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CLINICAL ARTICLE

Cervical spine chordomas: surgical outcome assessment in a multicenter cohort from the Primary Tumor Research and Outcomes Network

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OBJECTIVE Chordomas are rare, locally aggressive primary neoplasms. Resection with negative margins is the primary recommended therapeutic approach, while adjuvant radiotherapy and chemotherapy can also play a role in their treatment in certain situations, including lesions with positive margins or those that are poorly differentiated or dedifferentiated. Cervical spine chordomas pose significant surgical challenges given their proximity to critical anatomical structures and the mechanical constraints of the cervical spine. In the current case series, authors aimed to explore the clinical and patient-reported outcomes (PROs) of the surgical treatment of cervical chordomas in a large multicenter cohort.

METHODS This multicenter case series analysis utilized data from the prospectively collected Primary Tumor Research and Outcomes Network (PTRON) registry, from its inception (May 16, 2016) to data extraction (February 29, 2024). The study population was restricted to patients with histologically confirmed cervical chordomas involving levels C0–7, who

ABBREVIATIONS AE = adverse event; AOSKFT = AO Spine Knowledge Forum Tumor; DIR = deliberate intralesional resection; EA = Enneking appropriate; EBL = estimated blood loss; EBR = en bloc resection; EI = Enneking inappropriate; FEBR = failed en bloc resection; HRQOL = health-related quality of life; LOS = length of stay; LRFS = local recurrence–free survival; MCS = Mental Component Summary; OS = overall survival; PCS = Physical Component Summary; PEG = percutaneous endoscopic gastrostomy; PRO = patient-reported outcome; PTRON = Primary Tumor Research and Outcomes Network; RT = radiotherapy; SF-36v2 = SF-36 version 2.0; SOSGOQ2.0 = Spine Oncology Study Group Outcomes Questionnaire version 2.0.

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underwent surgical treatment at one of the participating centers, and for whom both the initially planned and postoperatively pathologically confirmed surgical margins were documented. Patient demographics, tumor characteristics, surgical and adjuvant treatments, local recurrence–free survival (LRFS), overall survival (OS), and perioperative adverse events were retrieved. PROs included the Spine Oncology Study Group Outcomes Questionnaire version 2.0 (SOSGOQ2.0), EQ-5D, and SF-36 version 2.0 (SF-36v2).

RESULTS Thirty-eight patients were identified, 12 of whom underwent true en bloc resection (EBR), 18 of whom underwent deliberate intralesional resection, and 8 of whom underwent EBR after intralesional surgery or in whom EBR failed. True EBR led to better LRFS (92% vs 83% vs 63%, respectively) and OS (83% vs 39% vs 50%, respectively). Surgical adverse events within 1 year were more frequent with true EBR (100% vs 39% vs 75%, respectively). EQ-5D, SOSGOQ2.0, and SF-36v2 showed improvement with true EBR, whereas the trends for PROs from the other groups were more variable.

CONCLUSIONS This multicenter case series analysis provides critical insights into the clinical outcomes and PROs in the largest cohort of surgically treated cervical spine chordomas described to date. It underscores the importance and challenges of wide resection for oncological control. It establishes the associated morbidity and provides an overview of PROs following surgery. These findings contribute valuable evidence to inform shared decision-making and optimize patient care.

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KEYWORDS cervical; chordoma; patient-reported outcome; oncology; tumor

Chordomas are rare primary bone tumors of the axial skeleton that exhibit high potential for local recurrence and metastasis.[1,2] The annual incidence of 0.1–0.8 cases per million persons makes studying chordomas a very challenging task, especially when designing comparative treatments or single-center studies.[1,3] The optimal therapeutic approach for disease control involves en bloc resection (EBR) to achieve negative surgical margins without tumor capsular breach.[4–8] EBR with wide or marginal excision is the recommended treatment, when technically feasible.[5] However, these surgeries carry significant morbidity and require a high degree of technical care and surgical experience.[9–11] When the anticipated surgical morbidity is high and the risk to life or function is considerable, other treatments are often considered. These include purposely aiming for an incomplete or intralesional tumor resection.[12] In cases of incomplete tumor resection, radiotherapy (RT) has been shown to provide a survival benefit.[13] The role of chemotherapy and targeted therapy is still unclear, and these options are currently reserved for recurrent and metastatic disease.[14]

The literature on surgical outcomes following resection of cervical spine chordomas is limited and primarily composed of retrospective case series with limited data on surgical margins, complication rates, and patient-reported outcomes (PROs). In this study, we aimed to investigate the clinical outcomes and PROs following the resection of cervical chordomas by utilizing a prospectively collected multicenter data registry from the AO Spine Primary Tumor Research and Outcomes Network (PTRON). The primary endpoints were PROs among 3 resection groups: 1) true EBR (EBR group; Enneking appropriate [EA]), 2) deliberate intralesional resection (DIR) group, 3) EBR after intralesional surgery (IS/EBR+IS) or failed en bloc resection (FEBR) group; Enneking inappropriate [EI]). PROs for health-related quality of life (HRQOL) were assessed using the EQ-5D,[15] Spine Oncology Study Group Outcomes Questionnaire version 2.0 (SOSGOQ2.0),[16] and SF-36 version 2.0 (SF-36v2)[17] when available. The secondary endpoints were oncological outcomes (local recurrence–free survival [LRFS] and overall survival [OS]), estimated blood loss (EBL), hospital length of stay (LOS), readmission and reoperation rates, associated intraoperative and postoperative adverse events (AEs), and incidence of tracheostomy and percutaneous endoscopic gastrostomy (PEG) in each resection group.

Methods

Data Source

Study data were obtained from PTRON, a prospective, multicenter international registry designed to evaluate the management and outcomes of primary spine tumors. PTRON, established and led by the AO Spine Knowledge Forum Tumor (AOSKFT; formerly known as the "Spine Oncology Study Group"), comprises 17 specialized spine oncology centers across North America, Europe, and Asia and is dedicated to advancing research on primary spine tumors. Only 10 centers participated in this study: Baltimore, Beijing, Bologna, Houston, Milan, Oxford, Providence, Rochester, San Francisco, and Vancouver. Center-specific ethics approval and informed consent from all patients were obtained. Demographic and clinical data were collected on REDCap (version 6.5.2, Vanderbilt University), a secure web-based research application.[18]

Study Population

Patients from 10 spine centers included in the registry, from its inception (May 4, 2016) until the date of data extraction (February 29, 2024), were eligible for inclusion in this study. Patients included in the study 1) had a histologically confirmed chordoma diagnosis, 2) had a cervical tumor (levels C0–7), 3) underwent surgical treatment within the PTRON, and 4) had a planned resection and pathological surgical margin data documented in the database. Patients were classified into 3 resection groups based on the surgical plan and postoperative confirmation of the histological margins: 1) EBR group (EA), 2) DIR group, and 3) EBR+IS/FEBR group (EI). The decision to proceed with EBR versus intralesional resection was based on a shared decision-making process between the patient and

the treating surgeon, as well as multidisciplinary review. Many criteria were considered when making that decision: anatomical characteristics of the tumor, feasibility of achieving EBR with a negative margin, expected morbidity from the resection, and age and fitness of the patient to undergo a given surgical plan. In all cases of attempted EBR, the surgical indication was oncological cure but could vary in cases in which an intralesional approach was favored. In some cases where an EBR was not feasible or when morbidity was considered too significant for the patient, an intralesional strategy was adopted, relying more heavily on adjuvant therapy. In other cases, intralesional resection was chosen as a palliative approach.

Description of Variables

Data on patient characteristics (sex, age, BMI, Charlson Comorbidity Index), tumor characteristics (tumor location, vertebral artery involvement, total number of vertebrae involved, largest tumor dimension, histological subtype, and single-nucleotide polymorphism genetic karyotyping), surgical and adjuvant treatments received, LRFS, and OS were retrieved. Clinical variables including EBL during surgeries, intraoperative and postoperative AEs, tracheostomy or PEG, LOS, reoperations, and readmissions were documented. Neoadjuvant and adjuvant chemotherapy and RT were administered according to local institutional protocols and documented accordingly. As previously shown, the use of adjuvant therapy is not standardized and varies greatly among institutions.[19] PROs included the EQ-5D, SOSGOQ2.0, and SF-36v2 when available.

Data Representation and Statistical Analysis

Continuous variables were described using the mean ± standard deviation. Categorical variables were described as the number (percentage). All statistical comparative tests used a predefined significance level (p value) of 5% and compared each resection group. The anatomical distribution heat maps were created on Affinity Designer vector-based editor software[20] and were based on a linear transparency scale from a single color that was adjusted to the relative proportion of patients with disease in a particular anatomical location. LRFS and OS were analyzed using Kaplan-Meier curves with data censoring to illustrate the probability of event-free survival over time, with all analyses performed using SAS version 9.4 (SAS Institute Inc.).[21] For the analysis of PROs, mean scores were calculated for each resection group at each time point (3, 12, 24, and 36 months). This was done by adding the mean baseline score to the difference observed at each corresponding time point. All analyses, including the trend graphs, were conducted using RStudio (Posit Software).[22]

Results

Illustrative Case

A 37-year-old healthy female presented with progressive neck pain and a 6-month history of dysphagia. She had intact motor strength, normal sensation, and symmetrical reflexes bilaterally in the upper and lower extremities. Initial CT and MRI, followed by a CT-guided biopsy, confirmed a conventional chordoma. MRI demonstrated a C3-based tumor invading C2 and C4 with epidural extension without compression of the spinal cord and abutting 270° of the vertebral arteries bilaterally (Fig. 1). Local and systemic staging were negative.

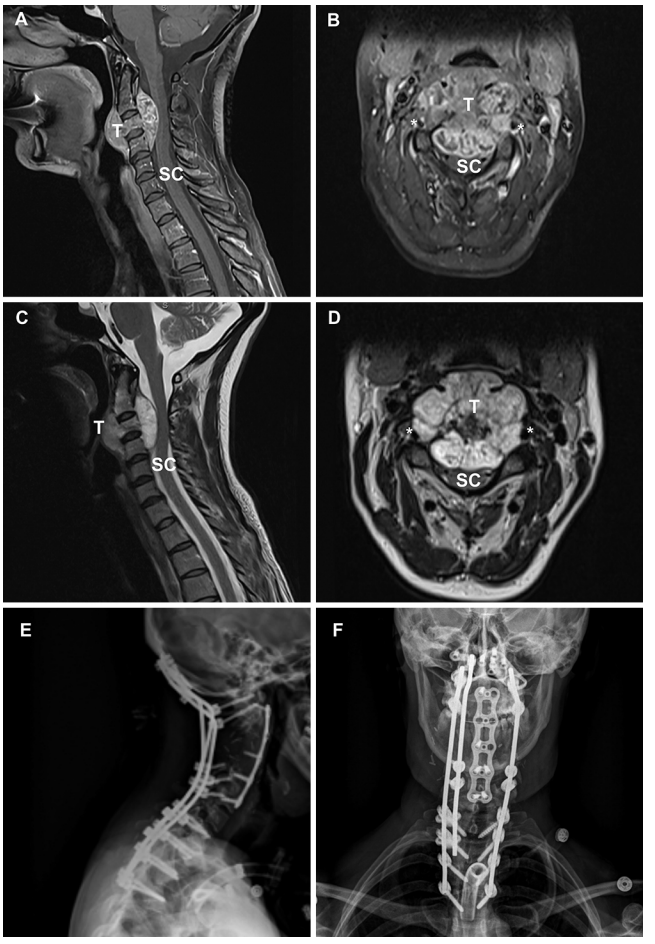

FIG. 1. Preoperative sagittal T1-weighted (**A**), axial T2-weighted (**B**), sagittal T1-weighted (**C**), and axial T2-weighted (**D**) MRI sequences of a C3 chordoma. Postoperative radiographs, lateral (**E**) and anteroposterior (**F**) views, of the construct from occiput to T2 anterior and posterior EBR, instrumentation, and fusion. *Asterisk* indicates the vertebral artery. SC = spinal cord; T = tumor origination from C4.

The patient subsequently underwent a three-stage EBR for a cervical spine chordoma. PEG and tracheostomy were placed preoperatively given the high cervical lesion. The first stage involved occiput to T4 posterior instrumentation, C2–4 tumor mobilization, bilateral C3 and C4 nerve root sacrifice, and bilateral vertebral artery skeletonization from C2 to C5. The second stage involved a wide anterior dissection, C2–4 EBR, reconstruction with iliac crest bone graft, and anterior plating from C2 to C6. The third stage involved posterior final rod placement (Fig. 1). Postoperatively, the patient remained in the ICU for 48 hours, had no postoperative AEs, was weaned off the PEG and tracheostomy, and did not have any diaphragmatic paralysis or neurological deficits. Surgical margins were marginal at the level of the vertebral artery, and adjuvant RT was recommended.

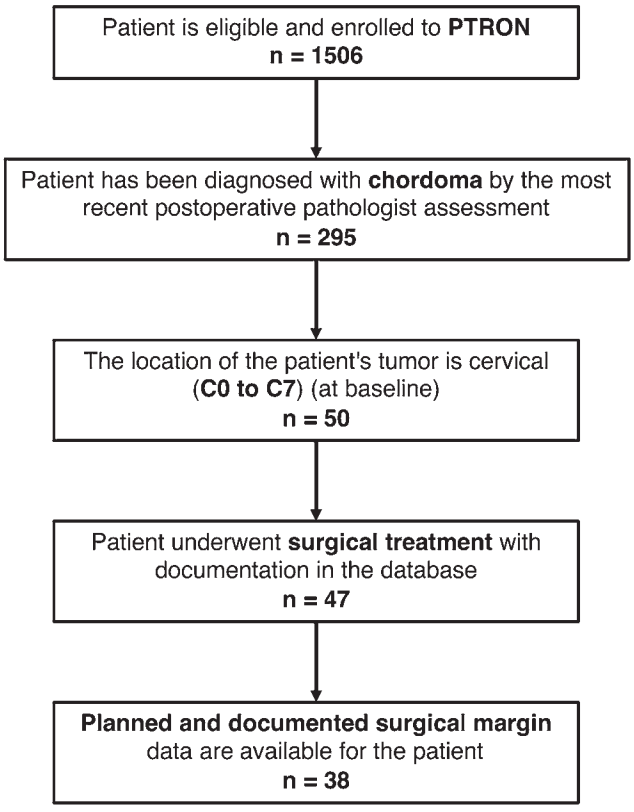

FIG. 2. Flowchart for patient inclusion in the study, fulfilling all criteria.

Patient Demographics

Of the 1506 patients potentially eligible and enrolled in PTRON, 38 were included in the final analysis (Fig. 2), distributed across the resection groups as follows: 12 (31.6%) in the EBR group, 18 (47.4%) in the DIR group, and 8 (21.1%) in EBR+IS/FEBR group (Table 1). Overall, the mean age at diagnosis was 51.7 ± 18.5 years, and the patients were predominantly male (n = 27, 71.1%). The mean BMI was 26.6 ± 4.8, and the mean Charlson Comorbidity Index was 2.2 ± 0.8. The differences in patient characteristics among the resection groups were not statistically significant. Patients were followed up from the first presentation to the last visit or death, with a mean follow-up of 3.1 ± 1.7 years.

Tumor Radiological, Histological, and Genetic Characteristics

Regarding the tumor anatomical distribution (Fig. 3, Table 2), the most frequently affected vertebral segments were C0–2 in the EBR group (n = 10, 83.3%) and DIR group (n = 11, 61.1%). The most affected vertebral segments in the EBR+IS/FEBR group were C3–6 (n = 5, 62.5%). Vertebral artery involvement was more common with DIR (n = 11, 61.1%) than with true EBR (n = 4, 33.3%) or EBR after IS or FEBR (n = 3, 37.5%). The overall mean number of affected vertebrae was 2.5 ± 1.5, with no statistical difference among resection groups (p = 0.48). The overall mean tumor volume was 40.3 ± 53.4 cm^3, with no statistical difference among the resection groups (p = 0.18). Most of the tumors were conventional chordomas. As shown in Table 2, their histological and genetic profiles did not differ significantly among the treatment groups.

Perioperative, Adjuvant, and Neoadjuvant Characteristics

Table 3 summarizes the perioperative characteristics. A greater proportion of patients in the EBR group and EBR+IS/FEBR group was managed with curative intent (100% in both groups) than in the intralesional group (72.2%), although this difference did not reach statistical significance (p = 0.07). Notably, staged procedures were significantly more common in the EBR group (n = 7, 58.3%) and EBR+IS/FEBR group (n = 5, 62.5%) than in the DIR group (n = 3, 16.7%; p = 0.01), reflecting greater surgical complexity.

Differences were also observed in the surgical approach. The posterior surgical approach (n = 21, 55.3%) was the most utilized initial approach across all resection groups. The number of instrumented levels did not dif-

TABLE 1. Patient characteristics stratified by resection group

Characteristic	EBR Group	DIR Group	EBR+IS/FEBR Group	Total	p Value
No. of patients	12	18	8	38	
Sex					0.21
F	2 (17)	8 (44)	1 (13)	11 (29)	
M	10 (83)	10 (56)	7 (88)	27 (71)	
Age at Dx in yrs	52.4 ± 15.4	48.6 ± 19.9	57.9 ± 20.3	51.7 ± 18.5	0.32
BMI	28.0 ± 4.8	26.0 ± 4.9	25.5 ± 4.7	26.6 ± 4.8	0.44
Charlson Comorbidity Index	2.2 ± 0.8	2.3 ± 1.0	2.3 ± 0.5	2.2 ± 0.8	0.94
FU in yrs					
Mean	3.3 ± 1.2	3.1 ± 2.0	2.7 ± 1.8	3.1 ± 1.7	0.74
Median	3.4 (2.5, 4.1)	3.2 (1.1, 4.2)	2.8 (0.8, 4.2)	3.1 (1.8, 4.2)	
Min, max	0.9, 5.2	0.1, 6.6	0.5, 5.3	0.1, 6.6	

Dx = diagnosis; FU = follow-up.
Values are expressed as number (percentage), mean ± standard deviation, median (interquartile range), or minimum and maximum, unless indicated otherwise.

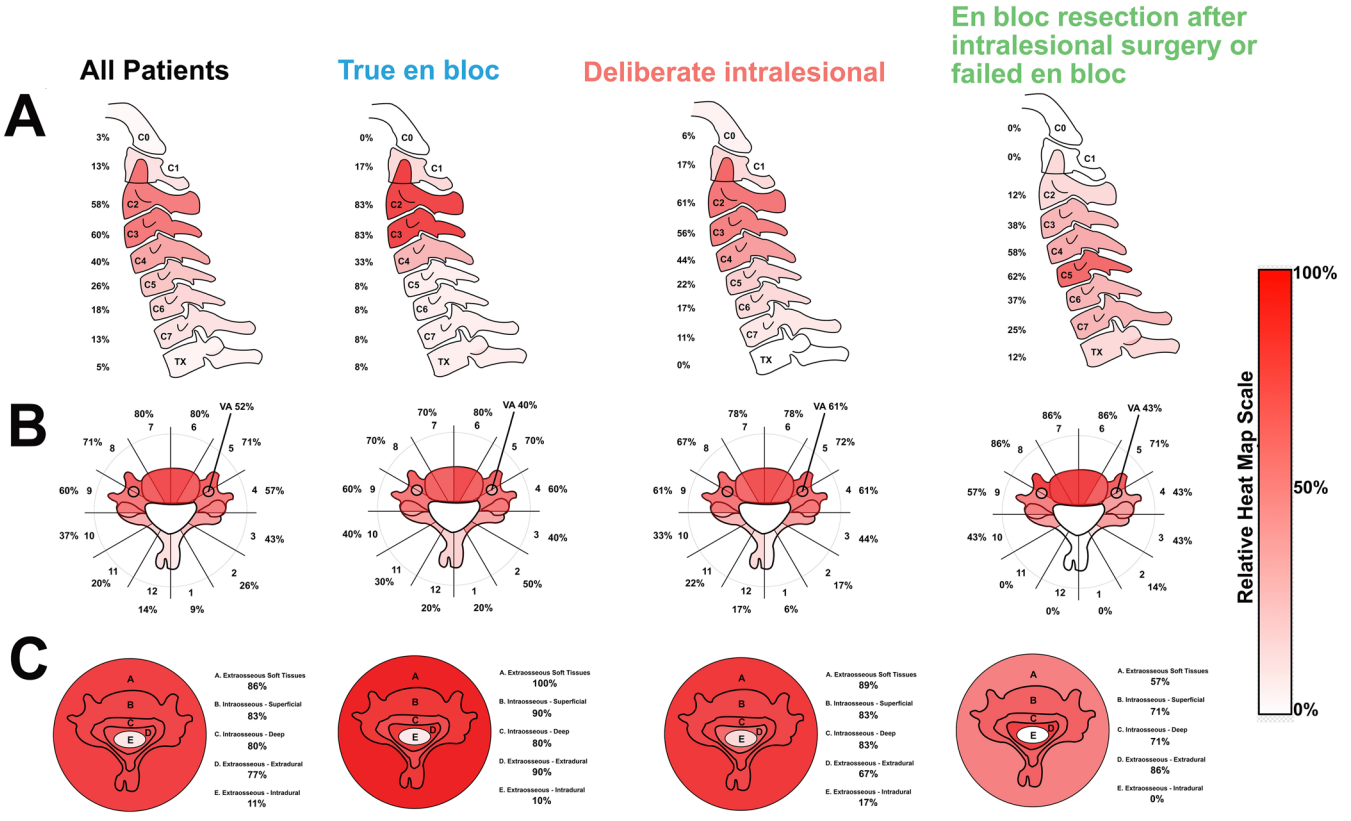

FIG. 3. Heat map relative distribution of tumor according to cervical spine level (**A**) and Weinstein-Boriani-Biagini classification (**B and C**). TX = thorax.

fer significantly among the groups (4.7 ± 2.9, p = 0.37). The mean EBL was comparable across groups, with no statistically significant differences (overall 0.8 ± 0.6 mL, p = 0.22). The hospital LOS was not significantly longer for patients in the EBR+IS/FEBR (43.9 ± 41.5 days, p = 0.27) than in those in the EBR (32.6 ± 26.2 days) and DIR (19.8 ± 14.8 days) groups. Readmission rates also differed significantly, with higher rates in the EBR group (1.3 ± 2.2 readmissions, p = 0.01) than in the DIR (0.1 ± 0.3 readmission) and EBR+IS/FEBR (0.8 ± 0.2 readmission) groups. There was no significant difference in reoperation rates across resection groups (overall 0.2 ± 0.4 reoperation, p = 0.35)

The data indicated no significant differences in the use of neoadjuvant RT, neoadjuvant chemotherapy, adjuvant RT, and adjuvant chemotherapy across the groups, with p values ranging from 0.36 to > 0.99 (Table 3). RT details are provided in Supplemental Table 1. Within 365 days, 25 (65.8%) patients experienced an AE (Table 4): 12 (100.0%) in the EBR group, 7 (38.9%) in the DIR group, and 6 (75.0%) in the EBR+IS/FEBR group. Tracheostomy was required in 4 patients (33.3%) in the EBR group, 3 (16.7%) in the DIR group, and 2 (25.0%) in the EBR+IS/FEBR group, with no significant difference among the groups (p = 0.620). The use of PEG tubes was more common in the EBR group (41.7%); however, there was no significant difference among the treatment groups (p = 0.137). Other complications included dysphagia/dysphonia, dural tears, wound infections, cardiac events, hardware malposition, pneumonia, and pulmonary embolism, with varying incidences across the groups.

LRFS and OS

LRFS and OS (Table 3) varied across the resection groups, but the differences did not reach statistical significance. Patients who underwent true EBR had the most favorable outcomes, with 91.7% showing no local recurrence and 83.3% alive at the last follow-up. In contrast, the EBR+IS/FEBR group had 62.5% of patients without local recurrence and 50.0% were alive within the study period. The DIR group had 83.3% of patients without local recurrence and a survival rate of 38.9%, which was the lowest survival rate. While these trends suggest better survival and lower recurrence with true EBR, the differences were not statistically significant (p = 0.23 for recurrence, p = 0.15 for survival). Kaplan-Meier curves (Fig. 4) illustrate the LRFS and OS tendencies for each resection group.

Patient-Reported Outcomes

The graphic trends for PROs are presented in Fig. 4. The EQ-5D scores improved across all resection groups, with the greatest increase observed in patients in the EBR+IS/FEBR group. SOSGOQ2.0 scores increased

TABLE 2. Tumor radiological, histological, and genetic characteristics among 3 resection groups

Characteristic	EBR Group	DIR Group	EBR+IS/FEBR Group	Total	p Value
No. of patients	12	18	8	38	
Main location of primary tumor					**0.02**
C0–2	10 (83)	11 (61)	1 (12)	22 (58)	
C3–6	1 (8)	5 (28)	5 (63)	11 (29)	
C7	1 (8)	2 (11)	2 (25)	5 (13)	
VA involved					0.33
No	8 (67)	7 (39)	5 (63)	20 (53)	
Yes	4 (33)	11 (61)	3 (38)	18 (47)	
No. of vertebral levels w/ tumor					
Mean	2.8 ± 1.6	2.3 ± 1.6	2.3 ± 0.9	2.5 ± 1.5	0.48
Min, max	1.0, 7.0	1.0, 6.0	1.0, 3.0	1.0, 7.0	
Tumor vol on preop images in cm^3					
Mean	52.8 ± 60.5	20.9 ± 27.4	60.4 ± 72.8	40.3 ± 53.4	0.18
Min, max	2.0, 205.3	1.0, 110.2	4.8, 197.7	1.0, 205.3	
Histological data available	11	13	6	30	
Chordoma subtype					0.15
Classic (conventional)	9 (82)	6 (46)	3 (50)	18 (60)	
Chondroid	0 (0)	0 (0)	1 (17)	1 (3)	
Dedifferentiated chordoma	0 (0)	2 (15)	0 (0)	2 (7)	
Other	1 (9)	0 (0)	0 (0)	1 (3)	
Not assessed	1 (9)	5 (38)	2 (33)	8 (27)	
SNP (rs2305089, GGT > GAT)					0.31
Negative (GG)	1 (9)	0 (0)	1 (17)	2 (7)	
Positive (AA)	0 (0)	1 (8)	1 (17)	2 (7)	
Positive (GA)	1 (9)	0 (0)	0 (0)	1 (3)	
Not assessed	9 (82)	12 (92)	4 (67)	25 (83)	

A = adenine; G = guanine; SNP = single nucleotide polymorphism; T = thymine; VA = vertebral artery.
Values are expressed as number (percentage), mean ± standard deviation, or minimum and maximum, unless indicated otherwise. Boldface type indicates statistical significance.

in all resection groups up to 10 months postoperatively; however, only the EBR group demonstrated continued improvement beyond this period. The SF-36v2 Physical Component Summary (PCS) scores declined across all resection groups during the first 3 months postoperatively. Although all groups showed improvement after this initial decline, only the EBR group surpassed its baseline score by the 36-month follow-up, whereas the other 2 groups remained below baseline. In contrast, the SF-36v2 Mental Component Summary (MCS) scores improved across all 3 resection groups.

Discussion

Our study delved into the feasibility of and the PROs (EQ-5D, SOSGOQ2.0, and SF-36v2) associated with the resection of cervical spine chordomas according to 3 surgical plans: 1) true EBR, 2) deliberate IS, and 3) EBR after IS or FEBR. Our findings underscored the challenging nature of these procedures, with true en bloc excisions demonstrating good feasibility and a tendency for better oncological outcomes (91.7% patients showing no local recurrence and 83.3% alive at the last follow-up). Deliberate ISs appeared to be associated with better LRFS and worse OS than those following EBR after IS or FEBR. Our results indicated that this less aggressive approach may be associated with a higher recurrence risk and decreased OS, although the difference between the two did not reach statistical significance. EQ-5D, SOSGOQ2.0, and SF-36v2 demonstrated similar trends in most treatment groups for up to 3 years, although this result should be interpreted with caution given the small number of patients and variable follow-up.

Our results highlighted the potential benefits of true EBR in terms of LRFS and OS within the first years after treatment. This tendency could become accentuated with a longer follow-up, as many of the patients are still very early in their follow-up. It has been reported that for sacral chordomas, the timing of recurrence is on average 47 months after wide resection compared to 35 months after a deliberate intralesional strategy.[23] Pham and Awad reviewed 76 articles and pooled 195 patients with cervical chordomas.[24] Despite their heterogeneous data, the authors concluded that EBR is associated with less recur-

TABLE 3. Perioperative, neoadjuvant, and adjuvant characteristics among 3 resection groups

Characteristic	EBR Group	DIR Group	EBR+IS/FEBR Group	Total	p Value
No. of patients	12	18	8	38	
Treatment intent					0.07
Curative	12 (100)	13 (72)	8 (100)	33 (87)	
Palliative	0 (0)	5 (28)	0 (0)	5 (13)	
No. of stages					**0.01**
1	5 (42)	15 (83)	3 (38)	23 (61)	
≥2	7 (58)	3 (17)	5 (63)	15 (39)	
1st stage approach					**0.05**
Anterior	5 (42)	5 (28)	0 (0)	10 (26)	
Posterior	7 (58)	10 (56)	4 (50)	21 (55)	
Combined	0 (0)	3 (17)	4 (50)	7 (18)	
No. of instrumented levels	5.3 ± 2.9	4.2 ± 3.2	5.0 ± 2.6	4.7 ± 2.9	0.37
Overall EBL in mL	1.1 ± 0.8	0.7 ± 0.4	0.8 ± 0.8	0.8 ± 0.6	0.22
Hospital LOS in days	32.6 ± 26.2	19.8 ± 14.8	43.9 ± 41.5	28.9 ± 26.9	0.27
No. of readmissions	1.3 ± 2.2	0.1 ± 0.3	0.8 ± 1.2	0.6 ± 1.5	**0.01**
No. of reops	0.3 ± 0.7	0.1 ±0.3	0.1 ± 0.4	0.2 ± 0.4	0.35
Adjuvant & neoadjuvant treatment					
Neoadjuvant RT					0.36
No	11 (92)	12 (67)	6 (75)	29 (76)	
Yes	1 (8)	6 (33)	2 (25)	9 (24)	
Neoadjuvant chemo					>0.99
No	12 (100)	17 (94)	8 (100)	37 (97)	
Yes	0 (0)	1 (6)	0 (0)	1 (3)	
Adjuvant RT					0.75
No	9 (75)	11 (61)	5 (63)	25 (66)	
Yes	3 (25)	7 (39)	3 (38)	13 (34)	
Adjuvant chemo					0.48
No	12 (100)	17 (94)	7 (88)	36 (95)	
Yes	0 (0)	1 (6)	1 (13)	2 (5)	
Local recurrence & survival					
Local recurrence					0.23
No	11 (92)	15 (83)	5 (63)	31 (82)	
Yes	1 (8)	3 (17)	3 (38)	7 (18)	
Survival status					0.15
Alive	10 (83)	7 (39)	4 (50)	21 (55)	
Censored	1 (8)	6 (33)	1 (12)	8 (21)	
Dead	1 (8)	5 (28)	3 (38)	9 (24)	

Values are expressed as number (percentage) or mean ± standard deviation, unless indicated otherwise. Boldface type indicates statistical significance.

rence and metastasis compared to those with intralesional resection, with an average follow-up of 37 months (range 3–108 months).[24] AOSKFT has demonstrated that patients with mobile spine chordomas who underwent EBR with EA margins had significantly lower recurrence rates at 10 years than the patients who underwent intralesional EI resection (14% vs 44%, $p < 0.001$).[5]

Our study demonstrated a higher complication rate in patients who underwent true EBR versus the other surgical approaches. In an international survey of experienced oncological centers, Dea et al. showed that when EBR was anticipated to result in acceptable morbidity, only 3% of surgeons would choose a DIR.[19] The high morbidity observed with EBR of cervical chordomas in the present study supports the concerns raised by experts who argue against EBRs in high-risk situations. Authors have suggested that irregular tumor growth most often prevents the possibility of EBR. Cervical chordoma resection can involve the sacrifice of vertebral arteries and cervical nerve roots. Sacrificing the vertebral artery can increase the risk of stroke, primarily when the tumor is associated with the dominant artery.[25] Similarly, sacrificing nerve

TABLE 4. Summary of AEs, stratified by resection group

Characteristic	EBR Group	DIR Group	EBR+IS/FEBR Group	Total	p Value
No. of patients	12	18	8	38	
AE w/in 1 yr					NA
Yes	12 (100)	7 (39)	6 (75)	25 (66)	
No	0 (0)	11 (61)	2 (25)	13 (34)	
Tracheostomy	4 (33)	3 (17)	2 (25)	9 (24)	0.620
PEG tube	5 (42)	2 (11)	1 (13)	8 (21)	0.137
Dysphagia/dysphonia	7 (58)	1 (6)	0 (0)	8 (21)	*
Dural tear	3 (25)	2 (11)	0 (0)	5 (13)	*
Wound infection/dehiscence	2 (17)	2 (11)	0 (0)	4 (11)	*
Cardiac event	2 (17)	1 (6)	0 (0)	3 (8)	*
Hardware malposition	1 (8)	1 (6)	0 (0)	2 (5)	*
Pneumonia	4 (33)	2 (11)	0 (0)	1 (3)	*
Pulmonary embolism	1 (8)	0 (0)	0 (0)	1 (3)	*

NA = not available.
Values are expressed as number (percentage), unless indicated otherwise.
* Statistical analysis was not performed because of insufficient sample size.

roots can lead to the loss of upper extremity function and significantly impact patient quality of life.[26] A study by Molina et al. associated the location of the tumor in the cervical spine with surgical morbidity, with surgery for chordomas involving the C1–2 vertebrae carrying significantly higher morbidity (71%) than those in the subaxial spine (21%).[27] Pooled data indicated that EBR is associated with higher complication rates, most commonly dysphagia, respiratory compromise, and wound dehiscence, leading to the conclusion that en bloc surgeries may be acceptable despite the higher complication rate.[24] Despite these challenges, our data suggested that when en bloc excision is feasible, it may confer better local control and better OS.

Importantly, we showed that LRFS, OS, and PROs can be impacted for up to 3 years postoperatively; however, we do not have longer-term data about this critical outcome at this time. Park et al. reported the outcomes for 45 patients with cervical spine chordoma over a median follow-up of 4.3 years. Local recurrence was noted in 46.7% of the patients, whereas distant metastasis was observed in 8.9%. Age was the only variable associated with mortality.[28] Yang et al. reported a 37.5% recurrence rate among 34 patients with sacral chordoma with a mean follow-up of 4.1 years.[29] Tumor level and margins were variables associated with recurrence in that study. In a systematic review of PROs and HRQOL in patients with primary tumors of the spine, the AOSKFT reported that at the short-term follow-up after surgery (up to 4 years), SF-36 PCS scores were still below normative data.[10] However, when longer-term follow-up data were available (up to 10 years), HRQOL scores returned to normative values, and only tumor recurrence was associated with lower HRQOL. This systematic review and our findings regarding a potential early difference in local control and overall mortality support the concept of an EA approach to minimize local recurrence.

Although our study focuses on the different surgical approaches and their impact on outcomes in patients with cervical spine chordomas, novel forms of RT are taking greater roles in the management of these complex cases and could significantly modify treatment paradigms in the future.[12] In the 2009 *Spine* Focus Issue on Oncology, AOSKFT advised the use of adjuvant RT at doses exceeding 60–65 Gy for chordoma following incomplete or intralesional resections.[30] In a study involving 76 patients with primary chordomas with at least 5 years of follow-up, Tobert et al. found that those who received proton RT at doses above 70 Gy had significantly better OS (82% vs 63%) and LRFS (93% vs 78%) than the patients who did not complete RT.[31] In contrast, in a multicenter analysis of 193 sacral chordoma cases, Houdek et al. reported no significant association between RT and enhancements in survival or local disease control.[32] The role of RT for chordomas in challenging locations is evolving and yet to be defined.

Our results contribute to the evolving understanding of optimal surgical approaches for cervical spine chordomas. The high morbidity associated with en bloc excision should not be overlooked, and efforts should be directed toward refining techniques and minimizing complications. Furthermore, the apparent correlation between en bloc excisions and improved local control highlights the need for personalized treatment plans that consider both short- and long-term outcomes and are tailored to every individual. Specifically, the role of RT based on the surgical margin and biology of the tumor is still controversial and needs further study.

Several limitations should be considered when interpreting data in this study. While this case series presents outcomes across different treatment groups, it is not sufficiently powered to detect statistically significant differences between them. The short follow-up period limits the evaluation and differentiation of outcomes, mainly local

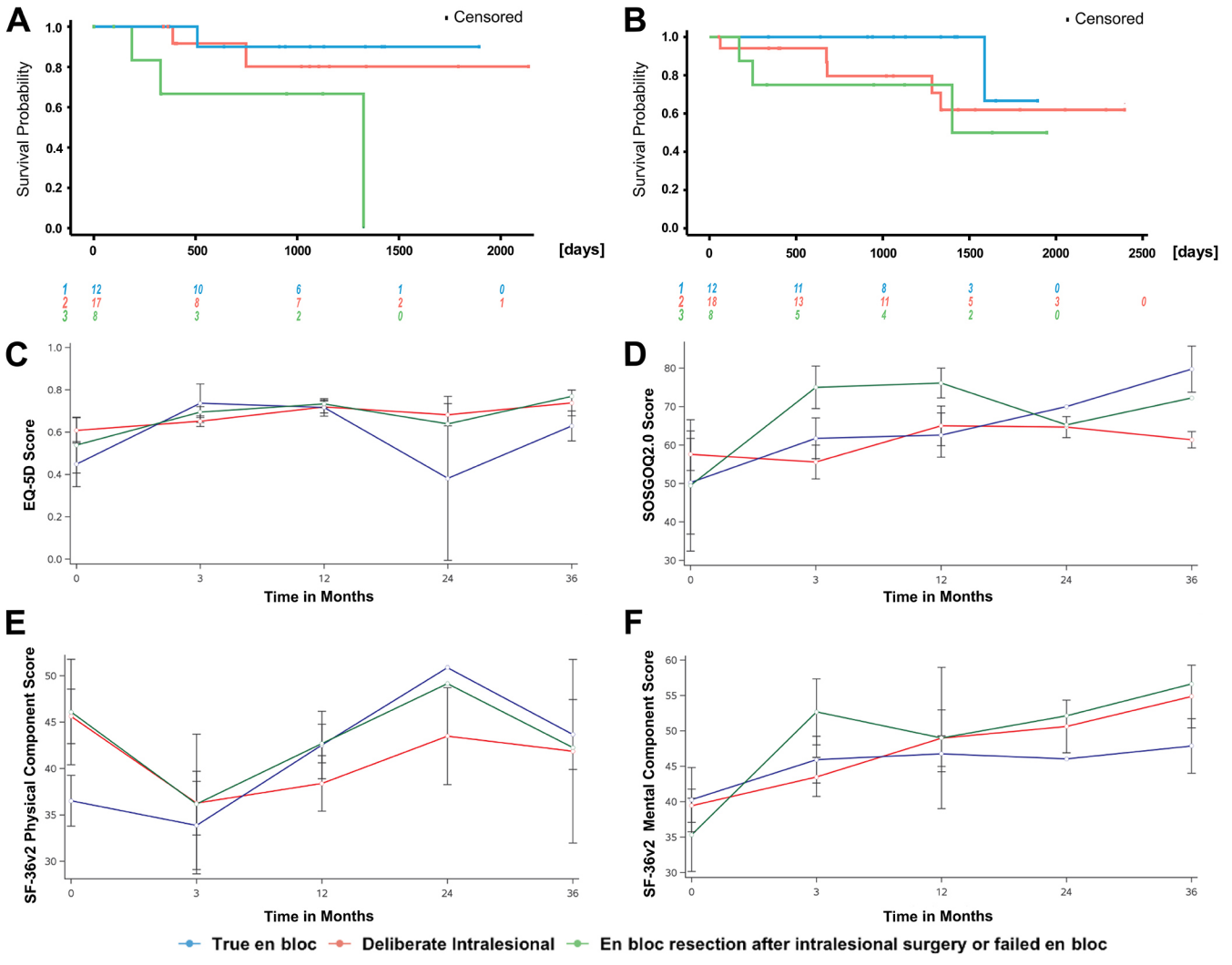

FIG. 4. Kaplan-Meier curves for LRFS (**A**) and OS (**B**). PROs for EQ-5D (**C**), SOSGOQ2.0 (**D**), SF-36v2 PCS (**E**), and SF-36v2 MCS (**F**).

recurrence and survival, in a disease known for delayed local recurrence and late mortality.[29] However, one of our primary objectives was to assess PROs and AEs after different types of treatment, and these outcomes are more adequately captured by this follow-up duration. The observational design and nonrandom sampling introduce potential selection bias. Cause-specific mortality was not analyzed, as data were unavailable in the PTRON registry, and the study did not aim to establish the superiority or inferiority of any treatment. Thus, the lack of statistical significance for many findings is not surprising, especially considering the rarity of this disease. To our knowledge, this is the most extensive study on the outcomes of different types of cervical chordoma resection. Furthermore, it is the first study on the subject to incorporate PROs, which we believe will become an invaluable tool for therapeutic decision-making once the database grows and the analysis becomes more powerful. We believe that this study constitutes the cornerstone of additional research on the subject. Indeed, as the PTRON registry matures, the number of patients and the follow-up time will increase. We believe that we will be able to clarify the potential outcomes associated with each surgical group more accurately in the coming years.

Conclusions

This retrospective study of prospectively collected data offers valuable insights into the clinical outcomes and PROs of the surgical treatment of cervical spine chordomas. Our study sheds light on the balance between en bloc tumor excision and less invasive ISs in managing cervical spine chordomas. The high morbidity observed with en bloc excision necessitates careful consideration, but the approach may offer superior local control when it is feasible. Conversely, ISs, while associated with lower morbidity, appear to carry increased risks of recurrence, progression, and mortality. Considering the nuanced interplay between surgical morbidity and long-term outcomes, these findings underscore the imperative for a personalized patient-cen-

tric approach. They underline the complexities of managing cervical spine chordomas, emphasizing the need for a nuanced approach in surgical shared decision-making.

A comprehensive evaluation of patient-specific factors should guide the choice between en bloc and ISs to achieve an optimal balance between surgical morbidity and long-term disease control. Further research is warranted to refine surgical techniques and approaches, improve patient selection, and enhance overall outcomes. As we strive to optimize patient care, ongoing research, multidisciplinary collaboration, and advancements in surgical techniques and adjuvant therapies remain pivotal in enhancing the effectiveness and safety of surgical interventions.

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Supplemental Information

Online-Only Content

Supplemental material is available with the online version of the article.

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