

Clinical characteristics of children with primary tracheobronchial tumors and SVM model in mucoepidermoid carcinoma Prediction

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Abstract

Objective

To determine the clinical characteristics, treatment, and prognosis of primary tracheobronchial tumors (PTT) in children.

Methods

The medical records of children with PTT who were hospitalized at the Children's Hospital of Chongqing Medical University from January 1995 through January 2020 were reviewed retrospectively. The clinical features, imaging, and bronchoscopic manifestations, treatment, and outcomes of these patients were summarized.

Results

Sixteen children were hospitalized with PTT during the study period: five (31.3%) with mucoepidermoid carcinoma (ME), three (18.8%) with inflammatory myofibroblastic tumors (IMT), two (12.5%) with sarcoma, two (12.5%) with papillomatosis, and one (6.3%) each with carcinoid carcinoma, adenoid cystic carcinoma (ACC), hemangioma, and schwannoma. Among them, ME was the most common type of tumor, and we used some statistical and machine learning techniques to predict ME, such as Gaussian naïve Bayes model, SVM(support vector machine) model, and decision tree model. The median age at diagnosis of PPTs was 9.3 years (range, 3–15 years), and the main clinical symptoms were cough (81.3%), breathlessness (50%), wheezing (43.8%), progressive dyspnea (37.5%), hemoptysis (37.5%), and fever (25%). The most frequent complication on chest imaging was atelectasis (56.3%), followed by bronchiectasis (25%) and emphysema (12.5%). Of the 16 patients, seven underwent surgical treatment, eight underwent bronchoscopic tumor resection, and one died. Of the 11 children followed-up, three experienced recurrence, including two who underwent subsequent tracheotomies, both of whom have varying degrees of post-activity shortness of breath. The other eight patients did not experience recurrence. No deaths were observed during follow-up.

Conclusion

PTTs are very rare in children, with ME having the highest incidence. And the SVM model showed high accuracy with respect to identifying ME. Chest CT and bronchoscopy can diagnose PTT effectively. Surgery is the treatment of choice, although bronchoscopic intervention can achieve good results in patients with unresectable tumors. The prognosis of the 11 followed-up children was good.

1. Introduction

Primary tracheobronchial tumors (PTT) are extremely rare in children and are misdiagnosed easily. Few reports describe patients with these tumors, making their clinical presentation, diagnosis, treatment, and outcome unclear. This study retrospectively reviewed the clinical data of children who were hospitalized with pathologically confirmed PTT in the Children's Hospital of Chongqing Medical University from January 1995 to January 2020, and then followed-up by telephone in July 2020.

2. Materials And Methods

The medical records of all children diagnosed with PTT from January 1995 to January 2020 at the respiratory department, thoracic surgery ward, and ear, nose, and throat department of the Children's Hospital of Chongqing Medical University were reviewed retrospectively. Data collected included demographic characteristics, clinical presentation, results of chest X-rays and computed tomography (CT) scans, pathological diagnosis, lesion location, treatment, follow-up, and clinical outcome. Patients were included if they were under 18 years old and had an intracavitary lesion of the trachea or large bronchus confirmed by chest CT and bronchoscopy, with pathology confirmed by biopsy^{1,2}. Patients with lung cancer, laryngeal cancer, and metastatic tracheobronchial tumors were excluded. At the same time, the clinical manifestations and imaging characteristics collected were used to establish a database to construct an identification model for the most common tracheobronchial tumor type, ME. We use Gaussian naïve Bayes model, SVM(support vector machine) model, and decision tree model to build the model respectively.

3. Results

3.1 General information

During the study period, 16 children (ten boys and six girls; median age, 9.3 years; range, 3–15 years) were diagnosed with PTT. Of these, ten were aged between 9 and 15 years. Five (31.3%) had mucoepidermoid carcinomas (ME), three (18.8%) had inflammatory myofibroblastic tumors (IMT), two (12.5%) had sarcomas, two (12.5%) had papillomatosis, and one (6.3%) each had carcinoid carcinoma, adenoid cystic carcinoma (ACC), hemangioma, and schwannoma (Tables 1 and 2). The personal and family histories of the 16 children were unremarkable.

3.2 Clinical manifestations

The main clinical manifestations in these 16 children were cough (81.3%), breathlessness (50%), wheezing (43.7%), progressive dyspnea (37.5%), hemoptysis (37.5%), and fever (25%) (Table 3). Other manifestations included chest pain, weight loss, hoarseness, and dysphagia. The average delay between symptom

appearance and initial diagnosis was 10 months (range, 7 days to 5 years). Tumors located in the trachea usually presented with dyspnea, hoarseness, dysphagia, or other neck compression symptoms (dyspnea occurred earlier than bronchial tumors).

3.3 Radiological examination

Most of these children underwent chest CT with enhancement and airway reconstruction, with chest CT usually showing bronchial luminal obstruction in the lesion, with soft tissue shadowing, with or without bronchiectasis, atelectasis, or compensatory emphysema (Fig. 1). The most common abnormalities were atelectasis (9/16, 56.3%), followed by bronchiectasis (4/16, 25%) and emphysema (2/16, 12.5%). Maximum tumor diameter ranged from 5 to 40 mm. Seven tumors were located in the left main bronchus, three in the right main bronchus, and six in the trachea. PET/CT scanning of one patient with ME (moderately malignant) and carcinoid carcinoma revealed an SUV in the primary lesion that was significantly higher than that in the surrounding tissue, which is consistent with manifestations of a malignant tumor. F-18 FDG accumulation in the lesion has been reported in high- and low-grade MEs³.

3.4 Diagnosis and treatment

Fifteen children underwent bronchoscopy and biopsy. Seven were treated surgically. Three (19%) underwent sleeve lobectomies: two underwent left upper sleeve lobectomies, and one underwent a right middle sleeve lobectomy. In addition, one (6%) patient underwent a right middle and lower lobectomy, and three (19%) underwent tracheal sleeve lobectomies with end-to-end anastomosis, including one who also underwent partial thyroidectomy. The remaining eight children underwent local tumor resection via bronchoscopy: five (31%) underwent tumor resection alone, and two (13%) underwent tumor resection followed by chemotherapy. Surgery was successful in all 15 patients.

One child died during hospitalization due to sudden onset of extreme dyspnea. Bronchoscopy revealed complete obstruction of the left main bronchus and two-thirds of the right bronchus by the tumor. An autopsy confirmed the diagnosis of rhabdomyosarcoma.

3.5 Follow-up and outcome

Of the 15 children, two were lost to follow-up after 5 and 60 months, and four were never followed-up. The remaining nine children were reached successfully by telephone in July 2020. The average follow-up duration was 46 months (range, 5–182 months). One child (patient no. 3) experienced tumor recurrence within 2 months after tracheal sleeve resection; this patient underwent local tumor resection twice, with no recurrence for 18 months thereafter. However, this patient still experiences wheezing after strenuous activity. Two children with intratracheal papillomatosis (patient nos. 13 and 14) experienced tumor recurrence 6–8 times and underwent repeated local tumor resection, with no new recurrence at the last follow-up. One still experiences shortness of breath after activity, whereas the other can participate in normal activities; both have hoarseness. Two patients (patient nos. 2 and 4) who underwent local tumor resection plus chemotherapy are still receiving chemotherapy, with patient number 2 being treated with ifosfamide, etoposide, vindesine, methotrexate, and prednisone; their activities are not affected. The remaining children show no evidence of disease recurrence. No deaths were observed during the follow-up period.

3.6 Recognition model of ME

ME carcinoma was the most common tracheobronchial tumor in children. A total of 16 characteristic parameters were selected to construct a database: ten clinical manifestations (cough, fever, progressive dyspnea, weight loss, wheezing, dysphagia, breathlessness, hemoptysis, chest pain, and hoarseness); three tumor locations (left bronchus, right bronchus, and trachea); and three imaging features (atelectasis, bronchiectasis, and emphysema). Then, a Gaussian naïve Bayes model, an SVM model, and a decision tree model were used to predict incidence of mucoepidermoid carcinoma (Table 4). All cases were assigned randomly to a training set (11 cases) and a test set (five cases). The training set was used to select characteristics and to construct recognition models, and the test set was used to evaluate the prediction performance of the models. The area under the ROC curve (AUC), accuracy, and F1-score were used to evaluate the models. We found that the SVM model showed the best performance, followed by the decision trees and the Gaussian plain Bayes model. The SVM model uses python sklearn, and the kernel is linear; the relaxation coefficient is 6.13, and the other parameters are default settings. The accuracy, AUC, and F1-score of the test set were all 1 (Fig. 2), which indicates that the model shows good predictive efficiency in the test set.

Table 1
Clinical profiles and outcomes of 16 patients with primary tracheobronchial tumors

Patient no.	Age/sex	Symptoms	Symptom duration (months)*	Pathology	Location	Size (mm)	Imaging characteristics	Treatment methods	Follow-up (months)
1	4/B	Cough, fever, dyspnea, weight loss	0.4	IMT	LM	16.3*4	Atelectasis	Upper sleeve lobectomy	48
2	5/B	Cough, fever, wheezing	1	IMT	LM	26.8*10.2	Atelectasis, bronchiectasis	Tumor resection + chemotherapy	6
3	11/B	Dyspnea, dysphagia	4	IMT (ALK positive)	T	18.4*16.5*14.5	No	Trachea sleeve resection	22
4	12/B	Cough, wheezing, breathlessness	2	ME (moderate malignancy)	LM	25.4*13.2*12.8	Atelectasis, emphysema	Tumor resection + chemotherapy	5
5	10/G	Cough, wheezing, breathlessness, hemoptysis	10	ME (low-grade malignancy)	RM	23.8*19.2*40.6	Emphysema	Tumor resection	16
6	9/B	Cough, fever, breathlessness, dyspnea	1	ME (low-grade malignancy)	LM	13*16*12	Atelectasis, bronchiectasis	Tumor resection	44
7	14/B	Cough, breathlessness, hemoptysis	60	ME (low-grade malignancy)	LUL	35*25	Atelectasis, emphysema, bronchiectasis	Upper sleeve lobectomy	lost
8	9/B	Cough, hemoptysis	5	ME	RIL	37.5*16.3*17.4	Atelectasis	Middle sleeve lobectomy	lost
9	13/G	Cough, wheezing, breathlessness, hemoptysis	24	Carcinoid	RIL	7.3*22.6	Atelectasis	Middle and lower lobectomy	19
10	13/B	Cough, fever, wheezing, breathlessness, dyspnea	1	ACC	T	13.2*11.5*24.7	Bronchiectasis	Trachea sleeve resection	lost
11	15/B	Cough, breathlessness, hemoptysis, chest pain	1	Schwannoma	T	12.7*8.2	No	Trachea sleeve resection + right partial thyroidectomy	25
12	8/G	Cough, hemoptysis	0.2	Hemangioma	T	5*3.6	No	Tumor resection	76
13	3/G	Hoarseness, dyspnea	24	Papillomatosis	T	10*10*10	No	Tumor resection	60
14	4/B	Wheezing, breathlessness	24	Papillomatosis	T	10*5	Atelectasis	Tumor resection	182
15	6/G	Cough, wheezing, breathlessness, dyspnea	1	Rhabdomyosarcoma	LM	20*18	Atelectasis	Dead	
16	13/G	Cough, hemoptysis, chest pain	6	Synovial sarcoma	RM	7.7*16.6	No	Tumor resection	lost

Abbreviations: G, girl; B, boy; ME, mucoepidermoid tumor; ACC, adenoid cystic carcinoma; IMT, inflammatory myofibroblastic tumor; ALK, anaplastic lymphom stem bronchus; LUL, left upper lobe; RM, right main stem bronchus; RIL, right intermiddle lobe; T, trachea; NED, no evidence of disease.

*Time from symptom appearance to initial diagnosis.

Table 2
Characteristics of the various types of tracheobronchial tumor

Pathological types[2]	Malignant tumors				Benign tumors				Total
	ME	Sarcoma	ACC	Carcinoid	IMT	Papillomatosis	Schwannoma	Hemangioma	
Number of patients	5	2	1	1	3	2	1	1	16
Left bronchus /trachea/right bronchus	3/0/2	1/0/1	0/1/0	0/0/1	2/1/0	0/2/0	0/1/0	0/1/0	7/6/3
Sex (B/G)	4/1	0/2	1/0	0/1	3/0	1/1	1/0	0/1	10/6
Average age (year)	10.8	9.5	13	13	6.7	3.5	15	8	9.3
Atelectasis	4	1	0	1	2	1	0	0	9
Death	0	1	0	0	0	0	0	0	1
Recurrence	0	0	0	0	1	2	0	0	3
Follow-up	3	0	0	1	3	2	1	1	11

Table 3
Main symptoms presenting in the 16 pediatric patients with tracheobronchial tumors

Symptoms	n = 16 (%)
Cough	13 (81.3%)
Breathlessness	8 (50%)
Wheezing	7 (43.8%)
Progressive dyspnea	6 (37.5%)
Hemoptysis	6 (37.5%)
Fever	4 (25%)
Chest pain	2 (12.5%)
Dysphagia	1 (6.3%)
Weight loss	1 (6.3%)
Hoarseness	1 (6.3%)
Asymptomatic	0

Table 4
Evaluation index of the three models

Measure	Gauss naïve Bayes	Support vector machine(linear)	Decision trees
Accuracy	0.60	1.00	0.80
F1-score	0.60	1.00	0.78
AUC	0.58	1.00	0.75
AUC, area under the ROC curve. F1-score is a weighted average of the precision and recall.			

4. Discussion

PTTs are very rare in children. Here, we found that the incidence of tumors in pre-school or school-age children was higher in boys than in girls, and that the most common tumor was ME (31.3%). Different from the most common types of tumor reported abroad⁵, ME carcinoma is the most common type of PTT reported in domestic studies, even accounting for up to 74% of cases⁴. Thus, ME is a malignant tumor with a high incidence rate, meaning that identification is important. The SVM model showed high accuracy with respect to identifying ME. Therefore, in the absence of invasive pathological biopsy, the model can be used to analyze clinical data from patients with tracheobronchial tumors and can help to identify the type of the tumor; this will be especially useful for young doctors or doctors in primary hospitals as it provides an important reference for follow-up treatment and clinical research. However, in view of the small number of cases in our group, the model shows some instability and requires further verification using other independent data.

No specific respiratory symptoms are present during the early stage of the PPTs⁶. In our cohort, the most common symptom was a cough, followed by breathlessness, wheezing, progressive dyspnea, and fever. The incidence of specific symptoms such as hemoptysis and chest pain is similar in children and adults⁴, whereas non-specific symptoms such as fever, cough, and wheezing are more common in children. X-rays of the 16 children identified lung infection in seven. The remaining nine were negative, resulting in misdiagnoses that included pneumonia, asthma, or foreign bodies, with one child being misdiagnosed

with tuberculosis for 5 years. Finally, these children were diagnosed with PTT by chest CT and bronchoscopy. The rate of misdiagnosis of PTTs has decreased gradually over time, as has the time from appearance of symptoms to a correct diagnosis; this is due to greater use of chest CT and bronchoscopy. Chest CT can assess the size and location of the lesion, the extent of infiltration into surrounding tissues, the enlargement of mediastinal lymph nodes, and the involvement of the lung parenchyma^{6,7}. The high calcification rate (50%) seen in ME⁸ can be confirmed by CT. Bronchoscopy can visualize the site, shape, and size of the lesion, enabling biopsies to determine the nature of the lesion; however, the positivity rate of bronchial lavage is low⁹.

Surgery is the treatment of choice for PTT in children, with sleeve lobectomy often chosen to preserve lung function⁴⁻⁶. Complete surgical resection is effective in patients with malignant tumors¹⁰; however, the benefits of adjuvant radiotherapy and chemotherapy are unclear, so they are used rarely. Chemotherapy and radiotherapy are still recommended for children with tumor recurrence⁷, and radiotherapy is a viable option for patients with highly malignant tumors with positive surgical margins¹¹. Studies of targeted therapy for PTT show that recurrent respiratory papillomatosis responds well to bevacizumab¹². In addition, adjuvant treatment with an ALK inhibitor such as cyclozolidinib improves the prognosis of children with IMT and partial ALK gene rearrangement¹³⁻¹⁵.

In addition to surgical resection, bronchoscopic intervention is effective in treating patients with PTT⁹. However, the benefits of endoscopic local resection of tumors are unclear because endoscopic resection has several limitations, including a high risk of transmural injury, an inability to coagulate larger diameter vessels, and a risk of airway tree dislocation¹⁶. CT angiography before fibrilloscopic intervention showed that two patients in the present cohort had large vessels deep within the tumor. After consultation with a cardiologist to reduce the risk of bleeding, both patients underwent minimally invasive vessel closure. In addition to the higher intraoperative risk, the recurrence rate was higher after local resection than after surgical treatment. Indeed, 43% of patients who underwent local resection experienced tumor recurrence during follow-up period⁴. Because some of these patients later required tracheal cricotomy, single local tumor resection through bronchoscopy is not recommended. At present, bronchoscopy is used mainly to assess the degree of airway narrowing, to take biopsies to confirm the diagnosis, and to perform temporary palliative treatment such as deburring, lavage, or APC plus cryotherapy⁷ in patients with unresectable lesions, both to reduce airway obstruction and improve patient quality of life.

Factors affecting the prognosis of children with PTT include histologic classification, pathologic grade, tumor size, lymph node status, distant metastasis, and the extent and ability of surgery¹⁷⁻¹⁹, with pathologic type being the most important factor¹⁹. For example, ME (the most common type of PTT) can be classified as low, intermediate, and high-grade tumors^{20,21}. The Adult Tracheobronchial ME Study reported that the 5 year survival rate of patients with low-grade ME was 57%, whereas the survival rate of patients with high-grade ME was significantly lower^{21,22}, with a 1 year survival rate of only 20%¹⁸. Moreover, survival rates differed significantly in patients who did and did not undergo surgery. By contrast, the 5 year survival rate of children with ME is estimated to be 95%; no deaths were reported in children with low- to intermediate-grade ME, with those having high-grade ME or distant metastases having a lower survival rate. The presence of a t(11;19) (q21;p13) translocation is also associated with a good prognosis in children¹⁸. Chemotherapy is associated with poorer late health and lower long-term survival rates²³. Except for one child in our cohort who was diagnosed with sarcoma and died during hospitalization, the prognosis of the 11 children who were followed-up was good. Three patients experienced tumor recurrence, with two undergoing subsequent tracheotomy. Their prognosis, however, remained good after a maximum follow-up of 182 months. The other eight patients, including three treated surgically and five treated by bronchial intervention, did not experience tumor recurrence after a maximum follow-up of 76 months. Guidelines suggest that bronchoscopy should be performed every 6 months for the first year and then annually thereafter²⁴.

In summary, pediatric PTT is relatively rare, with an insidious onset and an uncharacteristic clinical presentation^{6,7}. Increased tumor volume may cause tracheal and bronchial lumen narrowing and, when combined with respiratory tract infection, can result in fever, cough, breathlessness, and wheezing. These conditions can be confused easily with bronchial foreign body, asthma, pneumonia, and other bronchial diseases, leading to an incorrect or missed diagnosis^{8,25,26}. Lack of realization of the possibility of a tumor and the consequent need for chest CT and bronchoscopy can delay treatment, resulting in extensive adhesions and involvement of surrounding tissues, or even spread to distant airways, thereby worsening the prognosis^{13,15}. PTT should be considered in patients with chronic cough, wheezing, or progressive dyspnea who respond poorly to conventional treatment. A thorough examination of the respiratory tract, especially chest CT and bronchoscopy, may reduce the likelihood of misdiagnosis, and improve treatment and prognosis.

Declarations

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Data Availability Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request. Extra data are available by emailing t_dy@163.com

Author contributions

Chen Zhang and Wenlong Fu planned the study, monitored its progress, and drafted the manuscript based on comments provided by all authors. In addition, Chen Zhang was responsible for data collection and management, and Wenlong Fu was responsible for the follow-up of children. Chen Zhang and Wenlong Fu contributed equally to this study. Xiaofeng Ma was responsible for the design of identification models. Daiyin Tian designed the study and revised the manuscript. Jihong Dai, Yonggang Li, Xingye Tang, Gang Geng, Ying Li, Ting Yang, Li Yan, Jingyue Liu, Zheng Liu, and Xiaoping Yuan provided guidance with respect to important intellectual content. All authors approved the final version of the manuscript.

Ethics

Use of the database for this retrospective study was approved and permitted by the Ethics Committee of the Children's Hospital of Chongqing Medical University.

Conflicts of interest

The authors have no conflicts of interest related to this study.

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Figures

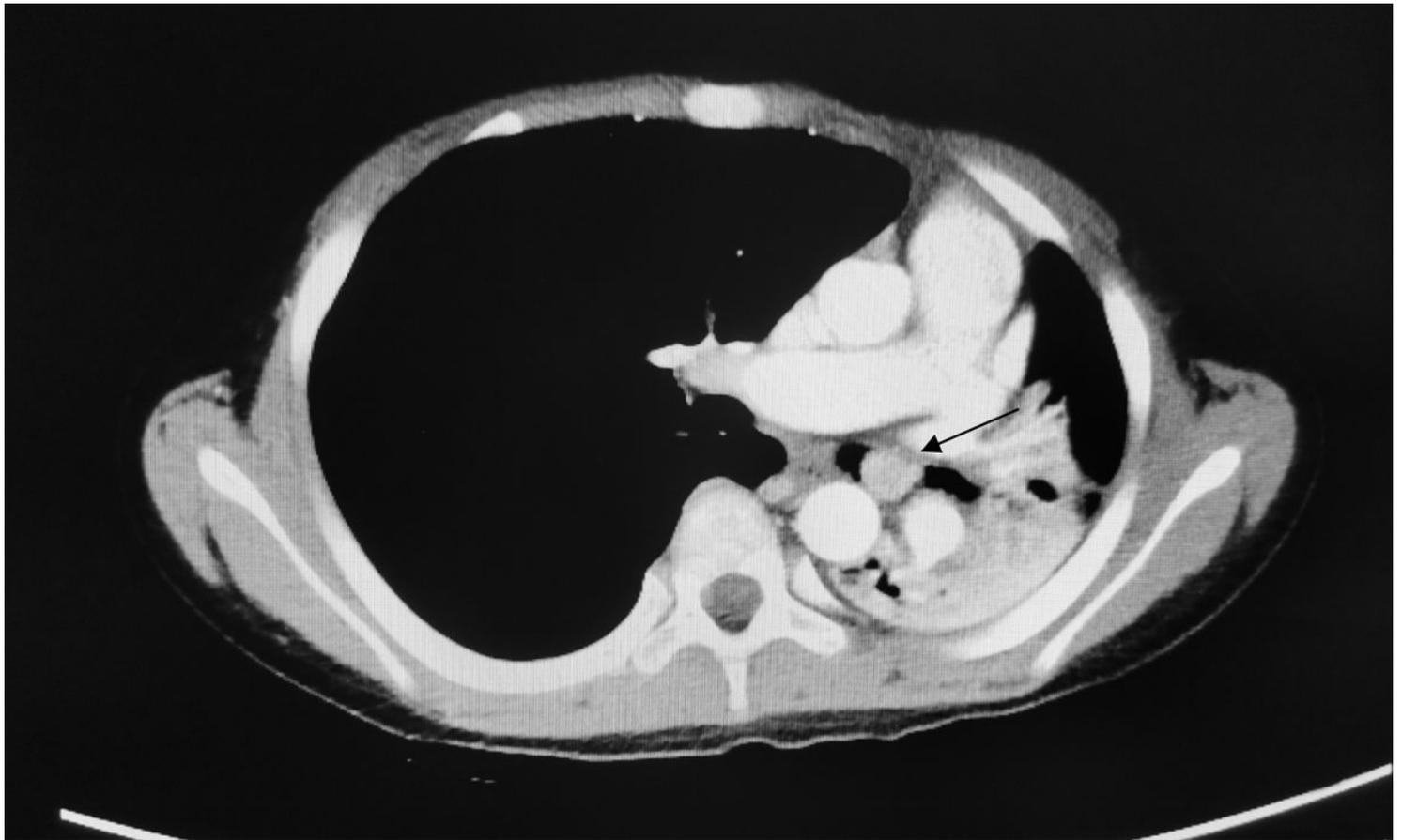


Figure 1

Preoperative computed tomography (CT) scan of patient no. 6, showing a lesion located in the left main stem bronchus (black arrow), obstructing the distal bronchus, as well as atelectasis of the left upper lung.

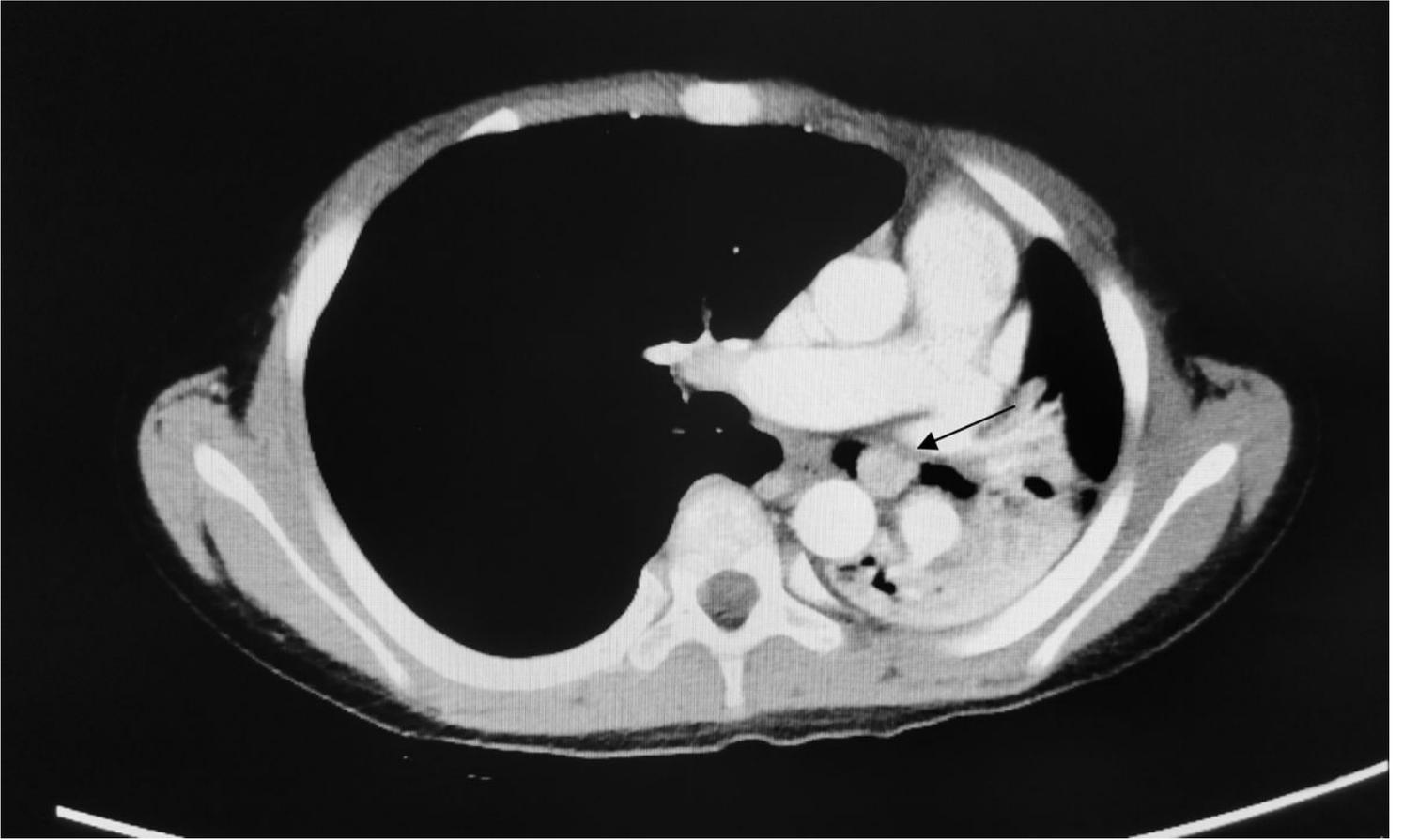


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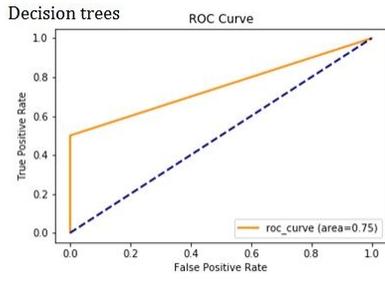
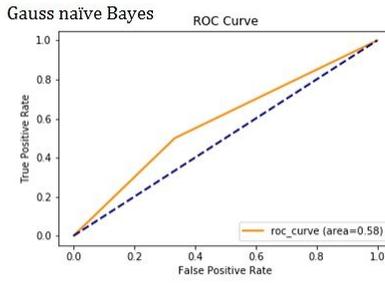
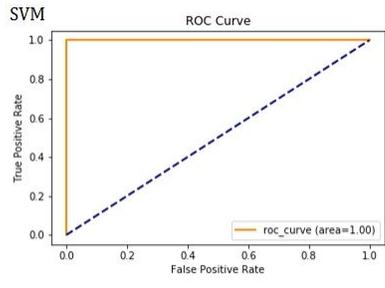


Figure 2

ROC curve for the three models; the area under the ROC curve (AUC) of the SVM is 1.

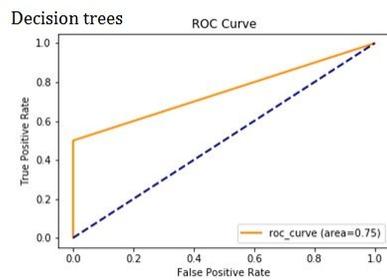
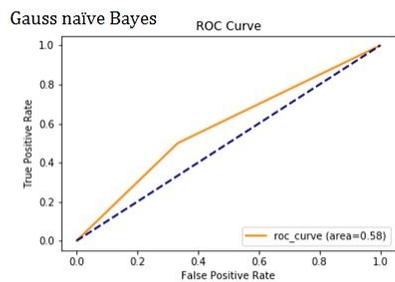
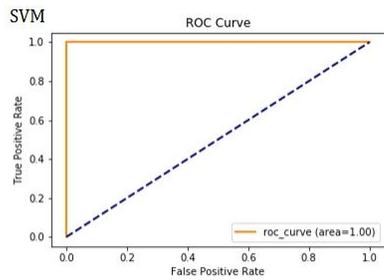


Figure 2

ROC curve for the three models; the area under the ROC curve (AUC) of the SVM is 1.