Case report

A 62-year-old man with stridor and dyspnoea

A 62-year-old man, without any past medical history, but a current 10-pack-year smoker, consulted a pulmonologist at the outpatient department for a gradually worsening dyspnoea over 1 year. Dyspnoea occurred first at exercise but, for 1 week, he felt short of breath, even at rest. He also reported a chronic cough with white sputum and frequent blood streaks. His voice was not affected and he had no swallowing difficulties. He lost 4 kg over the previous 3 months, along with a loss of appetite and tiredness. He did not have fever, night sweats, chest pain or choke.

As a farmer, he had been exposed to pesticides, but never to asbestos or other potentially toxic substances. He had no known tuberculosis contact within his family or workplace and he denied any past medical history of cervical trauma.

His physical examination revealed a stridor and signs of respiratory distress with polypnoea (respiratory rate 32 cycles·min$^{-1}$), and accessory inspiratory muscles use. His body mass index (20.3 kg·m$^{-2}$) and other vital signs (oxygen saturation measured by pulse oximetry ($S\text{\text{o}}_2$) 96% in room air, cardiac frequency 84 beats·min$^{-1}$, blood pressure 130/80 mmHg, temperature 36.8°C) were normal. He looked anxious, and examination of his face showed no cyanosis and a normal nasal and oral mucosa (especially no sign of heartburn, no oedema). Tonsils, soft palate, posterior pharynx and tongue were normal. Cervical spine and cranial nerves examination was normal, and there was no meningism. Examination of his neck revealed use of accessory respiratory muscles, but no asymmetry, mass, tracheal deviation or jugular venous distension. Thyroid and parotid glands were normal, and no cervical or sus-clavicular lymph node was palpable. Stethoscopic examination of the neck and lungs found an inspiratory wheezing. Cardiac and abdominal examination was normal.

Task 1
How can you describe a stridor?
How would you treat the patient in an acute setting and what is the key information to a diagnosis?
A 62-year-old man with stridor and dyspnoea

Main severity signs, common to all aetiologies and very easy to recognise, are described in figure 1. However, causes of stridor are numerous (table 1): anaphylaxis, vocal cord oedema, laryngeal stenosis by a tumour, or following irradiation, endotracheal intubation or trauma, and goitre are the most frequent causes of stridor due to upper airways anomalies in adults [2, 3]. On the tracheal level, causes of stridor can be malignant, especially primary lung cancer, or non-malignant, for example foreign body aspiration, tracheomalacia, complications occurring after a tracheotomy or endotracheal tubing, and non-malignant tumours of the central airways such as papillomas and hamartomas [2, 3]. The clinician should briefly ask very reliable questions to orientate the diagnosis hypotheses. Is it an adult or a child? Is there fever? Is there a witnessed episode of choke? Are there signs of allergy? Is it a traumatic context? Is it an acute and isolated, or a recurrent or progressive episode?

The next step is to secure the airways and assure adequate ventilation with measures depending on the severity signs and the aetiological hypothesis. For example, in the case of a life-threatening foreign body aspiration, a Heimlich manoeuvre should be attempted prior to intubation or tracheotomy (or cricothyrotomy); similarly, adrenaline should be administrated in the case of anaphylaxis; whereas, intubation is required in case of impeding respiratory distress complicating severe epiglottis. After stabilisation of the patient, detailed medical history, associated comorbidities, epidemiological factors, smoking or alcohol consumption, vaccination status, usual treatment, associated symptoms and complete physical examination will help the clinician to orientate towards a proper diagnosis.

Answer 1
A stridor is a monophonic, high-pitched and inspiratory sound, better heard on the anterior neck [1]. It is very important to recognise because it allows to reliably identify that there is an upper airways or central airways obstruction. Stridor should not be confused with wheezing, which is a continuous high-pitched expiratory sound, composed of single or multiple notes, and a feature of lower airways obstruction.

A stridor is a clinical finding that every doctor should know. Indeed, it is a sign of a life-threatening obstruction either on the upper airways or at the tracheal level, requiring emergency appropriate diagnostic and therapeutic interventions.

When hearing a stridor, the physician has to immediately make sure that intubation and tracheotomy material is available and ready to use in the patient’s room. Their second action is to administrate oxygen to the patient by nasal cannula or mask. They have to evaluate quickly, in parallel, the seriousness of the acute respiratory distress and the potential causes of stridor.

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Task 2
If the patient describes a choke, what is the most probable diagnosis, and how do you handle this emergency case?

Figure 1 Algorithm for management of stridor at the emergency department.
Table 1 Causes of stridor with their main diagnosis and therapeutic specificities

| Causes of stridor | Epidemiological factors | Unusual clinical presentation | Immediate management | Other useful tests and treatments |
|------------------|-------------------------|------------------------------|----------------------|----------------------------------|
| **Acute**        |                         |                              |                      |                                  |
| Foreign body inhalation | Children <3 year-old Swallowing disorders Loss of consciousness | Sudden non-febrile stridor Witnessed episode of choke | Heimlich manoeuvre Direct laryngoscopy ± Intubation or tracheotomy/ cricothyrotomy | Chest radiograph, CT scan of thorax Bronchoscopy |
| Anaphylaxis      | Allergies               | Sudden non-febrile stridor Swollen face and urticaria | Parenteral adrenaline injection | ± Intubation or tracheotomy/ cricothyrotomy |
| **Post extubation** |                         |                              |                      |                                  |
| Vocal cords oedema |                         | Stridor immediately after extubation Sore throat, dysphonia, dysphagia Improves in 24–48 h | Nebulisation of adrenaline and steroids |                                  |
| Laryngeal injury | Prolonged intubation Traumatic intubation Large endotracheal tube | Stridor immediately after extubation Hoarse voice or dysphonia | Direct laryngoscopy ± Reintubation | Various therapeutic options |
| Tracheal stenosis | High endotracheal tube balloon pressure | Progressive stridor and dyspnoea after extubation May remain silent for years | Bronchoscopy | CT scan of thorax Various therapeutic options |
| **Trauma/fire**  |                         | Think particularly to life-threatening injury in case of stridor after direct impact on the glottis or trachea | Intubation or tracheotomy/ cricothyrotomy are often needed | Most often surgical treatment |
| **Infections**   |                         |                              |                      |                                  |
| Epiglottis       | Children with immune deficiency Lack of Haemophilus influenzae B vaccination | Rapid respiratory febrile stridor and respiratory distress Upright posture – sniffing position Dysphagia, muffled voice | Do not lie down No tongue-blade examination ± Intubation or tracheotomy/ cricothyrotomy Empiric antibiotics | Neck soft tissue x-ray: thumb sign Epiglottis sample culture Blood cultures |
| Croup            | Children 6 months to 3 year old community epidemic | Progressive febrile stridor upright position barking cough | Oral or parental dexamethasone Nebulisation of adrenaline ± Intubation |                                  |
| Diphtheria       | Lack of vaccination for Corynebacterium diphtheriae | Progressive stridor low grade fever Preceded by rhinorrhoea, sore throat, cervical lymph nodes Grey pseudomembranes in the airways | Empiric antibiotics | Culture of pseudomembranes Droplet’s isolation for patient contacts’ investigation |
| Tracheitis       | Seasonal epidemic of parainfluenza, influenza and RSV | Acute stridor with high fever and respiratory distress Prefers to lie down Toxic appearance | Bronchoscopy Empiric antibiotics | Culture of bronchial aspiration |

(Continued)
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Table 1 Continued  Causes of stridor with their main diagnosis and therapeutic specificities

| Chronic/recurrent | Expansive process | Others |
|-------------------|-------------------|--------|
| **Endoluminal obstruction** | Airways compression | Glottic or tracheal stenosis (prolonged intubation, irradiation, congenital, idiopathic...) |
| Primary laryngeal or lung cancer | Mediastinal mass: | Tracheomalacia, relapsing polychondritis |
| Secondary metastasis: lung, renal, breast, thyroid, colon, sarcoma, melanoma | Malignant: thymus, thyroid, germ cell | Laryngomalacia |
| Benign tumours: papillomatosis, sarcoidosis, amyloidosis, hamartomas | Benign: goitre | Laryngospasm |
| Granulation tissue: post intubation/tracheostomy/stenting, foreign bodies, surgical anastomosis, granulomatosis with polyangiitis | Lymph nodes: | Bilateral vocal cord palsy |
| Mucus plus blood clots | Malignant: metastasis, lymphoma | Vocal cord dysfunction |
| | Benign: tuberculosis | Webs |

**Answer 2**
A witnessed episode of choking (sudden onset of cough and/or dyspnoea and/or cyanosis) is a very reliable sign for foreign body aspiration [4]. It occurs most often in children under 3 years old, and in adults with swallowing disorders or loss of consciousness. The usual clinical presentation is a sudden onset of non-febrile stridor, accompanied by a cough and decreased air entry in a previously healthy child. Adults more often have a chronic cough or recurrent aspiration pneumonia without stridor, but some patients present with an acute respiratory distress and stridor. In case of complete airways obstruction (patient unable to speak or cough, sniffing position, cyanosis, respiratory distress), a Heimlich manoeuvre should be attempted, prior to direct laryngoscopy, which localises the foreign body. The following steps depend on the localisation of the foreign body above or under the vocal cords, on the ability to maintain spontaneous ventilation, and on the success of endoscopic removal (figure 2).

**Task 3**
If your physical examination reveals urticaria and facial oedema, what is the most probable cause of stridor?
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In anaphylaxis [5], the onset of stridor is sudden with associated signs of allergies, such as urticaria and swelling face. Immediate intravenous or intramuscular injection of adrenaline is the first treatment to administrate. Endotracheal intubation or cricothyrotomy may be needed in case of impeding airway obstruction. Hereditary angioedema should be suspected in the absence of response to adrenaline, especially if the presentation is more progressive with lip swelling.

**Figure 2** Algorithm for management of a suspected foreign body aspiration. #: intentional right main bronchus intubation to advance the foreign body with the endotracheal tube, which is then pulled back into the trachea.

**Answer 3**
Anaphylaxis or hereditary angio-oedema.

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**Task 4**
If the stridor occurs after extubation, what causes do you suspect, and what are the key investigations for diagnosis?

**Figure 3** Computed tomography scan of thorax in frontal (a), sagittal (b) and axial (c and d) views. Post-intubation tracheal stenosis, 8 mm of diameter (a, b and c: black arrow; d shows the normal tracheal lumen diameter, below the stenosis) in a 19-year-old woman, which was misdiagnosed for asthma for several years and was not improved with inhaled therapies. Correct diagnosis was established after an episode of acute respiratory distress with stridor. The patient underwent surgical treatment, allowing complete regression of her symptoms and no recurrence of the tracheal stenosis at 4 years follow-up.
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A patient with vocal cord oedema will usually complain of sore throat, dysphonia and dysphagia in addition to stridor, occurring after extubation, and relieved by nebulisation of adrenaline and corticoids. It usually resolves in 24–48 h.

Laryngeal injury includes vocal cord ulceration, granuloma, paralysis and stenosis. It occurs more frequently in case of prolonged intubation, traumatic intubation and large endotracheal tube [6]. The patient usually presents a hoarse voice or a dysphonia immediately after extubation. A laryngoscopic examination is necessary to assess the diagnosis, the type of laryngeal injury, and to guide further therapeutic management.

Tracheal stenosis is caused by high endotracheal tube balloon pressure. The patient usually presents with a progressive onset of dyspnoea and stridor after extubation, but may remain paucisymptomatic with discovery of the tracheal stenosis years after the intubation (figure 3). Bronchoscopy and computed tomography of the thorax are cornerstones for diagnosis and treatment planning. Therapeutic options comprise various endoscopic interventions (stenting, balloon dilatation, laser resection) or surgical reparations.

In very rare cases, post-extubation stridor is due to obstructive fibrinous tracheal pseudomembranes (figure 4) [7]. The presentation is acute, immediately following extubation, but without laryngeal injury on laryngoscopy. Flexible bronchoscopy is sufficient to visualise the pseudomembranes, but rigid bronchoscopy may be necessary to remove them.

**Answer 4**

A stridor occurring shortly after extubation should orientate towards vocal cord oedema, laryngeal injury, obstructing secretions or tracheal stenosis (table 1).

**Task 5**

Why would you look for a stridor in the context of trauma or fire?

**Task 6**

If the patient is a febrile child, what are the most common causes of stridor you should look for?
A 62-year-old man with stridor and dyspnoea

Answer 5
A stridor occurring in the context of trauma or fire is a reliable sign of life-threatening injuries that may be not suspected otherwise, particularly in the case of direct impact on the glottis or on the trachea. If an upper or central airways injury is suspected, securing the airway is essential and often requires endotracheal intubation or tracheotomy.

Answer 6
Upper or central airways infections are very frequent causes of stridor, especially in children. The clinical presentation is usually an acute onset of febrile stridor associated with cough, change in voice and difficulty in swallowing. The most prevalent infections causing stridor are epiglottitis, croup, diphtheria and bacterial tracheitis (table 1) [8].

Epiglottitis [9] usually occurs in children with immune deficiency or lack of Haemophilus influenzae type b vaccination. The onset is rapid, with respiratory distress, upright posture in a sniffing position, dysphagia, muffled voice or aphonia and difficulty handling oral secretions. One should not attempt to visualise the epiglottis by tongue-blade, nor try to lie the patient down. In a life-threatening situation, bag-valve mask ventilation followed by endotracheal intubation or cricothyrotomy if failure to intubate, is needed. Empirical antibiotic treatment with a third-generation cephalosporin and an antistaphylococcal agent should be administered without delay.

Croup [10] is a laryngotracheobronchitis, often viral, occurring in children aged 6 months to 3 years, presenting with a progressive onset of fever, stridor, preference to upright position and typical barking cough. These clinical findings associated with epidemiological factors (community epidemic) are usually enough to assess the diagnosis. Stridor at rest is a severity sign indicating that in-hospital emergency management is required. All children with severity signs of croup should immediately receive intravenous or intramuscular dexamethasone and nebulisation of adrenaline. In cases of very severe respiratory distress, endotracheal intubation may be needed.

Respiratory diphtheria [11] is caused by toxins of Corynebacterium diphtheriae in patients without vaccination. The child presents with gradually increasing rhinorrhoea, sore throat, cervical lymph nodes and low-grade fever, followed by the apparition of an enlarged tonsil covered by grey pseudomembranes that extend rapidly to the pharynx, larynx, nasal passages and tracheobronchial tree. Culture of pseudomembranes and toxin detection allow the diagnosis. Empirical treatment with diphtheria antitoxin and antibiotics (erythromycin or penicillin) should be started without delay. Contact investigation for prophylactic treatment and patient’s droplet isolation are necessary to avoid epidemic spread of the disease.

Bacterial tracheitis [12] occurs in older children. It is a bacterial infection after a viral respiratory tract infection. It usually happens in the winter, during the seasonal epidemics of parainfluenza, influenza and respiratory syncytial viruses. Staphylococcus aureus is the most commonly involved bacteria. The child presents with high fever, stridor, toxic appearance and respiratory distress occurring progressively 1–3 days after signs of acute respiratory viral infection. In the case of respiratory distress, endotracheal intubation is needed. In contrast with other infectious causes of stridor, the child prefers to lie down. Definitive diagnosis is established by bronchoscopy showing oedema and erythema of the trachea with purulent secretions, and allowing sampling for microbiology studies. Empirical antibiotic treatment with antistaphylococcal agent and third-generation cephalosporin or penicillin should be started quickly.

Task 7
If the patient presents with a chronic recurrent history or a progressive onset of stridor, what diagnosis do you suspect?
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A benign or malignant expansive process can provoke endoluminal obstruction or extrinsic compression of the trachea or upper airways. On the tracheal level, the onset of stridor is progressive, and often preceded by a slowly growing dyspnoea. Indeed, dyspnoea occurs on exertion when the tracheal lumen is below 8 mm, and at rest when the tracheal lumen is below 5 mm. Most of the time, epidemiological factors (for example smoking or alcohol abuse for laryngeal, lung and oesophageal cancers), medical history, associated symptoms and constitutional signs, physical examination, cervical and/or thoracic computed tomography and airways’ endoscopies orientate the diagnosis and further management.

Tracheomalacia [13] is a tracheal weakness due to cartilage impairment or to excessive hyperdynamic collapse. It is most often a congenital disease in children and an acquired condition in adults. Recurring episodes of stridor associated with cough and acute onset of dyspnoea, triggered by an infection, in patients at risk of tracheomalacia is suggestive of this diagnosis. A history of prior prolonged endotracheal intubation or tracheostomy, tracheal trauma, or recurrent use of steroids in COPD patients are frequent causes of tracheomalacia. Thorax computed tomography with evaluation of the trachea in inspiration and expiration can demonstrate the airway collapse (posterior tracheal wall is flat, tracheal diameter is smaller and lung attenuation is increased). Spirometric flow–volume loop shows a low peak expiratory flow rate followed by a rapid decrease of flow. Bronchoscopy confirms the diagnosis when finding a >70% decrease of the tracheal lumen. Treatment of tracheomalacia depends on its extension and its cause and can include stenting, surgery or non-invasive ventilation.

Vocal cord dysfunction (paradoxical cord motion) [14] is an inappropriate movement of the vocal cords often associated with psychological disorders, in response to inducible triggers such as exercise, irritants, emotions and gastro-oesophageal reflux. It is often misdiagnosed as asthma. Direct laryngoscopy shows abnormal adduction of the true vocal cords at mid-inspiration.

**Answer 7**

Sometimes, the clinician faces up to stridor occurring in a patient with a history of progressive shortness of breath, often misdiagnosed as asthma or COPD. Several causes are possible, including compression or obstruction of the upper or central airways by a malignant or benign expansive process, tracheomalacia, and congenital diseases (table 1). Importantly, patients with chronic cause of stridor may present at any time an acute respiratory distress indicating a critical obstruction due to evolution of the underlying disease.

In our patient, blood tests showed normal total blood cell count, coagulation tests, electrolytes, creatinine and C-reactive protein. Blood gas analysis was not performed.

**Task 8**

What is the value of blood tests in the evaluation of a stridor?

**Answer 8**

Blood tests are not essential, and they should not delay appropriate emergency treatment. Increased white blood cells and C-reactive protein can orientate towards an infectious cause. Platelets, coagulation tests, clearance of creatinine are useful before invasive procedures and contrast computed tomography scanner. Blood gas analysis is important to look for respiratory acidosis, sign of severe respiratory failure.

**Task 9**

What is the value of thorax imaging in the evaluation of a stridor? What imaging test would you perform in our patient and why?
A 62-year-old man with stridor and dyspnoea

Figure 5 Computed tomography scan of thorax axial view (a), coronal view (b), sagittal view (c). A tissue-density mass (white arrow) arises from the right anterolateral tracheal wall and nearly completely obstructs the tracheal lumen.

Answer 9
As with blood tests, imaging should not delay emergency airway management of a life-threatening situation. A chest radiography can show mediastinal deviation due to a compressive mass, a lung collapse due to an obstruction of the homolateral main bronchus, or a radio-opaque foreign body. Computed tomography of thorax shows the same lesions as chest radiography, but with better accuracy and better assessment of their nature, origin, shape, vascularisation and relationship with the contiguous structures. Magnetic resonance imaging of the thorax would be appropriate in a non-acute setting, if a congenital vascular ring is suspected.

For our patient, computed tomography of the thorax was performed (figure 5). This radiology exam was preferred to chest radiography because it has a better accuracy to detect a subglottic or tracheal tumour or stenosis, which is the most probable diagnosis in this patient, regarding his age, risk factors, progressive onset of the stridor along with general status alteration, apyrexia, absence of choke and normal laryngoscopy findings.

Answer 10
Computed tomography of the thorax showed a large tracheal mass, developed on the right wall of the distal part of the trachea, drastically obstructing its lumen.

This tracheal mass can be a benign or malignant expansive process which provoke endoluminal obstruction of the trachea. Malignant process invading the trachea come most often from contiguous organ tumour (lung, larynx, oesophagus, thyroid, lymph nodes) but may also arise primitively from the trachea (most frequently squamous cell carcinoma and adenoid cystic carcinoma). Distant metastasis of melanoma, breast, renal and colon cancer can also be seen in the trachea. The most frequent benign tracheal tumours are hamartoma, leiomyoma, squamous cell papilloma and neurogenic tumours.

Task 10
Look at the computed tomography image (figure 5). What did it reveal and what are the subsequent diagnostic considerations?

Task 11
What is the diagnostic value of pulmonary function tests in the evaluation of a stridor?
A 62-year-old man with stridor and dyspnoea

Our patient did not perform pulmonary function test due to his life-threatening presentation at our facility, and he did not consult a respiratory physician before.

Compared with flexible bronchoscopy, rigid bronchoscopy allows mechanical resection of the tracheal mass, with very good control of bleeding; but also thermocoagulation, laser or cryotherapy. If needed, a tracheal endoprosthesis can be inserted during the same operating time.

In our patient, rigid bronchoscopy was performed (figure 6). It revealed a round, well-limited, regular, firm, non-friable, hypervascularised, white tumour, arising from the right tracheal wall, just above the carina, and nearly completely obstructing the tracheal lumen. This mass was removed by electrocauterisation, with no subsequent complication. Pathology examination (figure 7) found a grade I chondrosarcoma.

Discussion

A stridor is a “to-be-known” clinical finding. Indeed, it is a sign of a life-threatening obstruction either on the upper airways or on the tracheal level. This obstruction can be life threatening, requiring emergency appropriate diagnostic and therapeutic interventions. Correct management of the patient presenting with stridor depends on the presence of signs of severe respiratory distress, and on the presumptive cause.

Chondrosarcoma is a very rare cause of stridor. It is a primary malignant tracheal tumour consisting predominantly of cartilage [15–18]. Most tracheal chondrosarcomas arise from the posterolateral wall of cartilaginous rings. It can be located anywhere from the cricoid cartilage to the proximal bronchial tree. As other tracheal tumours, the clinical presentation is various, depending on the importance of the tracheal obstruction. Other symptoms are not specific and include cough, haemoptysis, hoarseness and recurrent chest infections. Often misdiagnosed as asthma or COPD, poor response to inhaled therapies should prompt medical attention. Chest radiography is often non-contributive; however, computed tomography of the thorax is a very useful investigation, allowing a precise evaluation of the lesion (location, size, calcifications, degree of obstruction and extra-tracheal extension of thorax) and of its environment (tracheal wall thickness, lymph node involvement). Bronchoscopy is the cornerstone investigation for diagnosis and sometimes treatment. Chondrosarcoma appears like a shaped, firm, multi-lobulated white mass arising from the cartilage rings. Both flexible and rigid bronchoscopy permits evaluation of the rapport between the vocal cords and the proximal pole of the lesion, and its distal extension. This information is essential to plan surgical treatment. Biopsies can be performed by flexible or rigid bronchoscopy; nonetheless, the latest allows better control in case

Answer 12
Rigid or flexible bronchoscopy.

Answer 11
In an acute setting, pulmonary function tests can often not be performed by a critically ill patient. However, if the patient presents with a chronic history of progressive dyspnoea, the presence of an impairment of the inspiratory and expiratory debit-volume curve (plateau aspect) is a clue orientating the clinician towards a tracheal obstruction.

Our patient did not perform pulmonary function test due to his life-threatening presentation at our facility, and he did not consult a respiratory physician before.

Task 12
What is the recommended investigation to propose for diagnosis and emergency treatment of our patient?
of bleeding. Moreover, rigid bronchoscopy allows urgent concomitant resection of the tumour, thus re-establishing airway patency. On pathology exam, chondrosarcomas can be classified into three grades, depending on cellularity, atypical and pleomorphic findings.

Complete surgical resection of the tumour with end-to-end anastomosis of the trachea is the gold-standard treatment of chondrosarcoma, preconised to avoid recurrence [15–18]. Nonresectable chondrosarcomas may benefit from palliative rigid bronchoscopic removal and radiotherapy.

Clinical course

Our patient’s symptoms disappeared immediately after bronchoscopy. He refused to have a complementary surgical treatment. He was regularly followed for 3 years by clinical examination, flexible bronchoscopy and computed tomography of the thorax, without any sign of local recurrence. He progressively regained weight and returned to a normal life.

Conclusion

Stridor is an emergency and a high-value clinical sign, immediately orientating the clinician towards an upper airways or central airways obstruction, avoiding non-useful investigations and time wasting; especially since some situations may be life threatening.

Patient age, detailed history, associated comorbidities, smoking or alcohol consumption, epidemiological factors, vaccination history and associated symptoms help the clinician to orientate towards proper diagnosis and towards deciding how to manage the patient.

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Conflict of interest

None declared.

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