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Endoscopic endonasal approach for skull base chordoma



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Abstract

Background Chordoma is a rare slow-growing malignant bone tumor that arises from embryonic notochordal remnants in the axial skeleton. Clival chordomas have a high propensity for extension through the skull base and a high proclivity for recurrence. Recently, resection of skull base chordomas through the endoscopic transnasal approach has become the standard way of managing these lesions with the new advancements in neuronavigation and reconstructive techniques in skull base surgery.

Methods It is a retrospective study of all patients with clival chordoma that were operated upon using endoscopic endonasal resection at the Main Alexandria University Hospital during the period from March 2008 to April 2013. The extent of surgical resection was assessed intraoperatively and confirmed by the postoperative MRI study performed 8–12 weeks, 6 months, and yearly after the surgery.

Results Twelve patients with clival chordoma were managed by endoscopic transnasal resection. Gross total resection confirmed by postoperative MRI was achieved in four cases, subtotal resection in six cases, and partial resection in two cases of clival chordoma. In cases where gross total resection was not achieved, residual tumors were adherent to vital neurovascular structures as confirmed with the utilization of an intraoperative imaging navigation device. A study of postoperative complications and possible recurrence was done for all cases.

Conclusions The findings of this study highlight the significance of gross total resection as a major determinant for preventing the recurrence of chordoma. Our findings also support the validity of the endoscopic approach for the management of clival chordoma especially when the approach is tailored based on the site and extent of the tumor.

Keywords Clival chordoma, GTR, MRI, Endoscopic transnasal, Endoscopic transoral

Background

Chordoma is a rare slow-growing malignant bone tumor that arises from embryonic notochordal remnants in the axial skeleton. It is usually presented in the third to fourth decades of life in the spheno-occipital region and around the fifth to sixth decades in the Sacro-coccygeal region [1, 2]. By virtue of their aggressive behavior, clival chordomas have a high propensity for extension through the skull base and a high proclivity for recurrence [3, 4].

Even with the recent technological advances and the emergence of new surgical approaches, effective and safe surgical resection of clival chordomas still poses a challenge to skull base surgeons. Transcranial, transfacial, and transnasal transsphenoidal approaches were used for chordoma resection [4–8]. Recently, the resection of skull base chordomas through the endoscopic transnasal approach has become more popular thanks to continuous



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improvements in various neuroendoscopic equipment and skull base reconstruction techniques [9–12].

This study retrospectively analyzed the outcome of 12 cases of clival chordomas that underwent endoscopic endonasal resection as a recommended treatment for these patients.

Methods

Twelve patients with clival chordoma were operated upon for endoscopic endonasal resection of clival chordoma at the Main Alexandria University Hospital during the period from March 2008 to April 2013. A surgical team included both an otolaryngologist and a neurosurgeon performing all procedures using 4 hands technique. Histopathological confirmation was available in each case. The extent of surgical resection was assessed intraoperatively and confirmed by the postoperative MRI study performed 8–12 weeks after the surgery. The second follow-up MRI was obtained after 6 months and then 12 months thereafter. The follow-up period ranged between 24 and 60 months for all cases.

The extent of tumor resection was categorized into total, subtotal, and partial resection. Total resection of the tumor was defined as complete surgical excision intraoperatively with no residual tumor on the postoperative MRI. Incomplete tumor resection was categorized as subtotal resection when less than 10% of the tumor was left behind and partial resection when more than 10% of the tumor was left as residual depending upon postoperative MRI (Fig. 1). Postoperative adjuvant radiotherapy was not administered to any of our patients.

Surgical technique

The surgical intervention was conducted under general anesthesia. The patients were positioned supine on the operating table with the neck supported and slightly flexed to facilitate surgical access and decrease intraoperative bleeding. Decongestion of the nasal cavity was achieved using cottonoid pledgets soaked with xylometazoline and left for 5 min in the nasal cavity. Intraoperative blood loss was reduced by inducing controlled hypotension and bradycardia as well. Visualization was facilitated by the use 0.30 and 45° Hopkins rod scope coupled to a high-definition camera and monitor (Storz Endoscopy, Tuttlingen, Germany). The surgical procedure was performed using skull base/neurosurgical endoscopic instruments (Storz), a high-speed drill with an angled handpiece, and diamond burrs.

The surgical approach was dictated by the clival region that was involved in the tumor and its contagious extension. Patients with clival chordoma involving the upper 2/3 were approached via a transnasal Para septal endoscopic approach. If the ethmoid labyrinth was involved, the approach was extended to perform ethmoidectomy. The involvement of the lower 1/3 clivus was approached using the trans-oral endoscopic approach via the oral cavity and oropharynx to ensure adequate visualization. The entire surgical procedure was conducted using a high-definition endoscopic camera system and often required a three- or four-hand technique to achieve adequate visualization, retraction, and safe resection of the tumor.

Transnasal approach

The transnasal para septal approach was initiated by excising the lower portion of the superior turbinate on each side and identifying the sphenoid sinus ostium. Sphenoidotomy was performed using mushroom forceps and enlarged on both sides with the microdebrider. A posterior septectomy was performed to widely open and communicate both sphenoid sinuses on each side. The inter-sinus septum along with the floor of the sphenoid sinuses was taken down using a high-speed drill and rongeurs to expose the Sella and upper portion of the clivus. The clival bone was drilled with a 4-mm diamond burr to achieve adequate exposure all around the tumor. The optic nerve and internal carotid artery were identified before tumor resection to avoid inadvertent injury to these structures during tumor resection.

Transoral approach

In trans oropharyngeal endoscopic approach, a Davis Boyle mouth gag was used to open the mouth, the palate was either retracted or split to achieve adequate exposure and a 45-degree angled endoscope was used to visualize the lower clivus. An inferiorly based mucosal flap was designed to approach the lower clivus and replaced in position at the end of the procedure to enhance mucosal healing.

Once all the margins of the tumor and the vital structures were identified, safe resection of the extradural tumor was achieved in a centrifugal fashion. In tumors with subdural extension, the basilar arteries and brainstem were carefully dissected using the four-hand endoscopic technique to safely excise the subdural component. Tumor tissue abutting close to the optic nerve, internal carotid artery, and sellar floor was removed at the end of the procedure. All the tumor tissue that could be resected without breaching vital structures was excised and sent for histopathological examination. At the end of the surgical procedure, adequate hemostasis was achieved using bipolar diathermy, and the surgical cavity was packed with oxidized cellulose (Surgicel). If surgical resection resulted in a dural defect, it was repaired in a multilayered fashion using fat, Fascia Lata, and a naso-septal flap. Lumbar drain was inserted at the end of the procedure



(A)



(B)

Fig. 1 Gross total resection of clival chordoma. A Preoperative MRI. B Postoperative MRI showing gross total resection

to help decrease CSF pressure postoperatively, which helped in the success of repair done in such cases.

Results

Twelve patients with clival chordoma were treated with endoscopic transnasal resection. 8 of them were females and other 4 were males, whose ages ranged from 25 to 65 years with a mean age of 44 years. All patients reported headaches. Eight cases were presented with nasal obstruction while 5 of them had dysphagia at clinical presentation. Manifestations of cranial nerve VI and diplopia were present in 6 cases and cranial nerve II affection resulting in visual dysfunction was present in 6 cases, while lower cranial nerves IX, X, and XI were affected in 4 cases. The clinical data of the patients and the extent of the tumor were summarized in Table 1.

According to pre-operative imaging, there was a single case with a tumor involving only the upper clivus, two cases with a tumor confined to the lower clivus, four cases with a tumor extended to the mid clivus, two cases in upper and mid clivus and three cases involving the three regions. Cavernous sinus invasion or carotid artery encasement was detected in six cases.

Gross total resection confirmed by postoperative MRI was achieved in four cases, subtotal resection in six cases, and partial resection in two cases of clival chordoma. In cases where gross total resection was not achieved, residual tumors were adherent to vital

	Age	Sex	Site	Headache	CN affection	Nasal obstruction	Dysphagia	CS invasion/ICA encasement	Resection
1	54 years	F	Lower clivus	+			+		GTR
2	60 years	F	Mid clivus	+		+			GTR
3	45 years	М	Upper, mid, and lower clivus	+	II, VI, IX and X	+	+	+	Subtotal
4	37 years	М	Lower clivus	+	IX, X, XI		+		Partial
5	56 years	F	Mid clivus	+		+			Subtotal
6	25 years	Μ	Mid clivus	+		+			GTR
7	28 years	М	Mid clivus	+		+			subtotal
8	60 years	F	Upper and mid clivus	+	II and IV	+		+	Subtotal
9	55 years	F	Upper and mid clivus	+	II and IV			+	Subtotal
10	50 years	F	Upper, mid, and lower clivus	+	II, VI, IX, X, XI	+	+	+	Subtotal
11	65 years	F	Upper, mid, and lower clivus	+	II, VI, IX, X, XI	+	+	+	GTR
12	47 years	F	Upper clivus	+	II and IV			+	Partial

Table 1 The clinical data and extensions of studied patients with clival chordoma

CN Cranial nerve, CS Cavernous sinus, ICA Internal carotid artery, GTR Gross total resection

neurovascular structures as confirmed with the utilization of an intraoperative imaging navigation device.

Blood loss during the surgical procedure ranged from 250 to 950 ml depending on the extent of the tumor resection; with an average of 600 ml. Intraoperative bleeding originated primarily from the cavernous sinus and was controlled by oxidized cellulose (Surgicel) without the need for blood transfusion.

Intraoperative CSF leakage was encountered in two of the patients who had intradural extension of the tumor (cases no. 3 and 12). Dural defect in both cases was < 2 cm and repair was accomplished in a multilayered fashion using fat, fascia lata, and nasoseptal flap.

There was no postoperative mortality in our series. The postoperative complications included nasal dryness in 4 patients (cases no. 3, 8, 9, and 10) and palatal fistula in one patient (case no. 3) which was successfully repaired in another separate surgical procedure. None of our patients developed diabetes insipidus, hydrocephalus, infection, secondary hemorrhage, or new neurological or visual deficits. Cranial nerve dysfunction did not improve postoperatively except for some improvement in the visual field of vision in cases no. 8 and 12 who experienced modest improvement 2 days postoperatively.

During the follow-up period which extended up to 24–60 months, no recurrence was observed when the tumor was completely excised (cases no. 1, 2, 6, and 11). Subtotal and partial resection was associated with an insidious growth of the residual tumor in all cases

and necessitated a revision surgery in 3 cases (cases no. 4, 8, and 12).

Discussion

Chordomas are locally malignant tumors of notochordal remnants that tend to metastasize late in the course of the disease [1, 13]. Complete surgical resection of clival chordomas is considered the best treatment option but is often hampered by poor surgical access and involvement of vital structures. Several surgical approaches have been described for the resection of chordomas including transcranial approaches [14, 15], transsphenoidal microscopic approach [8, 16, 17], trans oropharyngeal microscopic approach [18], and trans facial approaches [19, 20]. Each of these approaches has its merits and demerits; however the endoscopic approaches are getting more and more popular as they use normal apertures, carry less risk of morbidity, and provide a magnified panoramic surgical view even around corners [11, 20–23].

In the present study, the surgical approach has been tailored based on the part of the clivus involved and the extension of the tumor in an attempt to minimize morbidity, enhance visualization, and preserve vital anatomic structures. This individualization of the surgical approach for each patient has provided excellent surgical exposure, minimal morbidity, and maximal removal of the tumor without placing critical neurovascular structures in jeopardy.

Clival chordomas have been categorized based on the site of involvement of the clivus into upper, mid, and lower clival chordomas [10, 24]. Surgical access to clival chordomas is dictated by the site of origin of the tumor as well as the visualization provided by the surgical approach. The transnasal para septal endoscopic approach provides a panoramic view of the Sella, parasellar cavernous sinus, ICA, optic nerve as well as upper and mid clivus. It can be extended to tackle tumor extension in the ethmoid and provide adequate access to the posterior ethmoid roof. However, this approach is not particularly suited to manage tumor extension in the lower clivus. The trans-oropharyngeal approach provides adequate visualization of the lower clivus and ventral craniocervical junction but does not expose the upper and mid clivus adequately. Combined transnasal and transoral approaches can provide excellent surgical access to the entire clivus and with the aid of panoramic view provided by angles endoscopes, dissection in the vicinity of vital structures is unhampered [13].

Satio et al. [5] reviewed the results of case series of endoscopic resection of clival chordomas published in the literature. They reported that the major complications encountered during resection of clival chordomas included cranial neuropathies, internal carotid artery injury, stroke, intracranial hematoma, and hydrocephalus. The median rate of gross total resection was 70.4% and the median rate of postoperative CSF leakage was 14%. Gross total resection of clival chordoma was achieved in 4 of our patients (33%) and is comparable with the results achieved at other institutions. The overall gross total resection (GTR) rate reported in the literature by conventional as well as endoscopic approaches varies from 32.5 to 100%. The results of the case series published in the literature on the resection of clival chordomas are summarized in Table 2.

Gross total resection of clival chordomas (GTR) provides the best long-term survival but is often associated with injury to adjacent vital structures and catastrophic complications especially when the dissection is carried out around the corners without adequate visualization [24]. No major complications were encountered in small case series and when sequential intraoperative MRI was used [26, 28, 33]. In the present study, no major postoperative complication was encountered. Two of the patients had subdural extensions that necessitated resection of the dura and subdural resection of the tumor. Dural defects were successfully repaired in a multilayered fashion and none of the patients developed postoperative CSF leaks.

Although adequate visualization of the lateral portions of the tumor was attained in all cases in the present study, tumor adhesion to vital neurovascular structures was a limiting factor hindering complete excision. Clival chordomas often displace, encase, and infiltrate vital structures such as the brain stem and

Author(s), year	Patients	GTR	Major complications		
Jho and Ha. [25], 2004	3	100%	CSF leak, PE, AF, and Stroke (33%)		
Solares et al. [26], 2005	3	66.7%	_		
Frank et al. [27], 2006	9	33%	ICA injury (11%)		
Hwang and Ho. [28], 2007	3	100%	_		
Carrabba et al. [29], 2008	60	59%	Hematoma, hemiparesis, and hydrocephalus (6%) CSF leak (24%) Tension pneumocephalus (6%)		
Zhang et al. [30], 2008	7	85%	Subarachnoid hemorrhage (14%)		
Dehdashti et al. [31], 2008	12	58%	Hemiparesis (8%) CSF leak (33%)		
Hong et al. [11], 2009	9	66.7%	_		
Stippler et al. [32], 2009	20	45%	ICA injury (5%) Brainstem hemorrhage, quadriparesis, and lower cranial neuropathy (5%)		
Holzmann et al. [33], 2010	13	92%	_		
Tan et al. [34], 2012	14	50%	Basilar artery injury (7%) Hydrocephalus (7%) Pneumocephalus (7%) CSF leak (21%)		
Satio et al. [5], 2012	6	50%	Hydrocephaluls and brain stem infarction (16%)		
Ouyang et al. [35], 2014	77	32.5%	Cerebral infarction (3.2%) Lower cranial nerve palsy (4.8%) CSF leak (4.8%)		

 Table 2
 A summary of complications following clival chordoma surgery reported in literature

critical neurovascular structures. In such cases, gross total resection does not constitute the ideal surgical procedure to circumvent incapacitating postoperative sequelae [6, 35]. In the present study, gross total resection was attempted in every case provided that this would not jeopardize the postoperative functional status of the patient which seems to be a factual strategy to attain maximal tumor resection with minimal morbidity. The gross total resection achieved in this study is reasonable when correlated with the low complication rate. The low morbidity encountered in this study has been achieved by cautious dissection of the tumor adjacent to vital structures which was facilitated by the adequate visualization provided by individualization of the surgical approach. The angled endoscopes combined with the high-definition camera in addition to the use of an intraoperative imaging navigation system provided exquisite anatomical details of vital structures, the ability to see around corners, and offered an additional advantage of using the four-hand technique. This strategy has eliminated the development of major postoperative complications in our patients.

During the follow-up period of this study which extended to 60 months, all patients survived except one (case no. 3). This patient was a revision case who had been biopsied in another institution 1 year prior to the surgical intervention at our institution. The patient died 2 years after subtotal tumor excision because of second-ary regrowth of the tumor close to the basilar artery which resulted in brain stem compression. Regrowth of a tumor after initial surgical intervention has been associated with a poor outcome which has been partially attributed to the aggressive biological behavior of the recurrent tumor [1, 34, 36].

Conventional radiotherapy has a significant effect on prolonging progression-free survival, but does not increase the overall survival rate; hence, the use of conventional photon radiation in chordomas has been negated [35]. Chordomas are resistant to radiotherapy requiring very high doses of radiation to achieve control of these tumors. The proximity of clival chordomas to vital radiosensitive structures in addition to the high risk of complications associated with operating on tumor recurrence in irradiated patients limits the routine use of conventional radiation therapy [4, 13, 17].

The superiority of proton beam therapy has been highlighted in some studies [35]. Recently, Jahangiri et al. [24] challenged the concept that the type of radiation has an influence on recurrence. According to their results, there is no benefit of proton-based over photon-based radiation, contradicting conventional presumptions and underscoring the need for randomized trials addressing the efficacy of each radiation modality. Surgical intervention in patients who receive radiotherapy is associated with a high risk of injury to intracranial vascular and neural structures; therefore, we adopted a close follow-up policy to surgically salvage tumor regrowth reserving radiotherapy for patients who refused surgical intervention or were otherwise medically unfit [7, 17, 37]. A similar strategy was adopted by Maira et al. [17] who questioned the efficacy of radiotherapy in the management of chordoma. Residual tumor was associated with its regrowth in all our patients and four patients (25%) required a second-stage surgery (cases 4, 8, and 12).

The recurrence of the tumor after resection and survival have been well correlated with the extent of initial tumor resection. Al-Mefty and Borba [4] achieved gross total resection (GTR) in 10 patients (43.5%), subtotal resection in 11 patients, (47.8%), and partial resection in two patients (8.7%). Recurrence was observed in five patients, two of whom had recurrence within the first postoperative year. Forsyth et al. [38] reviewed 51 patients with chordomas. Based on the extent of removal of the tumor, 11% of the patients were biopsied and 78% of the tumors were subtotal excised. The survival rates for patients who underwent biopsy were 36% at 5 years while for those who had subtotal resections, it was 55% at 5 years. Gay et al. [7] reported their outcomes on 46 patients with chordomas. The tumor was grossly removed in 67%, subtotally in 23%, and partially in 10% of the patients. The recurrence-free survival rate of patients with skull base chordomas at 5 years was 65%. Recurrence was observed in 6 patients during a followup period ranging from 1 to 11 years with a median of 3.9 years. They concluded that gross total resection was associated with a lower risk of recurrence. Maira et al. [17] reported that after gross total resection, no recurrence was noted at 14 to 86 months (mean 37.5 months). At the time of analysis, no recurrence was observed in any of the patients who had undergone gross total resection (GTR). However, we are fully aware that a longer follow-up period is required to exclude recurrence.

Conclusion

The findings of this study highlight the significance of gross total resection as a major determinant for preventing the recurrence of chordoma. We also emphasize that gross total resection is often impossible to achieve without incurring significant morbidity to the patient. In such cases, a more conservative resection is a logical policy and tumor regrowth can be salvaged in a secondstage surgery. Our findings also support the validity of the endoscopic approach for the management of clival chordoma especially when the approach is tailored based on the site and extent of the tumor. An individualized endoscopic approach should be a prominent technique in the armamentarium of skull base surgeons that can achieve maximal tumor resection with minimal morbidity.

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Authors' contributions

AY: is main and corresponding author. HM: is a main author who participated in surgical cases. RB: is a main author who helped with the study design and literature review. AI: is a main author and surgeon. AF: is a main author and surgeon for cases from the neurosurgery team. MA: is a main author who helped with study design and literature review. All authors read and approved the final manuscript.

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Availability of data and materials

The dataset generated and/or analyzed during the current study is available from the corresponding author upon request.

Declarations

Ethics approval and consent to participate

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The article has been reviewed by the ethics committee of Alexandria University and has been approved with IRB No: 00012098. All patients have given full informed consent for participation.

Consent for publication

Written consent has been obtained from the patients for using their information for publication.

Competing interests

All authors declare that they have no competing interests.

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