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A Multicentre Cohort Study of Spinal Osteoid Osteomas: Results of Surgical Treatment and Analysis of Local Recurrence

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Abstract

Background Context

Spinal osteoid osteomas are benign primary tumours arising predominantly from the posterior column of the spine. These 'osteoblastic' lesions have traditionally been treated with intralesional excision.

Purpose

The purpose of this study was to review the treatment and local recurrence rates for symptomatic spinal osteoid osteomas.

Study Design/Setting

Multicentre ambispective cross-sectional observational cohort study.

Patient Sample

During the study period, a total of 84 patients (65 male, 19 female) were diagnosed with a spinal osteoid osteoma and received surgical treatment. The mean age at surgery was 21.8 ± 9.0 years (range: 6.7-52.4 years) and the mean follow-up was 2.7 years (13 days-14.5 years).

Outcome Measures

Local recurrence, perioperative morbidity and cross-sectional survival.

Methods

Utilizing the AOSpine Knowledge Forum Tumour (AOSKFT) multicentre ambispective database, surgically treated osteoid osteoma cases were identified. Patient demographic, clinical and diagnostic, treatment, local recurrence, perioperative morbidity, and cross-sectional survival data were collected and retrieved. Descriptive statistics were summarized using mean/standard deviation or frequency/ percentage.

Results

In our study most of the tumours were localized in the mobile spine (81/84 (96%)); all patients reported pain as a symptom. According to the postoperative assessment, 10 (12%) patients received an en bloc resection with marginal or wide margins, while two (2%) patients received en bloc resections with intralesional margins, 69 (82%) patients were treated by piecemeal 'intralesional' resection, and 3 (4%) patients were not assessed. A total of six patients (7%) experienced a local recurrence, all of which occurred in patients who had received an intralesional resection.

Conclusions

Benign bone forming tumours of the spine are uncommon. Most patients in our series underwent a piecemeal resection with intralesional margins. This remains safe with a low local recurrence rate. En bloc excision may provide more chance of complete excision of the nidus but is not mandatory. The importance of complete excision of the nidus cannot be over-emphasized.

Keywords: Spinal Osteoid Osteoma; Surgical Management; Recurrence; Primary Spinal Tumor; Cohort Study

Introduction

Osteoid osteomas are benign bone-forming tumours [1, 2]. They are relatively rare as they represent only 3% of all primary bone tumours [3]. In 10% of the cases they occur in the spine with 70-100% located in the posterior elements [2, 4-6]. The majority of osteoid osteomas occur in the 2nd and 3rd decades of life, with a male predilection of 2–3:1 [7]. Osteoid osteomas are characterized by a nidus of osteoid tissue or even mineralized immature bone, with a highly vascularised stroma and are often surrounded by sclerotic reactive bone [7]. The nidus is less than 1.5 cm in diameter by definition, with larger lesions being called osteoblastomas [7]. This is an important distinction as osteoblastomas can be aggressive and thus, careful radiological as well histological analysis is essential to distinguish these tumour types.

Bone scintigraphy is almost invariably positive and has been advocated for localizing the vertebral level in patients with clinically suspected osteoid osteoma [4]. Subsequent targeted CT is generally regarded as the preferred cross-sectional technique for the demonstration and precise localization of the nidus. Osteoid osteoma characteristically manifests as a low-attenuation nidus with central mineralization and varying degrees of perinidal sclerosis [4].

Patients typically present with pain, usually at night-time are relieved by salicylates [8]. There may be stiffness in 89% of cases [9] and scoliosis in 60-70% of adolescents [10, 11] which may become structural [12]

Neurological impairment in osteoid osteomas is rare. Often these tumours go into remission over 2 to 8 years. Surgical intervention is usually indicated in adults with pain not controlled with medication and in adolescents with a risk of developing a structural scoliotic curve [8] The surgical options include intralesional curettage, marginal or en-bloc excision [2, 13-15]. Minimally invasive options such as radio-frequency thermo-ablation [16-18] or CT guided core –excision [19] have also been reported. The purpose of this study was to review the treatment and local recurrence rates for primary symptomatic osteoid osteomas of the spine using the AOSpine Knowledge Forum Tumour (AOSKFT) multicentre database.

Material and methods:

This study was developed and funded under the umbrella of the AOSpine International Knowledge Forum Tumour – AOSpine is a non-profit, international organization of spine care professionals dedicated to delivering knowledge, experience, and evidence to improve patient care and outcomes.

Database Development:

AOSKFT investigators identified centres with experience in the treatment of primary spinal tumours (PSTs) and an established research track record. A feasibility questionnaire was sent to potential sites to gather data on infrastructure, epidemiology, tumour pathology, treatment modalities and outcomes. Centres treating ≥10 patients per year with multidisciplinary oncology care and prospectively collected data were identified. All centres obtained institutional review board or ethics committee approval prior to study commencement.

A PST database consisting of a secure, web-based application to support data capture (REDCap, Vanderbilt University, Nashville, TN, USA) was created. The

AOSKFT Steering Committee composed of Spine Oncology Surgeons, Oncologists, Musculoskeletal Pathologists, and Epidemiologists developed six data modules including: 1) demographic, 2) clinical, 3) diagnostic, 4) therapeutic, 5) cross-sectional survival and local recurrence, and 6) peri-operative morbidity. Data collection was performed using a combined ambispective, cross-sectional design with >75 % of cases being collected prospectively. Prospectively collected databases and clinical charts were used for retrospective data capture. When necessary, mortality information was obtained from government vital statistics databases.

Data Collection:

Between 1981 and 2012, data were collected on 1,495 surgically treated patients with specific primary benign and malignant spine tumours [20] performed at 13 spine oncology referral centres from three geographical regions. Cases were only included if they were admitted to one of the participating spine centres with a diagnosis of a PST and received surgical treatment and standardized follow-up. A study coordinator was employed to assist with data collection, data entry, and capture of cross-sectional survival data. Mandatory and quantifiable data points (i.e., dates, estimated blood loss, etc.) were screened for inconsistencies and incompleteness. Outstanding issues were reported to the centres for clarification.

The treating surgeon used the Weinstein-Boriani-Biagini (WBB) [21] classification system to classify the osteoid osteoma. This is an anatomical classification that incorporates the unique anatomic complexity of the spine with an underlying goal of identifying a safe surgical corridor for tumour excision that will ensure sparing of spinal cord without compromising surgical margins [21] (Figure 1). Postoperatively, the treating surgeon classified the surgical resection according to the following:

En bloc: wide or marginal

En bloc: intralesional (if the surgical intent was en bloc, but resulted in an intra-lesional resection

Piecemeal: intra-lesional

or not assessed

En bloc resection includes the removal of the tumour nidus as a single intact whole, fully encased by healthy tissue (circumferential margin of tumour-free normal tissue). A wide margin occurs when the plane of dissection has been peripheral to the reactive zone (sclerotic area) through normal tissue; a marginal margin describes that the plane of dissection has been performed along the reactive zone; and an intralesional margin is when the tumour has been violated and the dissection is through the nidus.

Results

Patient Demographics

A total of 84 patients (65 males and 19 females) with osteoid osteoma were identified as having received surgical treatment between July 1990 and June 2012 (Table 1). The mean age at surgery was 21.8 ± 9.0 years (range: 6.7-52.4 years) where 36 were children (0-18 years) and 48 were adults (≥19 years). The mean follow-up was 2.7 years (range: 13 days-14.5 years). Most cases appeared in the mobile spine (N = 81, 96%) where C2, T9, T11, L2, and L4 were the most affected vertebrae. All patients reported pain as a symptom.

Figure 2 illustrates a typical example of the presentation of an osteoid osteoma. In this case, a 21-year old male presented with a new onset of painful scoliosis. Radiographs and CT confirmed the diagnosis of an osteoid osteoma at T12 in the concavity. He underwent an uncomplicated intralesional resection.

Tumour Characteristics

The mean size of the osteoid osteoma in the anterior-posterior dimension was 1.40 ± 0.86 cm (range: 0.30-7.00 cm), 1.31 ± 0.60 cm (range: 0.20-4.00 cm) in the left-right dimension, and 1.27 ± 0.53 cm (range: 0.30-3.00 cm) in the cephalad-caudal dimension (Table 2). By using the WBB classification system and grouping the sectors by pedicle (sectors 2-4 and 9-11), spinous process (sectors 12-1), and

vertebral body involvement (sectors 5-8), we found that of the WBB classified tumours (N = 79), 92% involved the pedicles, 23% spinous process, and 25% vertebral body.

Treatment

Preoperative embolization was performed in 15 (19%) patients and the treatment strategies are shown in Table 3. Most patients underwent a posterior surgical approach (N = 69, 83%) and spinal fixation was not utilised in the majority of patients (N = 64, 76%).

Based on the postoperative assessment, 10 (12%) patients received an en bloc resection with marginal or wide margins, whilst two (2%) patients received en bloc resections with intralesional margins, and 69 (82%) patients were treated by piecemeal resection with intralesional margins (Table 4). The type of surgery was not assessed in three patients (4%). Only one patient treated by intralesional resection received adjuvant radiation therapy.

Complications

Complications were observed in five different patients. There were three intra/postoperative complications observed in three different patients: one incidental durotomy, a minor problem with postoperative ventilation and one superficial wound infection. In addition, three delayed complications were observed: postoperative neuropathic pain, pneumonia, and wound dehiscence. One patient suffered both intra/postoperative and delayed complications. These complications were observed in intralesional resected patients except for postoperative neuropathic pain (en bloc: wide resection) and wound dehiscence (unknown resection).

Local Recurrence & Mortality

Six (7%) patients experienced a local recurrence (Table 4) all of which occurred in patients with tumours located in the mobile spine who received a piecemeal 'intralesional' resection. All these recurrences were subsequently treated by further

intralesional resection. Four out of the six local recurrences happened within two years of the index surgery. Information regarding the date of local recurrence was not available for one of the patients. During follow-up, one patient death was observed due to an unknown cause (and not tumour-related), which occurred 2.8 years postoperatively.

Case Presentation

As an example of a patient with a recurrence, we present this case illustration of a 14 year old girl who presented with several months history of cervical night pain. Computed tomography (CT) showed an osteoid osteoma in the arch of C1 (Figure 3). She underwent an intralesional excision performed by a transoral approach with apparent complete removal (Figure 4). A post-operative CT revealed a small degree of residual tumour (Figure 5) but as she was asymptomatic, the surgeon decided to manage her non-operatively.

She did however, represent 5 months later with increasing pain when another CT scan showed clear evidence of the recurrence on the right border of the excision corresponding to the previous surgical site (Figure 6). As a result, it was decided to offer her a further re-excision (Figure 7) following which she had complete resolution of her pain (Figure 8). She had made excellent recovery at the 2 years follow-up without any further recurrence and full cervical motion.

Discussion

This study reports on, to our knowledge, the largest multicentre series of primary symptomatic spinal osteoid osteoma undergoing surgical intervention. It represents data collected from 13 international centres under the guidance of the AOSKFT. During the study period, a total of 84 patients were surgically treated with the majority of the tumours being located in the mobile spine (81/84 (96%)). Sixty-nine patients (82%) underwent a piecemeal ‘intralesional’ resection, 12 (14%) had en bloc (wide/marginal/intralesional) surgery with complete excision of the nidus. A total of six patients (7%) experienced a local recurrence all of which occurred in patients who received the piecemeal intralesional resection. Four of the 6 patients experienced a local recurrence within two years postoperatively and were treated by

further intralesional resection. Although limited to 12 cases, en bloc excision provides more chance of complete removal of the nidus than piecemeal intralesional excision. Whilst conventional intralesional excision surgery remains safe with a low local recurrence and complication rate, importance of excision of the nidus should be the goal.

Osteoid osteomas may be difficult to detect radiologically. Whilst osteoblastomas cause expansile lesions with a well-defined sclerotic margin, osteoid osteomas show reactive bone sclerosis around the nidus. The use of bone scans, CT and MRI has been emphasised with the latter showing high signal intensity in the tumour. Despite the responsiveness of osteoid osteoma to non-steroidal anti-inflammatory medication, surgical treatment is preferred when these agents fail, or to avoid the long term use of anti-inflammatory medication [2]. The need for stabilisation is perhaps only recommended in cases where there has been extensive resection of bone and there is pre-existing deformity.

In a large study of symptomatic osteoid osteomas, Boriani treated 80 cases with intralesional excision [2]. The recurrence rate was 6% and these were treated successfully with further surgery. In this study, there was only one additional patient who underwent percutaneous radiofrequency coagulation (PRC) for a lesion in the vertebral body [2]. These authors stated that there was limited use for this technique in the spine due to the potential damage to neural structures. Rosenthal et al., who were the early pioneers of this method, suggested that the thermal necrosis was less safe if neural tissues were within 13 mm of the probe [22, 23]. Despite the promising outlook for minimally invasive treatments, more data regarding outcomes and risks of minimally invasive treatments, and especially the risks in PRC are necessary [2]. Further, several studies show differing results in small numbers, and there has not been a large study on these minimally invasive treatments for spinal osteoid osteoma [16, 24-26]. There is potential advantage with less tissue trauma reducing post-operative pain and quicker recovery. The main disadvantages include a long learning curve associated with endoscopic surgeries (if used), risks to neurological structures and uncertainty around the histological verification of the specimens using

PRC [2]. A further study is underway by our group comparing treatments between symptomatic non-spinal osteoid osteoma (mostly managed with radiofrequency coagulation in the literature) versus spinal osteoid osteomas (largely treated by intralesional excision as per present study) [27].

Our results support the notion that intralesional resection can be performed safely and with a low recurrence for these benign tumours (which may be benign active) ; this would be inconsistent with Enneking's principles [28]. Goal of surgical treatment of osteoid osteoma remains complete removal of the nidus either by piecemeal or en bloc excision. Wide margin is not required for this benign condition but en bloc excision may provide more chance of complete removal of the nidus than piecemeal excision. However, it is not mandatory and is conceptually distinct from en bloc excision in the treatment of malignant tumours. Thus, complete excision of the nidus is more important than surgical margins in osteoid osteoma and 6 of the 69 patients who had a recurrence likely had this due to incomplete excision of the nidus.

Another interesting presentation of these tumors is scoliosis. Unfortunately, this was not studied in this study but would certainly be an area of future focus. Finally, embolization of the feeding arteries is not really necessary in osteoid osteomas; it was however used in cases where the lesions looked particularly aggressive and where the diagnosis was initially uncertain [29]. The main limitations of our study are the small number of patients undergoing en bloc resection with wide or marginal margins and lack of patient reported outcome measures. However, it remains an ambispective cohort study from 13 international centres in the largest series of primary symptomatic spinal osteoid osteoma treated by surgical intervention.

Conclusion

Most patients in this series underwent a piecemeal intralesional resection (69/84 (82%)) for symptomatic osteoid osteomas with low complication and recurrence rates. Whilst en bloc excision can be performed sometimes quite simply and safely (due to their location within the posterior elements), we do not recommend this form

of excision for all these benign tumours. It is important however, to excise the nidus completely which can be via intralesional resection.

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Figure Legends

Figure 1 – The Weinstein-Boriani-Biagini classifications of spinal tumours. The location of the lesion is described using the 12 radiating zones (1 to 12) and 5 concentric layers (A to E) with layers D and E representing epidural and intra-dural involvement, respectively.

Figure 2 – Antero-posterior radiographs (a) and axial CT (b) showing osteoid osteoma in the posterior elements of a T12 vertebra

Figure 3 – Computed tomography (CT) showed an osteoid osteoma in the arch of C1 (arrow).

Figure 4 – An immediate postoperative axial CT showing apparent complete removal of the osteoid osteoma (arrow).

Figure 5 – A follow-up axial CT revealed a small degree of residual tumour (arrow).

Figure 6 – An axial CT scan at the 5 month stage, showing clear evidence of the recurrence on the right border of the excision corresponding to the previous surgical site (circled by radiologist).

Figure 7 – Intraoperative picture of the surgical exposure through the transoral route.

Figure 8 – A postoperative axial CT scan after the re-excision, showing complete excision of the tumor.

Table 1: Summary of Patient Demographics

Variables	
Gender	
Female	19 (23)
Male	65 (77)
Age (years)	
At diagnosis	21.3 ± 9.0
Child (0-18 years)	14.5 ± 3.0
Adult (≥19 years)	27.2 ± 8.2
At surgery	21.8 ± 9.0
Child (0-18 years)	14.6 ± 3.0
Adult (≥19 years)	27.1 ± 8.3
Spinal Location	
Mobile spine	
Cervical	25 (30)
Thoracic	31 (37)
Lumbar	25 (30)
Fixed spine	
Sacral	3 (3)
Clinical Presentation Pain	
Yes	84 (100)
No	0 (0)

Data are presented as N (%) or Mean ± Standard Deviation

Table 2: Summary of Spinal Osteoid Osteoma Characteristics

Variables	
Tumor Size (cm)	
Anterior-Posterior	1.40 ± 0.86
Left - Right	1.31 ± 0.60
Cephalad-Caudad	1.27 ± 0.53
WBB Sectors[†]	
2 - 4	42 (53)
9 -11	31 (39)
12 -1	18 (23)
5 - 8	20 (25)

Data are presented as N (%) or Mean ± Standard Deviation

† WBB = Weinstein – Boriani – Biagini, Please note that numbers are higher as the tumors involve multiple segments

Table 3: Summary of Spinal Osteoid Osteoma Treatment

Variables	N (%)
Preoperative Embolization	
Yes	15 (19)
No	62 (81)
Surgical approach	
Anterior	10 (12)
Posterior	69 (83)
Anterior-Posterior	0 (0)
Posterior-Anterior	2 (2)
Other	2 (2)
Method of Spinal Fixation	
Anterior	2 (2)
Posterior	18 (21)
Both	0 (0)
None	64 (76)

Table 4: The Effect of Surgical Treatment on Local Recurrence

Variables	Local Recurrence		
	No	Yes	Total
	N (%)	N (%)	N (%)
Surgical Procedure (and margin)			
En bloc: wide or marginal	10 (12)	0 (0)	10 (12)
En bloc: intra-lesional	2 (2)	0 (0)	2 (2)
Piecemeal: intra-lesional	63 (75)	6 (7)	69 (82)
Piecemeal: not assessed	3 (4)	0 (0)	3 (4)