



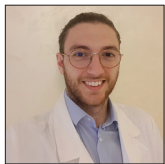
Review Article

## Solitary subdural osteomas: Systematic review of the literature with an illustrative case

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### ABSTRACT

**Background:** Subdural osteomas represent an extremely rare entity with only 20 cases described to date. Despite the typical benign behavior, these tumors can grow to compress the brain and occasionally detach from the dura mater.

**Methods:** A systematic search of the literature was performed in compliance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. After screening for duplicates, 179 publications met the eligibility criteria. Finally, 18 manuscripts were included in this review. Moreover, a detailed description of an illustrative case is provided.

**Results:** The median age at diagnosis was 43.2 years, showing a female prevalence. The inner table of the frontal bone was reported as the most frequent location, and in six cases, the lesions did not show any relation with the dura, which appeared intact. Surgical resection appears to be an effective and safe management option. In the present work, the case of a 60-year-old female who presented with persistent, diffuse headaches which had first occurred 6 months earlier is described. On admission, the physical and neurological examinations were unremarkable, and her medical history disclosed no systemic disease, meningitis, or head injury. Computed tomography showed a homogeneous, high-density nodule attached to the inner table of the left middle cranial fossa.

**Conclusion:** In addition to an in-depth case description, the first systematic and qualitative review of the literature on intracranial subdural osteomas using the PRISMA is provided.

**Keywords:** Brain tumor, Review, Subdural osteoma

### INTRODUCTION

Osteomas are benign tumors consisting of mature normal osseous tissue, commonly encountered on long bones, the mandible, sinuses of the facial bones, and the skull. When originating from the inner table of the skull, these tumors may grow to compress the brain's parenchyma and sometimes involve the dura mater.<sup>[1]</sup> Subdural osteomas, without any attachment to the skull or the dura, are infrequent, with only six cases reported in the literature to date.<sup>[1,3,11,14,15,21]</sup> In the present work, a systematic review of previously described subdural osteomas is provided along with an illustrative case.

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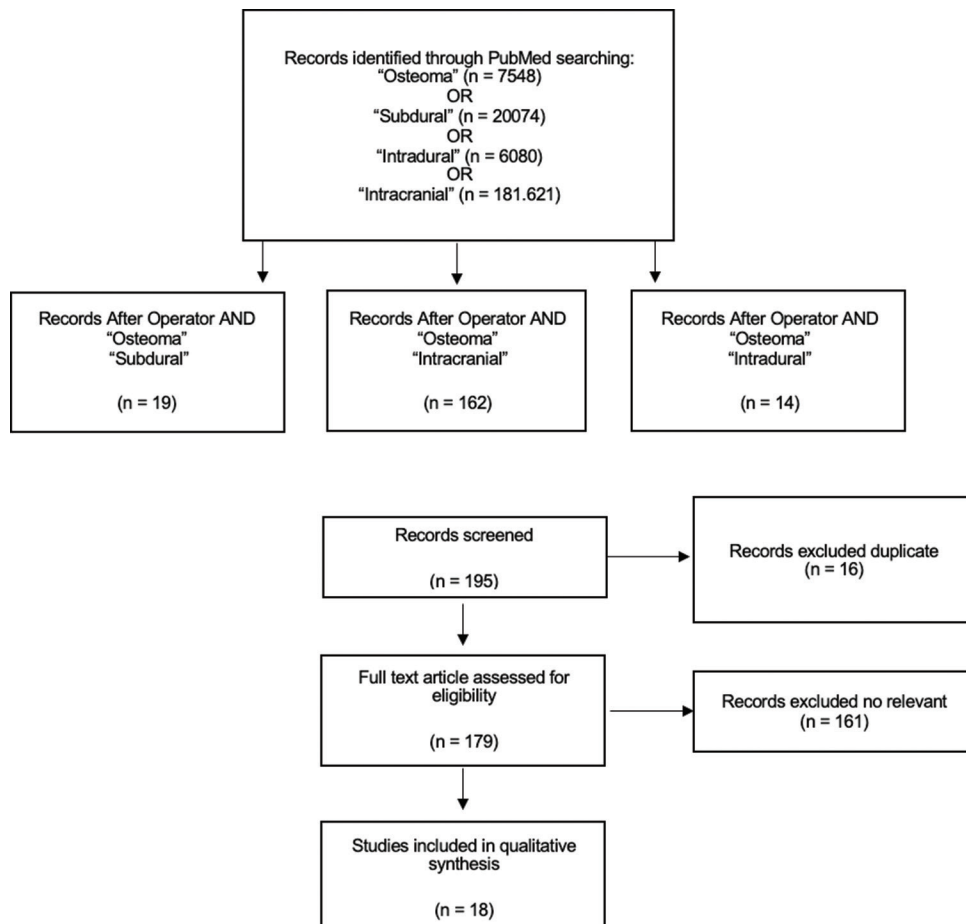
## MATERIALS AND METHODS

Patient's medical history including comorbidities, concomitant medications, diagnosis, and treatment were taken from clinical records. Written informed consent for the case publications was obtained. A systematic review of the literature was performed in compliance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. Screening was performed by reviewing article titles or full text up to December 2021 using the electronic database PubMed. The primary search terms included "Osteoma," "Subdural," "Intradural," and "Intracranial" in the article titles using operator "OR." The extracted citations were then screened for duplicates. Later, operator "AND" was applied to the extracted records by the use of the abovementioned terms to narrow the scope of the review. One hundred and seventy-nine articles met the eligibility criteria for our qualitative systematic review and 161 were excluded because not relevant. Finally, 18 papers were included in the qualitative analysis [Figure 1].<sup>[2,4-8,12-13,17,20]</sup>

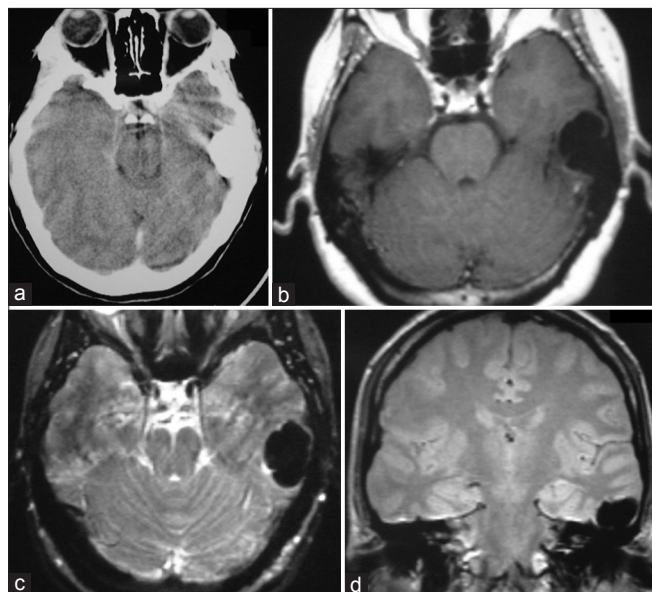
## ILLUSTRATIVE CASE

A 60-year-old woman presented with persistent, diffuse headaches first occurred 6 months earlier. On admission, the

physical and neurological examinations were unremarkable, and her medical history disclosed no systemic disease, meningitis, or head injury. Computed tomography (CT) investigation showed a homogeneous, high-density nodule attached to the inner table of the left middle cranial fossa without any evidence of a curvilinear lucent line between the inner layer of the skull and the calcified mass [Figure 2a]. A head magnetic resonance imaging (MRI) examination revealed a hypointense lesion in T1, T2, and FLAIR sequences [Figures 2b-d] causing a mild mass effect on the temporal lobe. The extra-axial lesion measured  $2.5 \times 1.9$  cm and was first believed to be either an osteoma stemming from the inner table of the cranium or a calcified meningioma. The patient underwent a left temporal craniotomy followed by the incision and exposure of the dura that appeared normal in color but slightly elevated. The tumor was noticed to be whitish in color and hard in consistency and it was firmly attached to the inner layer of the meninges, covering the posterior aspect of the petrous bone and showing no signs of adhesion to the subarachnoid membrane [Figures 3a and b]. A sulcus was observed on the surface of the mass due to the compression of the underlying brain parenchyma. The covering dura and the tumor were excised, and the latter was



**Figure 1:** Preferred Reporting Items for Systematic Reviews and Meta-Analyses protocol used for the systematic review.



**Figure 2:** Images of subdural osteoma. (a) Noncontrast computed tomography of the head showing a calcified, extra-axial mass within the left temporal lobe. (b) Axial T1, (c) T2, and (d) coronal FLAIR magnetic resonance images demonstrating significant signal loss in the region of the calcified mass.

found to measure  $2.5 \times 2.0 \times 1.8$  cm. The dural defect was closed using an artificial dural substitute.

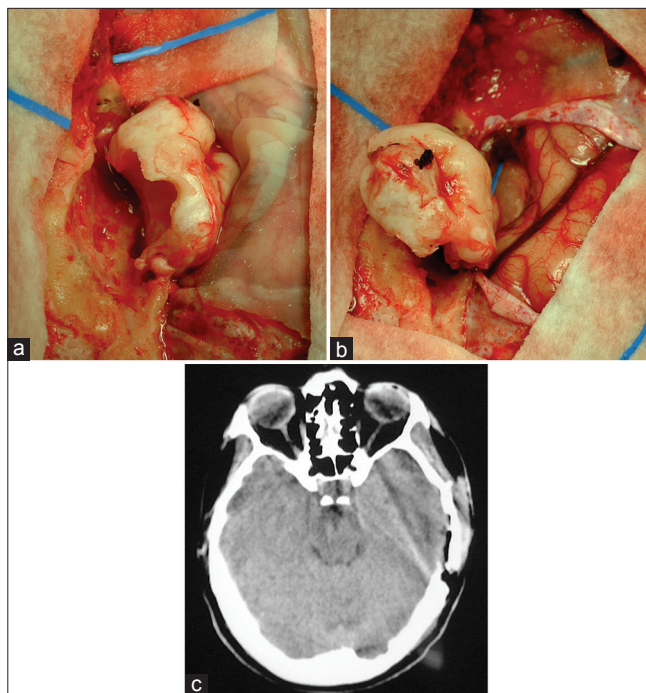
Postoperatively, the patient referred generalized headaches and nausea that persisted for 3 days after surgery. CT scans documented the complete resection of the tumor [Figure 3c], and she was, therefore, discharged.

Pathological examination of the decalcified paraffin sections demonstrated a very dense sclerotic bone covered by a thin layer of collagenous tissue. The lesion was composed of mature lamella bone and normal osteocytes between osteoid layers. The bony trabeculae encased marrow spaces, filled with adipose tissue without any active osteoclastic or osteoblastic activity.

At 2-year follow-up examination, the patient was found to be neurologically intact and reported no relapse of the headaches. A control CT scan was unremarkable for any sign of tumor recurrence.

## REVIEW OF THE LITERATURE

Subdural osteomas represent a rare entity, with only case reports published to date [Table 1]. The mean age of presentation was 43.2 years (range 16–66) with the female sex that appears to be more frequently affected by this tumor (17 F and 3 M). The age distribution suggests a congenital origin rather than an acquired pathology, especially when considering the peculiar slow growth rate of subdural osteomas.<sup>[10]</sup> Although the most commonly reported



**Figure 3:** (a) Intraoperative photograph of the lesion documenting the subdural location of the osteoma that appears nodular, whitish, and hard in consistency. (b) Of notice, the marked compression caused by the tumor on the adjacent parenchyma. (c) Postoperative computed tomography scan demonstrated the complete resection of the tumor.

locations were the frontal (13) and temporal (3) inner tables of the skull, some authors have documented lesions in the frontolateral (1), parafalcine (1), parietal (1), and sphenoid (1) areas. Six patients presented with parenchymal lesions localized in the frontal cerebral convexity, causing mass effects on the intact cerebral cortex. In one patient, the lesion was located in the frontal intracallosal region and extended to both the lateral ventricles. Intraoperatively, subdural osteomas were frequently adherent to the bone and the inner layer of the dura mater, not allowing for the identification of a clear plane for microdissection. Nonetheless, in six cases, “no connections with the dura mater or the skull” were reported with tumors enclosed within the arachnoid membrane, suggesting a subarachnoid origin.

The average duration of symptoms before the diagnosis was 21.8 months, with headache, mostly localized at the site of the lesion, representing the most common manifestation (14 cases). Other symptoms, although less common, included dizziness (4), Jacksonian seizures (1), tinnitus (1), altered mental status (1), and fatigue (1). At follow-up examination (average 21.8 months), the pain subsided in the majority of cases (15) following complete tumor removal, suggesting that the otherwise intractable headaches might be caused by the irritation or compression of the proximal

Table 1: Demographic, clinical, and surgical characteristics of subdural osteomas.

| Year and author                         | Sex | Age | Size (cm)                               | Symptoms                         | Duration of symptoms (months) | Location   | Surgery  | Surgical findings | Outcome  | Follow-up after surgery (months) |
|---|-----|-----|---|----------------------------------|-------------------------------|--|--|-------------------|--|----------------------------------|
| 1962<br>Dukes and Odom <sup>[8]</sup>   | F   | 43  | -                                       | Persistent diffuse headache      | -                             | Right frontal convexity  | Right frontal craniotomy                         | Subdural          | No neurologic deficits                                       | -                                |
| 1983<br>Vakaet et al.                   | F   | 16  | 5×5×6                                   | Headache, Jacksonian seizures    | 12                            | Frontal, intracallosal and extending into the lateral ventricles | -  | Subarachnoid      | Postoperative hemorrhage, death                              | -                                |
| 1995<br>Choudhury et al. <sup>[7]</sup> | F   | 20  | 1×1×1                                   | Persistent diffuse headache      | 4                             | Right frontotemporal convexity                                   | Right frontotemporal craniotomy                  | Subdural          | Recovery   | 10                               |
| 1997<br>Lee et al.                      | F   | 28  | 4×2.5×0.5                               | Persistent left frontal headache | 5                             | Left frontal convexity   | Left frontal craniotomy                          | Subarachnoid      | Reduction of headache frequency                              | -                                |
| 1998<br>Aoki et al. <sup>[2]</sup>      | F   | 51  | 1×1.5×0.7                               | Persistent diffuse headache      | 120                           | Right frontal paramedian convexity                               | Right frontal craniotomy                         | Subdural          | Recovery   | -                                |
| 2002<br>Cheon et al. <sup>[6]</sup>     | F   | 48  | 1.2×2.0×0.7                             | Persistent left frontal headache | 24                            | Left frontal convexity   | -  | Subdural          | Recovery   | 6                                |
| 2003<br>Pau et al.                      | F   | 33  | 3×2×2                                   | -                                | -                             | Left frontal convexity   | Left frontoparietal craniotomy                   | Subdural          | Recovery   | 24                               |
| 2007<br>Jung et al.                     | M   | 60  | -                                       | Persistent diffuse headache      | 36                            | Right frontal convexity  | -  | Subarachnoid      | Recovery   | -                                |
| 2013<br>Chen et al. <sup>[5]</sup>      | F   | 64  | 2.5×2.0×2.0                             | Tinnitus with dizziness          | 3                             | Right mesial-frontal lobe  | Right frontal craniotomy                         | Subdural          | The frequency and degree of dizziness and tinnitus decreased | 6                                |
| 2005<br>Akiyama et al.                  | M   | 24  | -                                       | Persistent diffuse headache      | 6                             | Right frontal convexity  | Right frontal craniotomy                         | Subarachnoid      | Recovery   | 24                               |
| 2012<br>Barajas et al.                  | F   | 63  | 4.5×3.7×2.5                             | Altered mental status            | 0.25                          | Right temporal convexity   | Right temporal craniotomy                        | Subarachnoid      | -  | -                                |
| 2015<br>Krisht et al. <sup>[1,3]</sup>  | F   | 22  | 1.5×2×2.4<br>0.5×0.7×0.4<br>0.4×0.5×0.4 | Persistent diffuse headache      | Several months                | Parafalcine  | Right side, high frontal parasagittal craniotomy | Subdural          | No residual tumor on CT; occasional headaches                | 3                                |
| 2016<br>Cao et al. <sup>[4]</sup>       | M   | 54  | -                                       | Intermittent dizziness           | 5                             | Right parietal convexity   | Right frontal craniotomy                         | Subdural          | Recovery   | 6                                |
| 2016<br>Kim et al. <sup>[1,2]</sup>     | F   | 29  | -                                       | Persistent diffuse headache      | 36                            | Right frontal convexity  | Frontoparietal craniotomy                        | Subdural          | Recovery   | -                                |

(Contd...)

Table 1: (Continued).

| Year and author                      | Sex | Age | Size (cm)   | Symptoms                    | Duration of symptoms (months) | Location                     | Surgery                      | Surgical findings | Outcome  | Follow-up after surgery (months) |
|--------------------------------------|-----|-----|-------------|-----------------------------|-------------------------------|------------------------------|------------------------------|-------------------|----------|----------------------------------|
| 2016 Takeuchi et al. <sup>[20]</sup> | F   | 40  | -           | Persistent diffuse headache | -                             | Right frontal convexity      | Bilateral frontal craniotomy | Subdural          | Recovery | 12                               |
| 2018 Yang et al.                     | F   | 64  | 3.5×3.3×4.5 | Dizziness                   | 24                            | Left temporal convexity      | Left temporal craniotomy     | Subdural          | Recovery | 3                                |
| 2020 Yang et al.                     | F   | 35  | 6.5×4×0.5   | Headache and fatigue        | 24                            | Right frontal convexity      | Frontoparietal craniotomy    | Subdural          | Recovery | -                                |
| 2021 Li et al.                       | F   | 47  | 2.1×1.8×1.1 | Headache and dizziness      | -                             | Left frontal convexity       | Left frontal craniotomy      | Subdural          | Recovery | 72                               |
| 2021 Li et al.                       | F   | 56  | 1.3×1.1×0.8 | -                           | -                             | Right great wing of sphenoid | Right pterional craniotomy   | Subarachnoid      | Recovery | 72                               |
| Present case                         | F   | 66  | 2.5×2.0×1.8 | Persistent diffuse headache | 6                             | Left temporal convexity      | Left temporal craniotomy     | Subdural          | Recovery | 24                               |

dural membrane.<sup>[14]</sup> Nonetheless, in the remaining patients, significant improvement was seen in four cases with the fifth unfortunately dying due to a postoperative hemorrhage.

## DISCUSSION

The pathophysiological mechanisms that lead to the formation of subdural osteomas are still debated, with some authors suggesting that trauma could be an initiating factor that would subsequently lead to a local ossification (a theory already disproved)<sup>[9,16]</sup> and others speculating on the role of enclosed primitive pluripotential mesenchymal cells or the unregulated proliferation of the connective tissue found within the wall of intracranial blood vessels.<sup>[18]</sup> More recently, various evidence suggested that subdural osteomas could originate from the uncontrolled mitosis of heteroplastic primitive cells located in the neural crest, whose uncontrolled proliferation might lead to the slow formation of such lesions. However, pathological examination showed that, although most subdural osteomas are attached to the inner layer of the dura, no dural cells are observed in these tumors.<sup>[15]</sup>

From a radiological standpoint, subdural osteomas appear as calcified, radiopaque lesions on CT scan which is, along with plain radiography, the most common imaging technique used for their identification.<sup>[1,11,14]</sup> The presence of a lucent dural line on CT scans could indicate a subdural origin, which is considered a key feature for the differential diagnosis between subdural osteomas and meningiomas.<sup>[22]</sup> On MRI examination, these tumors appear mostly as low-intensity lesions on T1-weighted images and show either a mixed low and high intensity or a homogeneous hyperintensity on T2WI. Adipose tissue within the lesion can sometimes cause a hyperintense appearance on T1WI and serve as an additional element in the differential diagnosis with meningioma.<sup>[19]</sup> Moreover, the administration of contrast can be useful in the differentials with meningeal ossifications or meningeal calcifications of the dura in elderly patients and is particularly useful when planning for surgical resection.<sup>[3]</sup>

## CONCLUSION

Solitary subdural osteomas represent rare entities, with only 20 cases reported to date. Although pathological examination represents the gold standard for their diagnosis, CT and MRI techniques allow for the identification and differentiation of these tumors from more common lesions such as ossified meningiomas. In the present work, the first systematic literature review along with an illustrative case is provided.

## Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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