Suboccipital and cervical chordomas: radical resection with vertebral artery control

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Object. Radical resection of chordomas seems to improve a patient's long-term prognosis. At the suboccipital and cervical levels, the vertebral artery (VA) is often considered as a limit in surgical possibilities. The authors report on the management of the VA in a series of 36 patients with chordomas located in the cervical region.

Methods. Over an 11-year period, 36 patients with chordomas located at the suboccipital (28 patients) or cervical (eight patients) level were treated in the authors' neurosurgical department. In 30 patients, the tumors extended laterally toward the VA and required surgical control of the VA. Sixteen of these 30 patients harbored primary tumor, whereas 14 were treated for recurrent disease.

The VA was encased in the tumor in 23 patients, with stenosis in six cases. A balloon occlusion test was performed in seven patients and the VA was resected in four. Extensive resection, via a lateral approach on one (22 cases) or on both (eight cases) sides, did not cause any permanent postoperative deficits. In five patients a complementary approach was performed: posterolateral in one and transoral in four. Spinal fixation was performed via the lateral approach (eight cases) or via a complementary posterior approach (five cases). All but two patients underwent radiotherapy, including 10 who underwent proton-beam radiotherapy.

Conclusions. Chordomas extending laterally to the VA can be radically resected via a lateral approach without causing significant morbidity. A complementary approach is often necessary. The best results are achieved in patients with primary compared with recurrent tumor.

KEY WORDS • chordoma • vertebral artery • radiation therapy • surgical approach

Chordomas are locally invasive tumors thought to arise from remnants of the fetal notochord.^{26,32} The most common location is the sacrococcygeal region followed in descending order by parasellar and clivus area and the mobile spine.^{3,4,9,18,30} In many articles, the authors do not distinguish among the various locations, however, each site poses particular problems during resection. In cervical and suboccipital locations, the VA is generally considered to be a limitation to resection. We report on the management of the VA in a series of 36 patients with chordomas located in the cervical region.

CLINICAL MATERIAL AND METHODS

Between 1989 and 2000, 36 patients harboring cervical chordomas were treated in the Department of Neurosurgery of the Lariboisière Hospital (Table 1).

There were 16 female and 20 male patients who ranged

in age from 11 to 63 years (mean 38 years). Most tumors were located at the upper cervical level, corresponding to the suboccipital segment of the VA from C-2 to the tip of the clivus; only eight tumors were located below C-2 at the level of the transverse segment of the VA from C-6 to C-2. In all but one case fewer than three vertebral levels were involved. In one case the lesion was an extensive epidural tumor extending from C-1 to T-1. In all patients the main part of the tumor was invading the VBs, the odontoid process, or the anterior arch of atlas anteriorly. In six patients the tumor extended only anteriorly toward the pharynx. In 30 patients, it also extended laterally toward the VA: on one side in 22 cases and on both sides in eight cases.

Of the 30 patients in whom the chordoma involved the VA, 16 were referred to our department with primary disease, whereas 14 were referred after one (11 cases) or two (three cases) recurrences.

All patients underwent MR imaging, with and without contrast injection, and CT scanning with bone windows; angiography was performed in 15 cases. On MR images, especially on T₁-weighted coronal views, the VA can be well identified; its diameter, its course along the spine, and

Abbreviations used in this paper: CCJ = craniocervical junction; CT = computerized tomography; MR = magnetic resonance; VA = vertebral artery; VB = vertebral body.

TABLE 1
Summary of general and treatment data

Tumor Type by Group	Patient Age (yrs)			No. of Operations		Radiotherapy (proton beam)		Mean Follow-Up Time (yrs)
	<15	15–40	>40	Range	Average	Yes	No	
primary (16 patients)	4	2	10	1–4	1.8	14 (7)	2	5
recurrent (14 patients)	3	5	6	1–2	1.3	14 (3)	0	3.5

its displacement by the tumor can generally be analyzed. However MR imaging cannot demonstrate an anterior radiculomedullary branch arising from the VA. Because this branch never arises above the C-3 level, angiography need not be performed in cases in which the tumor is located in the suboccipital region when MR studies are of good technical quality. Thus, angiography was performed only in the eight patients in whom the chordoma was located below C-3 and in seven patients treated for tumor recurrence.

RESULTS

The VA was surrounded by the tumor in most patients (23 cases), whereas it was displaced laterally at the lateral limit of the tumor in seven patients.

In six cases the tumor compressed the VA, causing a reduction in the diameter, but no case of complete VA occlusion was observed. The involved VA was equal in size to that of the contralateral one in 18 patients, larger in size in seven patients, and smaller in size in five patients.

Balloon occlusion tests were performed in seven patients, including six with recurrent disease, and all of the tests were well tolerated.

The 30 patients with laterally extending chordomas underwent surgery via one or several approaches, including at least one lateral approach. Overall 38 lateral approaches were performed: 22 unilateral and eight bilateral either the same day or in a two-stage procedure. In the group of patients with primary tumor, the lateral approach was performed on one side in 12 patients and on both sides in four; it was combined with a posterolateral approach in one patient and a transoral approach in four patients. In the group of patients with recurrent disease, the lateral approach was performed unilaterally in 10 patients and bilaterally in four; it was never combined with another approach.

Spinal fixation was performed in the surgical stage in which the lateral approach was used in eight patients (three primary tumors and five recurrences).⁵ A complementary posterior approach was necessary in five patients to achieve a posterior fusion. These five posterior fixation procedures were all performed in patients with primary tumor, but three patients with recurrent disease had undergone a posterior fixation before being referred to us.

Overall 48 surgical approaches were performed in the 30 patients; 18 procedures were performed in the 14 patients with recurrent tumors (average 1.3 operations per patient), and the 16 patients harboring primary tumors underwent 30 operations (average 1.8 operations per patient) (Table 1 and Fig. 1).

In all cases the resection was as radical as possible: all visible tumor was resected, including an intradural portion when present, and the bone was drilled beyond the tumor's margins. The completeness of resection was confirmed on postoperative CT and MR studies in all cases. In four patients the VA was sacrificed (in three cases of recurrent disease and in one case of primary tumor). The cervical nerves roots were preserved whenever possible, except for the C-1 and C-2 nerve roots that were divided in the VA exposure. The C-3 nerve root was frequently resected because of its limited neurological function. The C-5 nerve root was sacrificed in one patient with recurrent tumor and the C-6 nerve root was sacrificed in another patient with recurrent disease; both of these patient already had a corresponding sensory motor deficit.

Adjunctive radiotherapy was administered in 28 patients, including 10 who underwent proton beam radiotherapy (seven in the primary tumor group and three in the recurrent tumor group). Only two patients with the primary disease did not undergo radiotherapy.

Surgical Technique of Lateral Approach

Transverse Segment (C2–6). The patient is placed supine with the head slightly extended and rotated toward the opposite side. The skin incision follows the medial edge of the sternocleidomastoid muscle centered over the level of the tumor. The field between the medial aspect of the sternocleidomastoid muscle and the lateral aspect

Fig. 1. Diagram depicting the surgical and radiological treatments in 30 patients harboring chordomas with lateral extensions.

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of the internal jugular vein is then opened. This leads directly to the prevertebral muscles (longus colli and longus capitis) covering the lateral aspect of the VBs and the transverse processes. These muscles are cut after having identified and retracted the sympathetic chain, which runs beneath the aponeurosis of the prevertebral muscles. After fluoroscopic confirmation, the muscle cutting is conducted along the chosen transverse processes. Working subperiosteally, the anterior branch of the transverse processes is then removed; this provides to exposure of the VA while keeping intact the periosteal sheath that surrounds the VA and its venous plexus.

Suboccipital Segment. The patient position is similar to that used for the transverse segment level. The head is more or less rotated toward the contralateral side according to the location of the tumor. The skin incision follows the superior part of the medial edge of the sternocleidomastoid muscle, then the mastoid process and then along the occipital line. The sternocleidomastoid muscle is detached from the mastoid process, and the field between this muscle and the internal jugular vein is opened. The upper limit of the field is the mastoid process and the digastric muscle attached to it. Crossing the field is the accessory nerve, which must be identified and dissected from its junction with the sternocleidomastoid muscle up to the skull base. The fat sheath covering the depth of the field is separated from the deep muscles and rolled around the nerve to protect it during retraction. The transverse process of atlas is then exposed approximately 15 mm above and below the tip of the mastoid process. The little muscles (oblique and rectus muscles) inserted on it are detached, thus exposing the VA C1–2 segment and the above C-1 segment. The C-2 transverse process may then be exposed. Because of the particular length and oblique direction of the C-2 transverse process, it is difficult to approach it directly. It is recommended first to expose the C1–2 segment and the C-3 transverse process so that the VA exposure progresses along the VA bayonet shaped course from C-3 to C-1. The C-1 transverse process can be resected and the foramen opened by working subperiosteally just as any other transverse foramen. The VA can be mobilized away from the transverse foramen according to the location of the tumor. This was performed in 19 patients (in seven cases bilaterally).

Tumor Resection. The general principle is to control the VA one level above and below the tumor’s margins and then to progress toward the tumor. The periosteal sheath of the VA is generally invaded at the level of the tumor and has to be resected. Proximal and distal controls allow venous bleeding to be easily managed. Chordomas rarely extend beyond this periosteal sheath, which is a very resistant barrier to tumor extension just as is dura mater. Still more rarely, chordomas invade the VA wall. It has not been observed in cases of primary tumor but may be found in case of recurrence. In our series it occurred in four cases in which the VA was ligated and resected.

Via the lateral approach, the VBs and/or the anterior arch of atlas and the odontoid process can be almost entirely drilled out. The anatomical limits are the contralateral prevertebral muscles and the VA. It may be necessary to drill the VBs to gain access to the anterior aspect of the dural sac because of tumor extension or to achieve an

oblique corpectomy.^{12,15} Intradural extension can also be reached this way.

Extending the exposure laterally, the cervical nerve roots can be exposed. From their distal control, one may then progress toward the intervertebral foramen, working behind the VA and reaching the dural sac after having enlarged the foramen.

Posteriorly the tumor may be followed as far as to the laminae. At the C1–2 level, the posterior exposure can be extended to the midline. Superiorly the posterior fossa can be opened via different combined approaches: retrosigmoid (five cases in this series), presigmoid (none), or transpetrosal (none).

There were no surgery-related deaths. Morbidity was very limited. Transient cases of Horner syndrome were observed in four of the eight patients with lower cervical chordomas. Eleventh and 12th cranial nerve weakness was demonstrated in three and two patients, respectively. One patient, who underwent surgery via a lateral approach and in a second stage a transoral approach, presented with cardiopulmonary arrest 3 weeks postoperatively; she recovered but was in a vegetative state and eventually died at 1 year.

For the other patients, the follow-up period ranged from 1 to 11 years (average 4 years; a mean of 5 years in those with primary tumor patients and 3.5 years in those with recurrent disease) (Table 1).

In patients with primary tumor, six recurrences (40%) were observed with five deaths (33%) and there was a mean delay of 34 months before the first recurrence. In the primary tumor group, two of the five deaths were not related to the disease (pneumonia in one case and intracerebral hemorrhage in the other). In patients with recurrent tumor, a new recurrence was observed in all cases, leading to death in eight patients (57%) and with a mean delay of 15.2 months before the first recurrence. All deaths in the recurrence group were disease related and occurred after a last recurrence (Table 2).

ILLUSTRATIVE CASE

This 70-year-old man presented in September 1999. Based on CT and MR imaging evidence, a C1–3 chordoma was diagnosed. At this time he had a 1-year history of severe pain and stiffness of the upper neck. He had attended two previous neurosurgical consultations in which surgery was considered too dangerous. A biopsy sample was obtained transorally, and results of examination confirmed the diagnosis of chordoma. His medical history was significant for a myocardial infarction and systemic hypertension. A surgical therapy involving a lateral approach on both sides and a posterior fixation was pre-

TABLE 2
Summary of tumor recurrence data according to group

Tumor Type by Group	1st Recurrence and Op (mos)	No. of Recurrences (%)	No. of Recurrences Per Year	Disease-Related Death (%)
primary	34	6 (40)	0.13	3 (20)
recurrent	15.2	14 (100)	0.64	8 (57)

Fig. 2. Preoperative MR images. Sagittal (*left*) and axial (*right*) images demonstrating the chordoma extension from C-1 to C-3 and bilaterally to both VAs.

pared. At that time he refused operative treatment and but agreed to wear a rigid collar.

Three months later he returned with a tetraparesis predominating on the left side as well as left-sided hypesthesia. At this time he agreed to undergo surgical treatment. Repeated CT and MR imaging demonstrated moderate enlargement of the tumor. The tumor involved C1–3 and was destroying the C-2 VB, the base of odontoid process, and extending on both sides to the VA (Fig. 2 *left* and *right*). In a single sitting, he underwent surgery via a left- and then right-sided lateral approach; this was followed by a posterior occipitocervical fixation in which bilateral plates and bone grafts were placed (Fig. 3 *upper left* and *right*). The overall duration of the three procedures was 14 hours. Total blood loss was 400 ml. His postoperative course was very smooth, and he made a nearly complete recovery. At his most recent follow-up visit, 14 months postoperatively, he only complained of a slight weakness in the left thigh and hypesthesia of the left arm; however, he was fully alert, driving his car, hunting, and enjoying many activities.

Postoperative imaging studies (Fig. 3 *lower left* and *right*) demonstrated that complete tumor resection was achieved. Three months postoperatively he underwent a combination of conventional radiotherapy and proton-beam radiotherapy. The last control image study, obtained 1 year postsurgery, demonstrated a stable clinical and radiological condition.

DISCUSSION

Chordomas are neoplasms thought to arise from a remnant of the fetal notochord.^{9,26,32} Although it is often said that the lesion is a slow-growing process, actuarial survival rates at 5 and 10 years are approximately 50% and 35%, respectively,^{10,17,18,21} and metastases^{4,7,23,30} are not exceptional. Therefore, several authors have emphasized the need for an aggressive treatment in which radical resection is performed.

In fact the biological behavior of chordomas is highly variable. It is difficult to determine from the literature which factors clearly influence the prognosis of chordomas.

Fig. 3. *Upper Left* and *Right*: Postoperative CT scans revealing the extent of bone resection and the posterior fixation. *Lower Left* and *Right*: Postoperative sagittal (*left*) and axial (*right*) MR images confirming gross-total resection.

Histological features, especially those indicating malignancy such as mitosis, necrosis, or atypia, have no influence on the evolution of the tumor^{4,10,16,30,35} except in the very few cases of sarcomatous changes.³¹ The chondroid-type chordoma has been reported by several authors as being associated with a more favorable patient outcome; however, this is a controversial issue, and some authors argue that this subtype of chordoma does not exist.⁶ In the past, some chondrosarcomas may have been misdiagnosed as chondroid chordomas. Today, however, immunohistochemical studies permit differentiation between the two lesions.^{20,27} Furthermore, the chondroid type is more often observed in young patients^{2,7,8,11,16,35} and a better prognosis generally exists for younger patients as well.^{10,17,35} According to Forsyth, et al.,¹⁰ age less than 40 years is the only significant factor associated with better prognosis. Alternatively it seems that very young patients (< 5 years of age) have a very poor prognosis.^{20,35} Tumor recurrence and death very often occur in less than 1 year, and there is a high frequency of metastasis. Sex has also been proposed as a prognosis factor⁴ but this has not been confirmed.

Finally, in several papers the authors mention that chordomas of the spinal cord grow more rapidly and metastasize more often than those located in other regions.^{16,20,30,34} Sundaresan, et al.,³⁰ described a series of 18 vertebral chordomas, in which 39% of the cases had disseminated metastases. Harwick and Miller¹⁶ reported on five patients with cervical chordomas, and only two patients survived

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more than 18 months despite having undergone treatment. In a review of the literature published in 1975, Wellinger³⁴ found 46 cases of spinal chordomas in which five patients who went untreated were dead in less than 18 months. In Wellinger's review, 15 chordomas were located in the cervical region; nine patients died before 3 years and only two survived more than 5 years. In most recent papers the authors have included different locations (skull base and vertebral) and do not permit any separate analysis.^{2,19,22,28} In fact, it is difficult to anticipate the speed of growth of chordomas in general and even more difficult for those in a given location. In most publications, chordomas are described in all locations because of the rarity of this lesion. There have been few reports on chordomas of the cervical spine and suboccipital region.^{4,16,24,30,34,36} The number of cervical spine chordomas in our series reflects the particular interest of the Lariboisière Department of Neurosurgery in CCJ problems and VA surgery.

Survival of patients with chordomas varies from several months to 21 years³⁵ and even 46 years.²⁴ Recently Watkins, et al.,³³ have suggested that patients with chordomas be classified into one of two groups: "one group with high mortality within 5 years and a second group with an indolent disease process and near normal life expectancy;" however, there are no well-established criteria to permit such classification. Therefore, many authors recommend aggressive resection.^{1,9,11,28,33} Some authors recommend complex combined surgical approaches, claiming that the rates of morbidity and mortality are acceptable.^{1,11,28} They underscore also the fact that reoperations are generally more difficult, less efficient, and pose greater risk. Greater risk is also introduced if radiotherapy has already been conducted.¹¹ At the cervical level, the posterior laminectomy and the anterior approach are standard approaches. These approaches cannot reach the posterolateral corner of the VBs nor the transverse processes with the VA. The lateral approach, which we used in this series, is the only surgical route specifically designed to expose these structures. It also permits resection of any tumor that extends into the VBs as well as extension toward the spinal canal by using an oblique corpectomy technique.^{13,15} At the CCJ, standard approaches include the posterior approach and transoral approach. Again, however, these approaches place the lateral part of the CCJ out of reach. At this level, the lateral approach can be performed anteriorly (the anterolateral approach) or posteriorly (the posterolateral approach).^{12,14} In the anterolateral approach the upper extent of the lateral approach is provided at the cervical level. It can expose the CCJ from the anterior to the posterior midline. Even if it provides access to an intradural extension, the anterolateral approach is more specifically designed for lesions that develop in the extradural space and the bone elements. Conversely, in the posterolateral approach the lateral extent of the midline posterior approach is provided; this approach is specially designed for intradural lesions.¹⁴ In fact, the posterolateral approach is rarely indicated due to the typical development of chordomas. It was used only once in our series, after an anterolateral approach on the contralateral side; the lateral or the anterolateral approach was used in 30 patients because a lateral tumor extension with VA involvement occurred in 30 (83%) of 36 patients. This probably does not reflect the true incidence of the lateral extension of cervical chordo-

mas because our experience is certainly biased by the special referring pattern of our department.

In all patients with recurrent tumors a lateral location was demonstrated. This probably reflects an incomplete resection at this level during the primary treatment. We always used one or two lateral approaches in these patients with recurrent disease. In patients with primary tumor, only six had strictly anterior midline development and were treated via an anterior or a transoral approach. In the 16 other patients there were anterior and lateral developments that always required one or two lateral approaches either alone or combined with a transoral approach (four patients).

In our series outcome was much better in the primary tumor group in which patients benefited from a resection at first presentation. Disease-related mortality was 20% in this group compared with 57% in the recurrence group. The mean delay before recurrence was 34 months in the primary tumor group (an average recurrence rate of 0.13 per year) whereas it was 15.2 months in the recurrence group (an average recurrence rate of 0.64 per year).

All but two patients with primary tumor underwent radiotherapy, including 10 who underwent proton-beam radiotherapy (seven in the primary tumor group and three in the recurrence group). Although radiotherapy has been very controversial for a long time, it is was demonstrated to have a dose-related efficiency.^{22,25} At least 60 Gys are necessary to obtain a therapeutic effect on chordomas. Proton-beam radiotherapy permits the delivery of a high energy to a defined and limited area. Therefore, it has some advantages over conventional radiotherapy.^{2,19,29} Radiosurgery is another technique with advantages similar to proton-beam radiotherapy; the initial reported results have been good.²² Even these new techniques are limiting by the tolerance of anatomical structures to radiation. Especially high doses of radiation, regardless of the modality, cannot be used near by the brainstem or the spinal cord. This is why aggressive resection of chordomas, especially those located in the vicinity of the nervous tissue seems preferable.

CONCLUSIONS

Because there remains no definitive way to differentiate chordomas with benign evolution from those with rapid growth and invasive features, aggressive treatment with as radical a resection as possible followed by radiotherapy is recommended.

Cervical chordomas very often extend laterally toward the VA; those that do pose difficult surgical problems. We have shown that the lateral route with VA exposure and control is the approach of choice even though an additional approach to achieve extensive tumor resection and/or spinal fusion was necessary in almost 50% of the cases.

The resection of primary tumor provides better results than that of recurrent tumors. The results of our series underscore the need for extensive resection at a patient's first presentation.

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