histopathological analysis, surgical procedures, and follow-up, as described in Table 1. Though the majority of the papers reviewed were case reports or series, two studies focused on techniques for identification and post-operative management of MEP with little detail on the tumors themselves [7,8]. All but one of the studies described indications for surgery, which ranged from various clinical symptoms to evidence of trauma. Eight of the 12 studies described histological analysis of the excised tumors [3,4,9–14]. Ten of the studies, comprising 14 patients, described the surgical techniques used to excise the tumors [3,4,8–15]. Of those 10, four were total resections [3,12,13,15], five were subtotal [4,8–11], and one did not explicitly state the extent of tumor removal [3]. None of the studies classified the Simpson resection grade of the tumors. Fewer than half of the studies described post-operative follow-up [4,8–11], and only one described post-operative improvements in neurological deficits or symptom relief [16]. Four out of the nine papers did not mention histopathological analysis. Only four studies described tumor size [3,13–15], as described in Table 2.

3.1. Indications for surgery

Table 3 demonstrates the various symptoms that patients in the case series had on presentation for the 11 papers that described