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Conflict of interest

The authors declare that they have no conflicts of interest

Keywords

carotid body tumor; chemodectoma; neck paraganglioma; vascular formation; surgical treatment; neck surgery

CAROTID BODY TUMORS: SURGICAL TREATMENT AND PERIOPERATIVE RISK FACTORS OF ADVERSE EVENTS

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Abstract

A carotid body tumor (CBT), also commonly known as nonchromaffin paragangliomas and chemodectomas, is a slowly growing neoplasm originating from carotid body chemoreceptors. **Objectives:** To present the possible predictors of adverse events in the treatment of carotid body tumors. **Materials and methods.** The study was performed on basis of Scientific Central Surgery named after A.N. Syzganov. Only 8 patients with 8 tumors who underwent open surgical treatment of CBT over the past 10 years (from 2009 to 2019). Results. 6 (75%) patients had a painless mass in the neck, and 2 (25%) painful. According to the results of CTA and arteriography, the patients were classified according to the Shamblyn classification (Figure 1), of which 1 patient had the I type of tumor (12.5%), type II in three (37.5%) and type III (50%) 4 patients, one (12%) of which revealed hemodynamically significant stenosis of bifurcation of the common carotid artery (CCA). The average number of days a patient was in the hospital stay increased direct proportional difference and the type of tumor. The time and approach of the operation, intraoperative blood loss, the volume of blood transfusion increased in the same way as the size of the tumor increased, involvement of the carotid artery and malignancy of the tumors. One patient had a cranial nerve injury (temporal), which resolved after additional treatment. Pathology results showed that two patients (25%) had malignant cells in the tumor and one of them located in the lymph nodes. **Conclusion.** A rare occurrence, slow growth, asymptomatic course and a limited amount of information on the detection and management of the disease lead to an underestimation of the condition. In addition, the large size of the tumor, the involvement of the carotid arteries and cranial nerves in the process directly affect the surgical approach and increase the risk of complications.

Хемодектомалар: хирургиялық емдеу тәжірибесі және асқынулардың периоперативті қауіп факторлары

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Аңдатпа

Хемодектомалар, сонымен қатар хромоафинді емес параганглиомалар деп те аталады, бұл каротид синусының хеморецепторларынан шыққан баяу өсетін неоплазмалар. Параганглиомаларға 30 000 арасында мойын массасының шамамен 1-і кездеседі, оның ішінде 45%-ы хемодектомалар. **Жұмыстың мақсаты** ретроспективті талдау жүргізу және хемодектоманың емдеудің хирургиялық тактикасын көрсету болды. **Материалдар мен тәсілдер.** Зерттеу А.Н.Сызғанов атындағы Ұлттық ғылыми хирургия орталығының ангиохирургия бөлімінде жүргізілді. Жалпы алғанда, соңғы 10 жыл ішінде (2009 жылдан 2019 жылға дейін) хемодектомамен хирургиялық емдеуден өткен 8 формациясы бар 8 пациенттің ауру тарихы зерттелді. Нәтижелер. 6 (75%) науқаста мойын аймағында ауырсынусыз ісік тәрізді түзіліс байқалды, 2 (25%) пациентте ауырсыну пайда болды. КТ ангиографиясының нәтижелері бойынша науқастар Шамблин ұсынған классификацияға сәйкес бөлінді, оның I

типті ісігі бір науқаста (12,5%), II типте үшеуінде (37,5%) және III типте (50%)) 4 пациент болды, олардың біреуі (12, %) жалпы ұйқы артериясының бифуркациясының гемодинамикалық маңызды тарылуы болды. Науқастардың стационарда жатқан күндерінің орташа саны ісік мөлшері мен түріне тікелей пропорционалды түрде өсті. Ісік мөлшерінің ұлғаюымен, каротид артерияларының процеске араласуымен және түзілудің қатерлі ісіктерімен бірге жүргізілген операцияның көлемі, операцияның уақыты, қан жоғалту, қан құю көлемі де өсті. Операциядан кейінгі кезеңде бірнеше рет араласқан бір пациенттің уақытша жұтылу бұзылысы болды, ол физиотерапияны қолданғанда симптомдар жойылды. Гистологияның нәтижелері көрсеткендей, екі пациенттің (25%) қатерлі жасушалары бар болды, олардың біреуі лимфа түйіндеріне метастаздармен. **Қорытынды.** Операцияға дейінгі кезеңде пациенттің жағдайын және ықтимал көпсалалы тәсілі бар науқастың қосарланған фонын, әсіресе Шамблин классификациясы бойынша III типті науқастарда толық бағалау жүргізу ұсынылады. Сирек пайда болу, баяу өсу, асимптоматикалық ағым және ауруды анықтау туралы шектеулі ақпарат жағдайды бағаламауға әкеледі. Ісіктің үлкен мөлшері болған кезде, ұйқы артериялары мен бас сүйек нервтерінің қатысуы хирургиялық тәсілге тікелей әсер етеді және асқын қаупін арттырады.

Түйін сөздер

хемодектома; каротидті параганглиома; мойын ісіктері; тамырлы түзілімдер; хирургиялық ем; мойын хирургиясы

Хемодектомы: опыт хирургического лечения и периоперативные факторы риска осложнений

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Аннотация

Хемодектомы, так же широко известные как нехромоафинные параганглиомы, это медленно растущие новообразования, происходящие из хеморецепторов каротидного синуса. Приблизительно 1 из 30000 образований головы и шеи соответствует параганглиоме, 45% из которых являются хемодектомами. **Целью** работы было провести ретроспективный анализ и показать опыт нашего центра в менеджменте и хирургической тактике лечения хемодектом. **Материалы и методы.** Исследование проводилось на базе отделения ангиохирургии Национального Научного Центра хирургии имени А.Н. Сызганова. Всего ретроспективно были изучены истории болезни 8 пациентов с 8 образованиями, перенесших оперативное лечение хемодектом за последние 10 лет (с 2009 по 2019 годы). Результаты. У 6(75%) пациентов отмечалось безболезненное опухолевидное образование в области шеи, у 2(25%) болезненное. По результатам КТ-ангиографии пациенты были распределены по классификации, предложенной автором Shamblin, из которых I тип опухоли был у одного пациента (12,5%), II тип у троих (37,5%) и к III (50%) типу относились 4 пациента, у одного (12, %) из которых было выявлено гемодинамически значимое сужение бифуркации общей сонной артерии. Среднее число дней нахождения пациентов в больнице повышалась прямопропорционально размеру и типу опухоли. Объем проведенной операции, время операции, интраоперационная кровопотеря, объем гемотрансфузии повышались так же по мере увеличения размеров опухоли, вовлечения в процесс сонных артерий и злокачественности образования. В послеоперационном периоде у одного пациента с повторным вмешательством было временное нарушения глотания, которые с применением физиолечения симптомы были разрешены. Результаты гистологии показали, что у двоих пациентов (25%) были обнаружены злокачественные клетки, у одного из них с метастазами в ближайшие лимфоузлы. **Заключение.** В предоперационном периоде рекомендуется провести полную оценку состояния пациента и коморбидного фона пациентка с возможным мультидисциплинарным подходом, в особенности у пациентов с III типом по классификации Shamblin. Редкая встречаемость, медленный рост, бессимптомное течение и ограниченный объем информации по выявлению заболевания приводят к недооценке состояния. Тогда как большие размеры опухоли, вовлечение в процесс сонных артерий и черепно-мозговых нервов напрямую влияют на хирургический подход и увеличивают риск осложнений.

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Конфликт интересов

Авторы заявляют об отсутствии конфликта интересов

Ключевые слова

хемодектома; каротидная параганглиома; образования шеи; сосудистые образования; хирургическое лечение; хирургия шеи

Introduction

A carotid body tumor (CBT), also commonly known as nonchromaffin paragangliomas and chemodectomas, is a slowly growing neoplasm originating from carotid body chemoreceptors [1,2]. It occurs in 0.5 - 0.5% of cases of head and neck zone formations [3,4]. About 1 in 30,000 head and neck formations corresponds to a paraganglioma, 45% of which are CBT. It was found that bilateral location occurs in 5% of sporadic cases, while in family cases the percentage of CBT on both sides reaches

30% [5]. The frequency of occurrence in women is 2.5-3 times higher than in men [4.6]. CBT occurs at any age, but is usually diagnosed between the third and sixth decades of life [7].

Malignant chemodectomas are observed in 15–20% of patients [8]. In various observations, from 2–9%, the carotid chemodectoma can be malignant with the formation of metastases [8]. The presence of metastases to the lymph nodes, as well as the presence of distant metastases, indicates a malignant nature of the tumor and has an unfavorable prognosis [9]

The first anatomical mention of a carotid glomus tumor was made by Albercht von Haller in 1743 [10]. The first attempt at surgical removal of a chemodectoma was made by Reigner in 1880, which culminated in the death of the patient [11]. In 1886, Maydl first performed a successful operation to remove a chemodectoma, with a subsequent complication in the form of aphasia and hemiparesis [12]. The first operation to remove the tumor with the preservation of the internal carotid artery and without the development of complications was performed in the USA by Scudder in 1903 [13]. Since 1970, with the development of cardiovascular surgery technologies, the widespread introduction of neuroimaging methods, the mortality rate due to surgical removal of a chemodectoma has been less than 10% [14,15].

Treatment is primarily based on surgical removal. A significant role in the choice of surgical treatment approaches was played by a study by a group of authors Shamblin et al, who proposed their CBT classification based on carotid artery involvement. The first group is CBT without involvement of the carotid arteries, in this position it is easy to remove the tumor from the vessels, the second group has partial involvement of the vessels, in this category partial excision of external layer of the artery is possible, the third group has complete involvement of the vessels and transmural invasion of the tumor, in this case, tumor resection is required along with the carotid arteries due to the inability to separate them from each other [16].

Despite the fact that monitoring of undesirable consequences during surgery, improvement of surgical technique and postoperative monitoring yielded results and reduced the risk of mortality closer to zero, the percentage of postoperative neurological complications remains high and reaches from 10 to 40%. However, due to the locally invasive nature of CBT, the surgical approach is still effective [17-21]. Along with surgical treatment in the literature, there are publications by authors suggesting transarterial embolization (TAE) in the preoperative period, which will reduce the risk of complications [22,23].

Objectives

To present the possible predictors of adverse events in the treatment of carotid body tumors.

Materials and methods

The study was conducted on the basis of the Department of Vascular surgery of the National Scientific Center of Surgery named after A.N. Syzganov. In total, case histories of 8 patients with 8 formations undergoing open surgical treatment of CBT over the past 10 years (from 2009 to 2019) were retrospectively studied. After signing the informed consent, the patients were invited for examination and questionnaire, including complaints, family history, diagnostic tests, type of treatment and others. The search for relevant literature was carried out in the databases Web of Knowledge, Scopus, PubMed, Google scholar, Cochrane Library. Collection of all patient data, statistical calculations were performed using Microsoft Excel.

Table 1.
Main characteristics of patients

Features of patient	Mean or No (%)
Age, (years)	45,4±12,6 (range 31-73)
Gender male/female (n)	2(25)/6(75)
Presentation	
• Painless lump	6(75)
• Painful lump	2(25)
• Dysphagia	1(12.5)
• Horner's syndrome	1(12.5)
• Headache	4(50)
• Stroke/TIA	0/0
• Arterial hypertension	3(37.5)
• Previous operation	2(25)
• Family history	0(0)
• Thyroid neoplasm	1(12.5)
Preoperative imagine	
• Ultrasound	8(100)
• CTA	5(62.5)
• Angiography	8(100)
Shamblin classification	
• Type I	1(12.5)
• Type II	3(37.5)
• Type III	4(50)

TIA, transient ischemic attack; CTA, computed tomography angiography.

Results

The total number of patients was 8, of which 6 (75%) were women, the average age was 45.4 years, and none of them had bilateral lesions. Genetic tests for the SDH gene mutation have not been conducted, but there was no familial CBT in medical history. 6 (75%) patients had a painless tumor formation in the neck, and 2 (25%) painful. It was noted that the latter had larger tumor sizes compared with asymptomatic patients, and one of them underwent CBT excision surgery 8 years ago. 3 (37.5%) patients complained of an increase in blood pressure to 140/90 mm Hg. One patient had a thyroidectomy for thyroid cancer 1 year ago. All patients underwent ultrasound duplex angioscanning, computed tomography with angiography of blood vessels (6) according to indications, as well as angiography of carotid arteries (Table 1).

According to the results of CTA and angiography, patients were classified according to the Shamblin classification (Figure 1), of which type I tumor was in one patient (12.5%), type II in three (37.5%) and type III (50%) 4 patients were treated, in one (12%) of which hemodynamically significant narrowing of bifurcation of the common carotid artery (CCA) was revealed. Also, blood serum was taken from all patients for routine laboratory tests (blood test, biochemistry, blood coagulation function, lipid profile). Laboratory test results did not reveal clinically significant changes.

After the distribution of patients into groups, a comparative analysis of the perioperative parameters of the patients was carried out (Table 2). Thus, the average number of days a patient was hospitalized increased in direct proportion to the size and type of tumor.

In addition, we performed several types of surgical treatment (Figure 2). As shown in the above table, the volume of the operation, the time of the operation, intraoperative blood loss, the volume of blood transfusion increased in the same way as the size of the tumor increased, involvement of the carotid arteries and malignancy of the formation.

In the postoperative period, one patient with repeated intervention had temporary swallowing disorders, which, with the use of physiotherapy, the symptoms were resolved.

Pathology results showed that two patients (25%) had malignant cells, one of them with metastases to the nearest lymph nodes. Patient data were subsequently referred to oncologists and radiation therapy courses were applied to them in the affected area. The average follow-up for these patients was 1 year. There were no other complications such as cerebrovascular disturbances (stroke, TIA) and death.

Discussion

CBT usually manifests itself as a painless, slowly growing neck formation, laterally to the apex of the hyoid bone [24]. Sometimes the patient complains of hoarseness, dysphagia and even less often dizziness and coughing may occur as a result of compression on the vagus or sympathetic nerves [25]. In our study, only one patient had symptoms of grade 1 dysphagia, which was presumably due to the large size of the tumor (8.0 x 6.5 cm).

As a rule, the asymptomatic course of the disease in the initial stage and the slow growth of CBT can underestimate the condition and late treatment



Figure 1. 31-year-old woman with CBT on the right: A - Computed tomography: formation of the right neck with dimensions 5.0x4.0 cm; B - arteriography: there is a hypervascular formation in the area of bifurcation of the right common carotid artery

Table 2.
Distribution of patients according to Shamblin classification

	Type I	Type II	Type III
Hospital stay, mean (days)	8±0	11,3±2,2	12,6±3,8
Tumor size, mean (cm)	4±0	5,3±1,54	6,3±2,8
Side, left/right	0/1	1/2	3/1
Bilateral	0	0	0
Type of excision			
• Excision with ligation of ECA	0	1	1
• Excision without ligation	1	0	0
• Excision and patch repair	0	2	2
• Excision and prosthesis with graft (vein)	0	0	1
• Excision with ligation of CCA	0	0	0
• Operation time, mean (min)	78±0	155±42,2	189,5±31,5
• EBL, mean (ml)	200±0	382±68,4	443±79,2
• Blood transfusion, mean (ml)	0	304±127,3	488,5±201,3
Complications			
• Hematoma	0	0	0
• Wound infection	0	0	0
• Horner's syndrome	0	0	0
• Dysphagia	0	0	1
• Cranial nerve injury	0	0	1
• Stroke/TIA	0/0	0/0	0/0
• Death	0	0	0
Pathology			
• Benign CBT	1	3	2
• Malignant CBT without metastases	0	0	1
• Metastases to lymph nodes	0	0	1

ECA, external carotid artery; CCA, common carotid artery; EBL, estimated blood loss; TIA, transient ischemic attack; CBT, carotid body tumor.

Figure 2.
31-year-old woman with CBT on the right: A - Intraoperative view of the carotid bifurcation after removal of CBT; B - View of formation after excision



of patients. So, in our center there was only one case with the first type of tumor according to the Shamblin classification, while the third type was in half of the patients (4), although many of them noted the presence of a tumor-like formation in the

neck area was sick for 10 years. Moreover, in 5 out of 8 patients, the average education size was more than 5 cm, and in two patients a malignant chemodectoma was revealed according to the results of histology.

Currently, the Shamblin classification system, which is based on the degree of involvement of the adjacent carotid arteries, is used to assess the risk of intraoperative bleeding and damage to cranial nerves and can indicate the potential need for resection of the carotid artery with revascularization [26].

Also in the literature is the work of Kim et al, who offer new predictors of complications of CBT resection. The study is based on measuring tumor volume and distance from the base of the skull (DTBOS) in conjunction with the Shamblin score [27]. According to the results of the study, each 1 cm decrease in DTBOS is associated with a 1.8-fold increase in the risk of blood loss > 250 ml by a factor of 1.8 (CI 95%, 1.25-2.55) and a 1.5-fold increase in the risk of cranial nerve damage (CI 95%, 1.19-1.92). In our study, the assessment was carried out only according to the Shamblin classification.

The prolonged existence of even a benign tumor poses a threat of stenosis of the larynx, trachea, esophagus, involvement of the cranial nerves and great vessels of the neck with impaired cerebral circulation, and its spread into the cranial cavity. Surgical resection is recommended for all CBT due to the risk of local complications associated with tumor size and malignancy [22,28,29].

Conclusion

Despite the small number of patients analyzed at our center, we came to some important conclusions and developed steps to reduce the risk of complications.

In the preoperative period, it is recommended to conduct a full assessment of the patient's condition and the patient's comorbid background with the

involvement of the necessary specialists. We also recommend preparing a dose of blood for transfusion, mainly in patients with types II and III according to the Shamblin classification. For these patients, it is recommended to perform ultrasound scanning of the great saphenous vein in order to determine the "suitability" of the venous graft for prosthetics. In the case of a decision to resect the carotid artery or when there are prerequisites for prolonging the time of surgery, it is recommended to establish a temporary intravascular shunt (TIVS) in order to reduce the risk of cerebral ischemia. In case of detection of malignant cells in a tumor or neighboring lymph nodes, it is recommended that an oncologist is monitored immediately after surgery for the possibility of radiation or chemotherapy. After surgery, it is recommended that the patient is monitored at the ICU to monitor blood pressure and function of vital organs. For patients who underwent excision of the tumor with resection and subsequent prosthetics, or excision of the vessel wall with patching, it is recommended to perform ultrasound for the first 48 hours, as well as monitoring the patient's neurological status after waking up. We recommend that you allow the patient to drink water after waking up in small sips strictly in the presence of a doctor in order to determine possible damage to the cranial nerve.

Thus, the rare occurrence, slow growth, asymptomatic course and a limited amount of information on the detection and management of the disease lead to an underestimation of the condition. In addition, the large size of the tumor, the involvement of the carotid arteries and cranial nerves in the process directly affect the surgical approach and increase the risk of complications.

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