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SURGICAL TREATMENT OF AGGRESSIVE HEMANGIOMAS OF THE TRANSITIONAL CERVICAL-THORACIC SPINE IN CHILDREN (CLINICAL OBSERVATIONS, PRELIMINARY RESULTS)

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Introduction. Vertebral hemangioma is a common pathology, in which 3.7% cases are aggressive. One of the pathogenetic factors contributing to the growth of vertebral hemangioma is mechanical overload. The transitional parts of the spine are the most loaded. Lesions of the transitional cervical-thoracic spine by hemangioma are rare (2%–4% of all vertebral hemangiomas). A common treatment for aggressive hemangiomas is puncture vertebroplasty. Currently, the number of pediatric patients with vertebral hemangiomas has increased, with an incidence close to 8% for individuals aged below 18 years. Exaggeration of the possibilities of conservative treatment for hemangiomas in children often leads to unsatisfactory results. Clinical research on this problem is relevant due to the lack information about the surgical tactics in aggressive spinal hemangiomas in children.

Clinical observation. Two patients aged 15 and 17 years old with aggressive hemangiomas of the transitional cervical-thoracic spine underwent operation with the use of open-puncture vertebroplasty. There were no postoperative complications, and good preliminary results were obtained.

Discussion. Various approaches in the treatment of aggressive hemangiomas of the transitional cervical-thoracic spine in children, including open-puncture vertebroplasty, were presented and analyzed.

Conclusion. Due to the limited information about surgical treatment for pediatric vertebral hemangiomas, the presented clinical cases of surgical treatment for aggressive hemangiomas of the transitional cervical-thoracic spine may be of interest to a wide audience.

Keywords: spine; hemangioma in children; cervical-thoracic transition; vertebroplasty.

ОПЕРАТИВНОЕ ЛЕЧЕНИЕ АГРЕССИВНЫХ ГЕМАНГИОМ ПЕРЕХОДНОГО ШЕЙНО-ГРУДНОГО ОТДЕЛА ПОЗВОНОЧНИКА У ДЕТЕЙ (КЛИНИЧЕСКИЕ НАБЛЮДЕНИЯ, ПРЕДВАРИТЕЛЬНЫЕ РЕЗУЛЬТАТЫ)

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Введение. Гемангиомы позвоночника — достаточно распространенная патология, 3,7 % из них имеют агрессивный характер. Одним из патогенетических факторов роста гемангиомы тела позвонка считается его механическая перегрузка. Наибольшую нагрузку испытывают переходные отделы позвоночника. Поражение гемангиомой переходного шейно-грудного отдела позвоночника встречается достаточно редко (2–4 % случаев всех гемангиом позвоночника). Распространенным методом лечения агрессивных гемангиом является пункционная вертебропластика. В настоящее время наблюдается тенденция к увеличению числа пациентов детского

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возраста, частота встречаемости гемангиом в возрасте до 18 лет приближается к 8 %. Переоценка возможностей консервативного лечения гемангиом у детей зачастую приводит к неудовлетворительным результатам. Клиническое исследование по данной проблеме актуально в связи с недостаточной освещенностью в доступных источниках литературы вопросов по хирургической тактике при агрессивных гемангиомах позвоночника у детей.

Клинические наблюдения. Представлены результаты лечения двух пациентов в возрасте 15 и 17 лет с агрессивными гемангиомами переходного шейно-грудного отдела позвоночника, прооперированных с использованием открытой пункционной вертебропластики. Осложнений в послеоперационном периоде не отмечалось, получены хорошие предварительные результаты.

Обсуждение. Описаны и проанализированы различные подходы к лечению детей с агрессивными гемангиомами переходного шейно-грудного отдела позвоночника, в том числе с применением открытой пункционной вертебропластики.

Заключение. Представленные клинические случаи оперативного лечения агрессивных гемангиом переходного шейно-грудного отдела позвоночника могут вызвать интерес у широкой аудитории вследствие недостаточной освещенности хирургического лечения гемангиом позвоночника в детском возрасте.

Ключевые слова: позвоночник; гемангиома у детей; шейно-грудной переходный отдел; вертебропластика.

Introduction

Spinal hemangioma (SH) is a common pathology that occurs in every tenth individual in the population [1]. As per our literature review, 3.7% of SHs are aggressive in nature [2]. The aggressiveness of a SH is determined based on the radiological and clinical signs. The criteria for aggressiveness include the presence of the extravertebral component of the hemangioma, compression fracture or compression deformity of the vertebral body affected by hemangioma, bone expansion with protrusion of the cortical layer (swelling of the vertebra), lesion with hemangioma of > 60% of the vertebral body, damage (thinning and/or destruction) of the cortical layer, uneven trabecular structure of the hemangioma, expansion of the hemangioma from the body to the vertebral arch, the absence of adipose tissue in the hemangioma structure (low signal from hemangioma at T1 and high signal at T2-WI on magnetic resonance image (MRI); high signal on T2-WI in the mode of the adipose suppressing of the signal, local pain syndrome, and neurological manifestations. A certain number of points (1–5) are assigned to each clinical and radiological sign listed. The hemangioma is considered aggressive if the total score for all the signs is ≥ 5e [3].

With the introduction of radiological examination methods in clinical practice, SHs have shifted from the category of random findings to urgent problems of contemporary medicine [4]. The primary diagnostic methods for spinal diseases are computed tomography (CT) and MRI [3]. CT is the most effective method for diagnosing SH because its accuracy and sensitivity reach 100%, while the accuracy of MRI is 94.7%, and its sensitivity is 96.7% [5]. One of the pathogenetic growth factors of hemangioma is mechanical overload [6]. The transitional spinal sections are known to be subjected to the greatest load [7]. Their functional significance and exposure to increased mechanical stress increase the risk of aggressive growth and pathological fracture of the vertebral body in hemangiomas of these localizations [8]. Majority of the hemangiomas are those of the transitional thoracolumbar spine (Th_{11}–L_2) and constitute approximately 56%–62% of all cases, lesions of the transitional cervicothoracic spine (C_7–Th_1) constitute 2%–4%, while lesions of the transitional lumbosacral spine (L_5–S_1) constitute 8%–12% [9].

SHs are extremely rare in pediatric patients in the first decade of life [10]. It is noteworthy that currently, the number of pediatric SH patients is increasing [11]. Many authors associate this with genetic predisposition and a carcinogenic environmental situation [12]. In pediatric patients, SHs mostly affect the thoracic region (76%) followed by the lumbar region (21%); lesions of the cervical and sacrococcygeal spines are extremely rare (1%–1.5%) [13]. Thus far, the prevalence of SH among those aged < 18 is approximately 8%, considering all the identified cases [10]. In pediatric patients with aggressive SH, clinical signs, such as pain in the area of the affected vertebra or along the entire spine, increased pain after physical exertion and at the end of the day, and frequent numbness of the limbs, is noted [13].

According to most authors, puncture vertebroplasty (PV) is highly efficient in the

treatment of spinal pathology, including aggressive forms of hemangiomas [3]. The main purpose of vertebroplasty is to restore the support ability of the damaged vertebra and to achieve an analgesic and anti-tumor effect [6]. In case of PV in the cervical and transitional cervical-thoracic spine, the percutaneous anterolateral approach is used; however, it has its drawbacks [3]. In particular, the visualization with the electron-optical converter of the main radiography reference points at the C_7–Th_1 level can be significantly limited due to the effect of the radiography shadow created by the shoulder girdle [14]. This hinders the control over the puncture of the vertebra with a needle, thereby increasing the risk of complications. In conjunction with the previously mentioned, the PV at this level is recommended to be performed from the open anterolateral approach [15].

Limited Russian and foreign studies have investigated the surgical approaches in aggressive SHs among pediatric patients. Thus, we present the clinical cases of two patients aged 15 and 17 years with aggressive hemangiomas of the C_7 and Th_1 vertebrae, respectively.

Clinical cases

The method of open puncture vertebroplasty. The open anterolateral approach to the vertebral bodies C_7 and Th_1 was performed under general anesthesia with the patient in the supine position with a small roller under the shoulder girdle and the head in the extension state, slightly turned to the side opposite to the surgical incision at an angle of 15°. The right-hand approach was used. A skin incision, approximately 5 cm long, was made. The subcutaneous tissue, subcutaneous muscle, fascia of the neck were dissected in layers. The surgical wound was deepened in the space between the larynx, pharynx, esophagus on one side and the carotid artery on the other. The midline organs of the neck (larynx, pharynx, esophagus, and thyroid gland) were displaced medially, and the main neurovascular bundle of the neck was displaced laterally. In the depth of the wound, the body of the affected vertebra was exposed, into which a puncture needle was inserted and placed on the front surface of the vertebra closer to the midline. A puncture needle with a length of 10 cm and a diameter of 13 G with a conical circular distal end was used. The volume of the injected cement of high viscosity was 3 mL and 4 mL, respectively.

Clinical case 1. Patient A., 15 years old, presented to the advisory and diagnostic department of the Research Institute of Traumatology, Orthopedics and Neurosurgery, Saratov State Medical University, in March 2018, complaining of pain in the cervical spine radiating to the lateral surface of the shoulder girdle. The pains were persistent and intense, aggravated by head movement. The patient

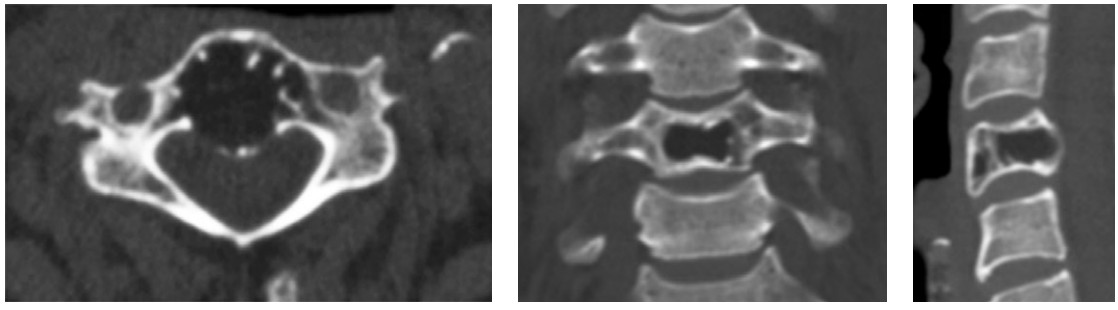

Fig. 1. CT scans of vertebra C_7 of patient A. before the surgery

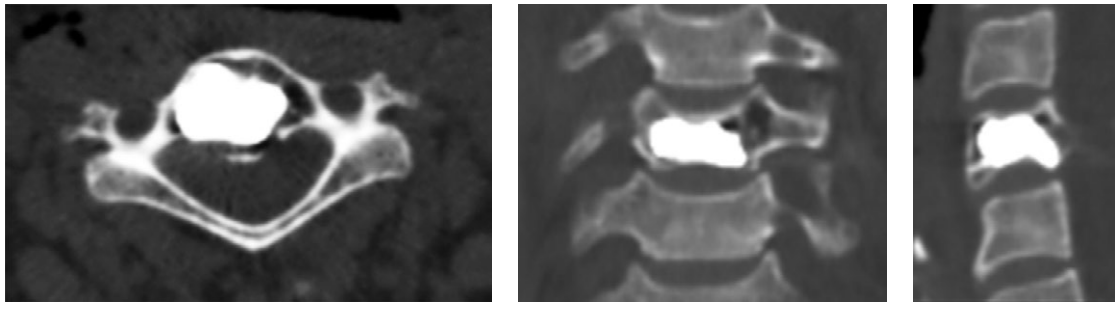

Fig. 2. CT scans of vertebra C_7 of patient A. after the surgery

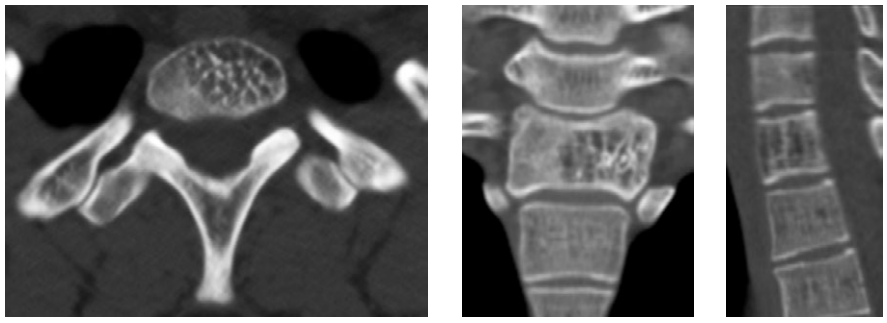

Fig. 3. CT scans of the vertebra Th_1 of patient V. before the surgery

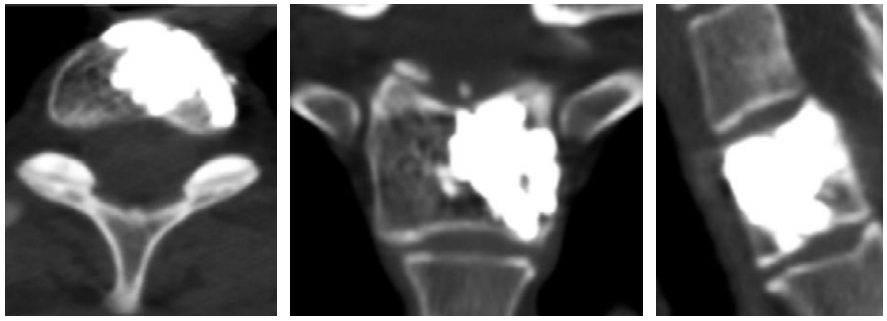

Fig. 4. CT scans of the vertebra Th_1 of patient V. after the surgery

estimated the pain at 8 points based on the visual analogue scale (VAS). The medical history showed that the patient had been experiencing pain over the previous 6 months. A sharp increase in the pain was noted on palpation of the spinous process of vertebra C_7. Using CT examination of the cervical spine, we diagnosed total aggressive hemangioma of the vertebral body C_7 (Fig. 1).

The patient was hospitalized in the in-patient facility where he underwent open PV of the vertebral body C_7. After the surgery, complete regression of pain syndrome was registered. No unplanned migration of polymethyl methacrylate was detected in the postoperative CT images (Fig. 2).

Clinical case 2. Patient V., 17 years old, presented to the institute in May 2018, due to complaints of pain in the cervical and thoracic spine with irradiation to the right upper limb. The pains were dragging and periodic, aggravated by physical exertion. The patient estimated her pain at 6 points as per the VAS. On palpation of the spinous processes of the C_7–Th_1 vertebrae, an increase in pain syndrome was noted. CT examination showed an aggressive hemangioma of the Th_1 vertebral body (Fig. 3). In addition to the clinical symptoms, the aggressiveness of hemangioma was characterized by the CT signs, such as bone expansion with protrusion of the cortical layer; a lesion larger than $^2/_3$ of the vertebral body; damage (thinning) of the cortical layer; and uneven trabecular structure of the hemangioma.

Open PV of the body of the vertebra Th_1 was performed for the patient. After the surgery, the pain syndrome regressed. On postoperative CT images, the cavity was densely filled with polymethyl methacrylate (Fig. 4).

The efficiency of PV was evaluated as per the clinical data of complete regression of pain syndrome (up to 0 points as per the VAS in both patients). After the surgery, both patients underwent a check-up CT examination. On CT images, the completeness of filling the cavity with hemangiomas with bone cement was visualized, from 86% to 94% of filling. The duration of hospital stay was 2–4 days. Subsequent CT control was performed after 6 months, and the absence of relapsing growth of hemangiomas was confirmed.

Discussion

The choice of therapeutic approach for pediatric patients with aggressive SH in the presence of persistent pain syndrome is debatable. It is believed that with the SH identified in a child, it must be observed dynamically before the onset of puberty, and if the hormonal balance changes, the neoplasm

may disappear [16]. However, the literature suggests an exaggerated possibility of conservative treatment of aggressive SHs in pediatric patients, leading to unsatisfactory long-term results, particularly with the development of pathological compression fractures of the vertebral bodies and neurologic impairment due to compression of the vertebral canal contents with a bone or soft-tissue component of the tumor, subsequently requiring complex reconstructive stabilizing surgeries [17, 18]. In this regard, in the presence of severe clinical symptoms, surgical treatment is proposed [10]. The authors believe that the hormonal imbalance contributes to the rapid growth of SH, leading to complications in a growing organism [12]. However, the limitation of the active surgical approach is that patients sometimes experience adverse effects due to inadequate surgical intervention [17].

Currently, PV with bone cement based on polymethyl methacrylate is a highly effective method for treating aggressive SHs [3]. Analyzing the cases of PV used in pediatric patients, it can be said that it provides [17] a persistent analgesic effect, enables the restoration of the support ability of the affected vertebra, and helps prevent tumor recurrence. However, the effect of polymethyl methacrylate on vertebral growth after vertebroplasty has not been fully studied. In foreign literature, two cases are described with a two-year follow-up period of the operated pediatric patients [10], indicating the absence of growth abnormalities.

In the course of percutaneous PV with the antero-lateral approach in the cervico-thoracic spine, the risk of damage by the sharp puncture needle to important neck anatomical structures increases, leading to iatrogenic complications, such as esophageal perforation with the development of mediastinitis with time, damage to large vessels with considerable blood loss, and damage to the respiratory tract [19]. When PV in the cervico-thoracic transitional spine, it should be considered that visualization with an electron-optical transducer of the spine and major reference points can be significantly limited due to the effect of X-ray shadow which is the layering of shoulder joints, shoulder blades, ribs, and apices of the lungs [3]. In such cases, intraoperative CT or special functions of the angiograph can be used. These options enable the performance of CT emulation and track in real time the direction and depth of insertion of the puncture needle using multiplanar plane and volumetric reconstructions [8, 14]. PV can be performed with an open approach that increases the manipulation efficiency and safety [15]. This is definitely more related to vertebroplasty in the transitional cervico-thoracic spine. The open antero-lateral approach provides a broad overview of the anterior and antero-lateral surface of the vertebral bodies, thereby reducing the risk of complications that may occur with the percutaneous approach. Cement with high viscosity and with sufficient working time is more appropriate for vertebroplasty at this level to reduce the risk of extravertebral composite exit.

Conclusion

With aggressive SHs in pediatric patients, timely diagnostics and adequate treatment, including surgery, are crucial. The clinical observations presented may be of interest to a wide audience because of insufficient current literature on SH during childhood and rare localization as well as the fact that until now, this disease in pediatric patients has commonly been treated conservatively.

Additional information

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Ethical review. Legal representatives of the patients agreed to the processing and publication of the personal data. A positive decision to conduct the study was obtained from the ethics committee of the Razumovsky Research Institute of Traumatology, Orthopedics and Neurosurgery, Saratov State Medical University (Protocol No. 2 dated 25.02.2019). The drugs applied passed clinical trials and are registered in the State Register of the Russian Federation. The bone cements used are approved for import, sale and use in the Russian Federation by the order of Federal Service on Surveillance in Health Care and Social Development of the Russian Federation (dated 21.12.2009 and 06.07.2015), namely VertaPlex Stryker Radiopaque Bone Cement (marketing authorization No. FSZ 2009/05816), SynicemVTP Synimed (marketing authorization No. FSZ 2012/11776).

Contribution of the authors

V.V. Zaretskov was involved in the concept and design of the study, phase and final editing of the text of the article.

V.B. Arseniyevich performed the data analysis, interpretation of results, and surgical treatment of the patients.

S.V. Likhachev prepared the text of the article, performed supervision, and surgical treatment of the patient.

S.V. Stepuhovich performed supervision, surgical treatment of the patient, and data analysis.

S.A. Mizyurov analyzed literary sources, collected material, and prepared the text of the article.

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