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CLINICAL CASE REPORT

A case of a fifteen-year-old patient suffering from rare adenocystic lung carcinoma bronchoplastic surgery

D. A. Kharagezov, E. A. Mirzoyan✉, Yu. Yu. Kozel, G. A. Mkrtchyan, A. A. Antonyan, T. G. Ayrapetova, I. A. Leyman, A. G. Milakin, O. N. Stateshny

National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
✉ ellada.mirzoyan@yandex.ru

ABSTRACT

Adenoid cystic carcinoma of the lung is a relatively rare malignant tumor, accounting for 0.04–0.2 % of all primary malignant tumors of the respiratory system. This carcinoma can occur at any age, but it is more common in the 40–60 age group and usually in women. The main treatment method for adenoid cystic carcinoma is surgical. Since tumors of this histological form are often centrally located, options for bronchoplastic operations are considered. In childhood, adenoid cystic carcinoma is extremely rare, and performing bronchoplastic lobectomies in children is associated with several difficulties, such as the smaller diameter of the bronchi compared to adult patients, complicating surgical intervention and subsequent rehabilitation.
This clinical case demonstrates the experience of performing a bronchoplastic operation on a 15-year-old patient at the Department of Thoracic Oncology of the National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation. The patient was hospitalized complaining of prolonged cough, shortness of breath, and chest pain. Adenoid cystic carcinoma of the central type was identified during diagnostics, which included bronchoscopy, computed tomography, and biopsy.
The surgical intervention involved performing a bronchoplastic lobectomy, during which the affected lobe of the lung was removed with resection and reconstruction of the bronchus. The operation was performed taking into account the anatomical features of the child's body, which required high precision and surgical skills. The postoperative period proceeded without significant complications, and the patient was under the supervision of a multidisciplinary team of specialists.
This clinical case provides a detailed description of the results of preoperative diagnostic measures, the stages of the operation, and the postoperative follow-up results. Special attention was paid to the difficulties associated with the small diameter of the bronchi in children, which required the use of specialized instruments and techniques. The importance of using modern diagnostic and treatment methods, as well as close interdisciplinary interaction, is emphasized for a successful treatment outcome.
The experience of performing such operations in childhood is extremely important for improving the quality and safety of surgical treatment of adenoid cystic carcinoma and other rare tumors in children. Further observations will be described in stages.

Keywords: lung cancer, adenocystic carcinoma, bronchoplastic lobectomy in children

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For correspondence: Ellada A. Mirzoyan – Cand. Sci. (Med.), MD, oncologist, researcher at the Department of Thoracic Oncology, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
Address: 63 14 line str., Rostov-on-Don 344037, Russian Federation
E-mail: ellada.mirzoyan@yandex.ru
ORCID: https://orcid.org/0000-0002-0328-9714
SPIN: 2506-8605, AuthorID: 1002948
ResearcherID: AAZ-2780-2021
Scopus Author ID: 57221118516

Compliance with ethical standards: this study followed the ethical principles outlined in the World Medical Association Declaration of Helsinki (1964, revised in 2013). The research was approved by the Ethics Committee of the National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation (protocol No. 16 dated 12.10.2021). Informed consent was obtained from all study participants

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КЛИНИЧЕСКОЕ НАБЛЮДЕНИЕ

Случай выполнения бронхопластической операции пациентке пятнадцати лет с редкой аденокистозной карциномой легкого

Д. А. Харагезов, Э. А. Мирзоян✉, Ю. Ю. Козель, Г. А. Мкртчян, А. А. Антонян, Т. Г. Айрапетова, И. А. Лейман, А. Г. Милакин, О. Н. Статешный

ФГБУ «Национальный медицинский исследовательский центр онкологии» Министерства здравоохранения Российской Федерации, г. Ростов-на-Дону, Российская Федерация
✉ ellada.mirzoyan@yandex.ru

РЕЗЮМЕ

Аденокистозная карцинома легкого является относительно редкой злокачественной опухолью, на долю которой приходится 0,04–0,2 % всех первичных злокачественных опухолей органов дыхательной системы. Эта карцинома может возникнуть в любом возрасте, однако чаще встречается в возрастной группе 40–60 лет и, как правило, у женщин. Основным методом лечения аденокистозной карциномы является хирургический. Поскольку опухоль данной гистологической формы часто располагается центрально, рассматриваются варианты выполнения бронхопластических операций. В детском возрасте аденокистозная карцинома встречается очень редко, а выполнение бронхопластических лобэктомий у детей сопровождается рядом трудностей, таких как меньший диаметр бронхов по сравнению со взрослыми пациентами, что усложняет оперативное вмешательство и последующую реабилитацию. Настоящий клинический случай демонстрирует опыт выполнения бронхопластической операции 15-летней пациентке на базе отделения торакальной онкологии ФГБУ «Национальный медицинский исследовательский центр онкологии» Министерства здравоохранения Российской Федерации, г. Ростова-на-Дону. Пациентка была госпитализирована с жалобами на длительный кашель, одышку и боли в грудной клетке. В ходе диагностики, включающей бронхоскопию, компьютерную томографию и биопсию, была выявлена аденокистозная карцинома центрального типа.

Хирургическое вмешательство включало проведение бронхопластической лобэктомии, при которой была удалена пораженная доля легкого с резекцией и восстановлением бронха. Операция была выполнена с учетом анатомических особенностей детского организма, что требовало высокой точности и навыков хирурга. Послеоперационный период протекал без значительных осложнений, пациентка находилась под наблюдением мультидисциплинарной команды специалистов.

В данном клиническом случае были подробно описаны результаты предоперационных диагностических мероприятий, этапы проведения операции, а также результаты послеоперационного наблюдения. Отдельное внимание было уделено сложностям, связанным с малым диаметром бронхов у детей, что требовало использования специализированных инструментов и техники. Подчеркивается важность использования современных методов диагностики и лечения, а также тесного междисциплинарного взаимодействия для успешного исхода лечения.

Опыт проведения подобных операций в детском возрасте крайне важен для повышения качества и безопасности хирургического лечения аденокистозной карциномы и других редких опухолей у детей.

Ключевые слова: рак легкого, аденокистозная карцинома, бронхопластическая лобэктомия у детей

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Для корреспонденции: Мирзоян Эллада Арменовна – к.м.н., врач-онколог, научный сотрудник отделения торакальной онкологии, ФГБУ «Национальный медицинский исследовательский центр онкологии» Министерства здравоохранения Российской Федерации, г. Ростов-на-Дону, Российская Федерация
Адрес: 344037, Российская Федерация, г. Ростов-на-Дону, ул. 14-я линия, д. 63
E-mail: ellada.mirzoyan@yandex.ru
ORCID: https://orcid.org/0000-0002-0328-9714
SPIN: 2506-8605, AuthorID: 1002948
ResearcherID: AAZ-2780-2021
Scopus Author ID: 57221118516

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INTRODUCTION

Lung cancers occupy the first place in the structure of morbidity and mortality from cancer among the male population [1–3].

Adenocystic carcinoma (ACC) of the lung is a relatively rare malignant tumor, which accounts for 0.04– 0.2 % of all primary malignant tumors of the respiratory system [4]. ACC can occur at any age, but it is more common in the age group of 40–60 years and is usually found in women [5–6]. The main method of treatment of ACC is surgical, and since the tumor of this histological form is often centrally located, options for performing bronchoplastic operations are being considered [7, 8].

Bronchoplastic lobectomy with systematic mediastinal lymphodissection is a good choice for the treatment of endobronchial tumors in both children and adult patients in order to preserve lung parenchyma [9]. Primary lung tumors in childhood are very rare, and performing bronchoplastic lobectomies in children is accompanied by a number of difficulties, in particular, the diameter of the bronchi is much smaller than in adults [10].

The purpose of the study was to present a clinical case of a 15-year-old patient with rare adenocystic lung carcinoma who underwent bronchoplastic surgery with a good long-term treatment result.

Clinical observation report

A patient born in 2006 applied to the National Medical Research Center for Oncology, Rostov-on-Don in February 2022 with complaints of periodic cough, shortness of breath, feeling of lack of air, hemoptysis. According to him, he has been ill since 2021, with a history of repeatedly suffering from pneumonia.

Since June, after suffering a new coronavirus infection, there have been frequent attacks of shortness of breath. In November, the condition has worsened, followed by hemoptysis, increased shortness of breath, feeling of lack of air. He went to the doctor at the place of residence. SCT performed on 12/02/2021 showed: a volumetric formation of an irregular shape of 1.3 × 0.7 × 1.2 cm of a heterogeneous structure is determined in the lumen of the left main bronchus.

12/28/22 SCT in the lumen of the main bronchus on the left showed a multi-node formation of 1.3 × 1.2 cm narrows its lumen, the remaining bronchi are passable in the visible extent (bronchial lumen of the 3rd-4th order is visualized), their walls are not thickened, without signs of bronchiectasis, on the left in the lower lobe there are areas of pneumosclerosis, on the right along the interlobular pleura a single dense focus requiring dynamic observation (Fig. 1).

SCT scan with angiography from 01/24/2022. The CT picture is more consistent with the endobronchial formation (carcinoid) of the left main bronchus.

FBS dated 12/28/2021: the larynx is mobile, the trachea, carina and bronchi of the right lung are without features. On the left: the main bronchus is 3/4 encircled by a tuberous exophyte about 1.5 × 1.2 cm in size. The exophyte originates from the interlobular spur and is located on a narrowed base. The spur at the exophyte outlet was expanded due to the sub-

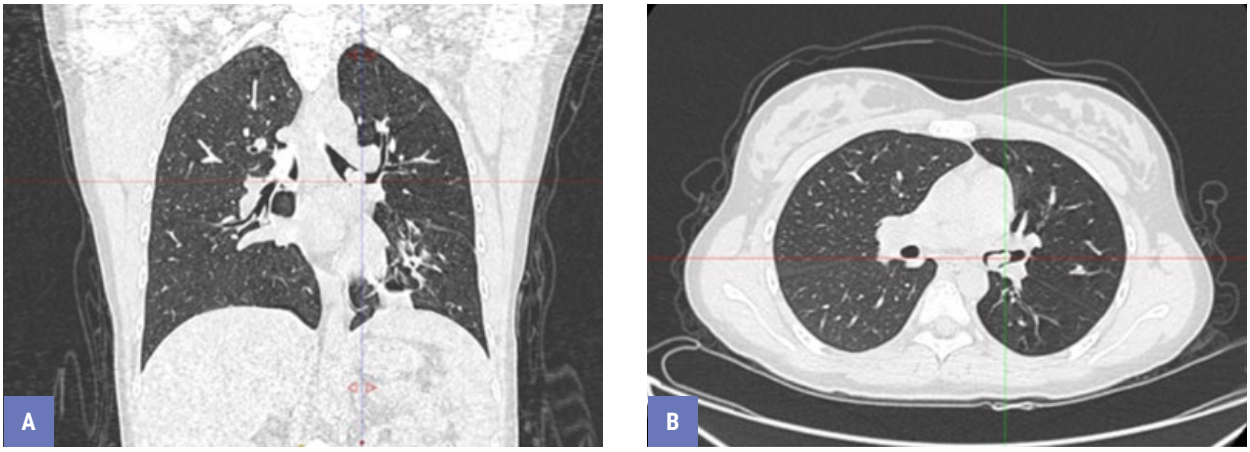

Fig. 1. Chest CT scan of the patient G.: A – frontal section; B – sagittal section

mucosal component of the tumor, and a biopsy was taken. Conclusion: exophytic tumor of the upper lobe spur of the left lung with signs of invasion of the interlobular spur. Subcompensated stenosis of the left main bronchus (carcinoid?) (Fig. 2).

Histological analysis 145380–81 /21 dated 01/13/2022: The morphological picture in the volume of biopsies is more typical for a typical carcinoid/ neuroendocrine tumor. To clarify the immunophenotype of tumor cells, an IHC study is recommended.

IHC dated 01/21/2022: morphological picture and immunophenotype of tumor cells in the volume of biopsies (PanCK+, CK7+, CD117+, p63+ in the myoepithelial layer) are characteristic of adenocystic carcinoma.

Upon admission to the Department of Pediatric Oncology No. 1 02.02.2022, the PCR test for SARS-CoV-2 is negative, PS ECOG (Eastern Cooperative Oncology Group scale, designed to assess the general condition of cancer patients) 1 point. The superficial lymph nodes are not enlarged. The chest is not deformed, both halves of it are evenly involved in the act of breathing. The breathing rate is 15 per 1 minute at rest. Percussive clear pulmonary sound, the same on the right and on the left. Auscultatively vesicular breathing on the right, and weakened on the left. Spirometric parameters are normal. On the ECG: Sinus rhythm with a heart rate of 68 beats /min, an ECG variant of the age norm. The clinical diagnosis was made: (C34.1) Adenocystic carcinoma of the left upper lobe bronchus T1bNxM0, stage IA, cl. gr. 2.

The patient was taken to the operating room on 02/07/2022. The patient was positioned lying on her right flank. An anterolateral thoracotomy was performed in the 5th intercostal space on the left. According to a comprehensive examination, the patient has a central malignant tumor of the distal part of the left main bronchus of 1.3 × 1.0 cm, spreading to the upper lobe bronchus. It was decided to perform an upper bronchoplastic lobectomy on the left with wedge-shaped resection of the main, lower lobar bronchus, mediastinal lymphadenectomy. The pulmonary ligament is excised. A posterior mediastinotomy was performed. Bifurcation lymphodissection was performed using an electrosurgical instrument. Anterior mediastinotomy was performed, the diaphragmatic nerve was visualized, isolated, and placed on a turnstile. With the help of Thunderbeat, the superior pulmonary vein is mobilized and crossed by a suturing device. The anterior mediastinal fiber and a group of 4L-6 lymph nodes were removed in a single block. Hemostasis was performed. The visceral pleura was dissected in the projection of the interstitial fissure. A2, A1–3, A4–5 were isolated, sequentially ligated and crossed. The lower lobe is mobilized from the upper lobe by a linear stitching device. Using the Thunderbeat electrosurgical instrument, the lymph nodes of the lung root were removed with the removal of the l/nodes of the root of the lower lobe with the exposure of the upper lobe, lower lobe bronchus, interlobular spur, distal part of the left main bronchus, where a tumor formation up to 1.5 cm in diameter is contoured. Wedge-shaped resection of the distal part of the main, interlobular spur, and proximal part of the inferior lobe bronchus was performed, with suturing of the defect with separate nodular sutures with atraumatic monofilament thread. The drug has been removed. The line of tantalum sutures on the lung parenchyma is additionally coagulated with bipolar tweezers. Hemostasis was performed. Control of pneumostasis by underwater breakdown: pneumostasis was stable. Drainage of the left pleural cavity at 7 and 9 i/c 2. Layered suturing of a thoracotomy wound.

The result of a planned histological examination: 11136–37/22: The morphological picture (taking into account IHCNo.83/22) is characteristic of bronchial adenocystic carcinoma (salivary gland-type tumor), with an exophytic growth pattern, invasion of the submucosal layer, the presence of foci of carcinoma in the adventitial layer. There were no signs of

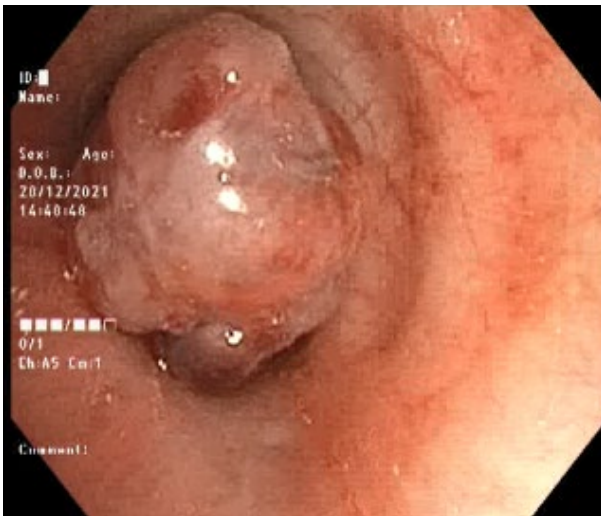

Fig. 2. FBS from 28/12/2021

perineural and lymphovascular invasion. No tumor cell resection lines were found in the adjacent bronchopulmonary 4L-7, 10–14 lymph nodes.

The postoperative period was uneventful. On the control fibrobronchoscopy from 02/14/2022: a wide interbronchial suture was determined. The seam line was consistent. Its mucous membrane was edematous, hyperemic, and there was a fibrin plaque on the anterior and posterior walls. The bronchi of the lower lobe were not deformed, freely passable. The mucous membrane was smooth, pale, shiny (Fig. 3).

The FBS dated 02/21/2022 showed: on the left, an anastomosis of the main and lower lobe bronchi in the form of an annular roller narrowing the lumen by 1/3, with suture ligatures, four red spots and one spot of fibrin. Bronchi of the lower lobe of the usual type. The anastomosis is in the resolution stage (Fig. 4.).

On the 15th day after surgery, the patient was discharged from the hospital in a satisfactory condition with the diagnosis: C34.1 Adenocystic carcinoma of the left upper lobe bronchus pT1bN0M0, stage IA, condition after thoracotomy, combined upper bron-

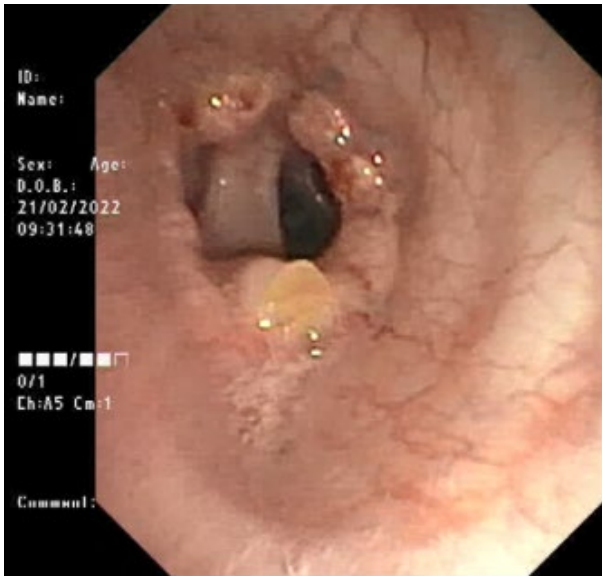

Fig. 3. FBS patient G. from 02/14/2022 on the 7th day after surgery

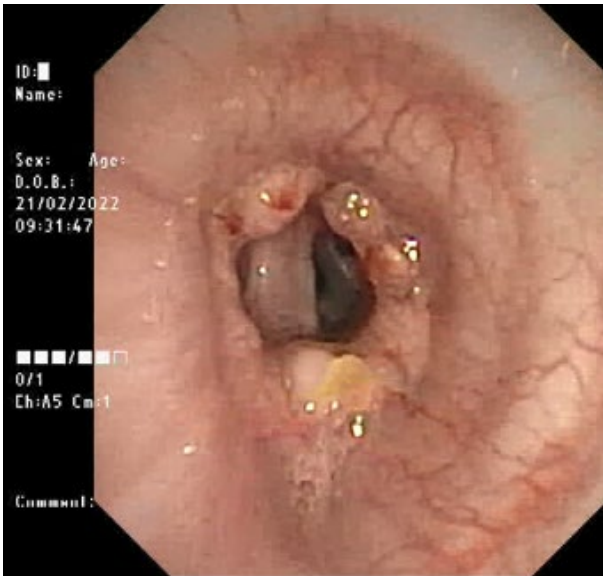

Fig. 4. FBS patient G. dated 02/21/2022 on the 14th day after surgery

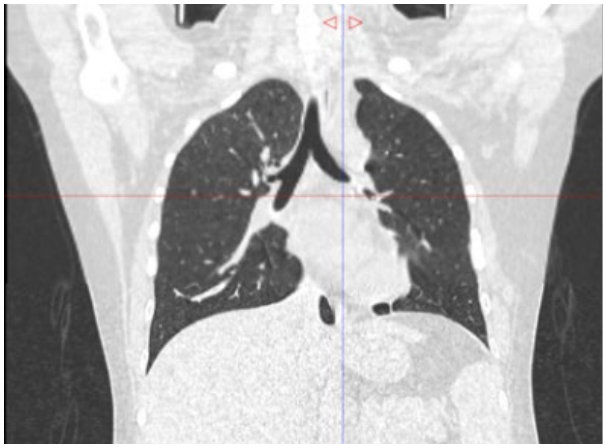

Fig. 5. Chest CT scan of patient G. from 28/11/2023, 1 year 9 months after surgery: frontal section

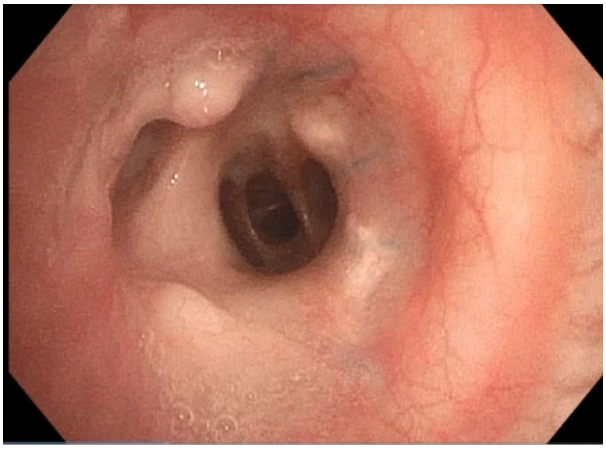

Fig. 6. FBS patient G. from 11.28.2023, 1 year 9 months after surgery

choplastic lobectomy on the left with wedge-shaped resection of the main, lower lobe bronchus, mediastinal lymphadenectomy from 02/07/2022, cl. gr. 3.

At the control visit on 11/28/2023, 1 year and 9 months after the operation, the patient does not complain, feels healthy, and leads an active lifestyle. PS ECOG 0 points, no pathology was detected during physical examination. On CT of the chest organs and FBS without signs of progression of the process (Fig. 5, 6).

DISCUSSION

Improvements in surgical techniques and anesthetic aids have led to the introduction of broncho- and angioplasty operations, which demonstrate better immediate and long-term treatment results compared to the pneumonectomies [11–14].

In the paperwork of E. V. Levchenko et al. A comparative analysis of the long-term results of surgical treatment of 198 patients with stage I–III non-small cell LC was carried out: bronchoplastic operations were performed in 99 patients, pneumonectomies – 99. The median overall and recurrence-free survival was 51.4 and 55.2 months after bronchoplastic lobectomies, and in patients after pneumonectomies 46.2 and 41.0 months, respectively. One-year, 3- and 5-year recurrence-free survival in the group of bronchoplastic resections was 87.9 %, 64.2 % and 52.3 %, respectively, versus 88.1 %, 61.6 % and 37.9 % in the group after pneumonectomies [15].

The results of bronchoplastic surgeries and pneumonectomies are considered in detail in the meta-analysis of Z. Li with co-authors, which presents the results of treatment of 14194 patients: 4145 performed bronchoplastic operations, 10049 – pneumonectomies. Overall survival was higher in the group of patients who underwent bronchoplastic lobectomies (OR: 1.53; 95 % CI: 1.31–1.80; $p < 0.00001$), and in the group of patients after pneumonectomies, there was a higher level of postoperative and 30-day mortality, as well as the frequency of distant metastases (5.86 % and 2.78 %, respectively) [16].

Various variants of bronchoplastic surgeries have also been used in pediatric practice, where the preservation of maximum lung tissue is also an important aspect [17]. Yu et al. presented the largest study evaluating the effectiveness of bronchoplastic interventions in children and adults. The authors found that this technique has a good prognosis in the pediatric population [18]. However, there are few reports in the literature about performed bronchoplastic surgeries for lung cancer in children [19].

CONCLUSION

A clinical case of a 15-year-old patient with a rather rare adenocystic lung carcinoma who underwent bronchoplastic lobectomy has been presented in this article. There is no data for the progression and recurrence of the process, the patient is currently under dynamic observation.

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Information about authors:

Dmitrii A. Kharagezov – Cand. Sci. (Med.), MD, Head of the Department of Thoracic Oncology, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0003-0640-2994, SPIN: 5120-0561, AuthorID: 733789, ResearcherID: AAZ-3638-2021, Scopus Author ID: 56626499300

Ellada A. Mirzoyan ✉ – Cand. Sci. (Med.), MD, oncologist, researcher at the Department of Thoracic Oncology, of National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0002-0328-9714, SPIN: 2506-8605, AuthorID: 1002948, ResearcherID: AAZ-2780-2021, Scopus Author ID: 57221118516

Yulia Yu. Kozel – Dr. Sci. (Med.), MD, Head of the Department of Pediatric Oncology, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0002-6681-3253, SPIN: 6923-7360, AuthorID: 732882, ResearcherID: AAZ-2780-2021, Scopus Author ID: 57221118516

Gulnara A. Mkrtchyan – MD, pediatric surgeon at the Department of Pediatric Oncology No. 2, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0002-5202-7681, SPIN: 1861-5165, AuthorID: 794720

Artur A. Antonyan – PhD student, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0001-6449-7026, ResearcherID: JWQ-0734-2024

Tamara G. Ayrapetova – Cand. Sci. (Med.), MD, Oncologist at the Department of Thoracic Oncology, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0001-5287-5800, SPIN: 8121-4039, AuthorID: 794672

Igor A. Leyman – Cand. Sci. (Med.), MD, Oncologist at the Department of Thoracic Oncology, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0003-2572-1624, SPIN: 2551-0999, AuthorID: 735699

Anton G. Milakin – MD, oncologist at the Department of Thoracic Oncology, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0002-2589-7606, SPIN: 7737-4737, AuthorID: 794734, Scopus Author ID: 57192109933

Oleg N. Stateshny – MD, oncologist at the Department of Thoracic Oncology, National Medical Research Centre for Oncology, Rostov-on-Don, Russian Federation
ORCID: https://orcid.org/0000-0003-4513-7548, SPIN: 9917-1975, AuthorID: 1067071

Contribution of the authors:

Kharagezov D. A., Kozel Yu.Yu. – scientific editing.
Mirzoyan E. A., Antonyan A. A. – writing the draft; data processing.
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