

## Novel diagnostic procedure

## Psittacosis infection and tracheobronchomalacia in a patient undergoing thyroidectomy

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A 34-year-old woman with papillary thyroid carcinoma underwent total thyroidectomy with central and lateral lymphadenectomy. Immediate airway compromise required re-intubation immediately after surgery. Marked tracheal and bronchial collapse of greater than 50% of the lumen indicated tracheobronchomalacia. Subsequent attempts at extubation failed over the next week. The patient soon developed evidence of a lower respiratory tract infection. Empirical treatment with penicillins was unsuccessful. A clinical suspicion of chlamydia infection prompted initiation of macrolide treatment followed by resolution of both the patient's respiratory infection and tracheobronchomalacia. Serology returned positive for *Chlamydophila psittaci* infection. It later transpired that the patient had symptoms of an upper respiratory tract infection just prior to surgery. This case demonstrates an interesting and unreported cause of tracheobronchomalacia as well as providing a good lesson on the importance of preoperative screening for infection.

**BACKGROUND**

Optimising perioperative care is paramount in any surgical case. Pulmonary vulnerabilities and associated complications have significant impact on perioperative morbidity and mortality.<sup>1–2</sup> Preoperative screening for respiratory tract infections routinely informs a deferral of elective surgery. Consequently, data analysing its impact on outcome are limited.

Psittacosis is a zoonosis caused by the obligate intracellular bacterium *Chlamydophila psittaci*.<sup>3</sup> Its name pertains to the parrot family of Psittacidae. The atypical respiratory tract infection is usually directly acquired from close contact with infected birds.<sup>4</sup> Confirmed cases of human psittacosis are relatively rare. In 2008, only 61 cases were reported in England and Wales, though this is thought to be a marked underestimation.<sup>5</sup> Underlying a likely misrepresentation is the wide spectrum of presentations from asymptomatic to life-threatening – as defined by the Centres for Disease Control (CDC) and the UK Health Protection Agency.<sup>4–7</sup> Subclinical infections are usually self-limiting. They have uncertain impact in the context of surgery.

We report the case of a young woman who, during an elective total thyroidectomy and modified radical neck dissection, developed an unusual life-threatening complication secondary to a co-existent initially undiagnosed respiratory tract infection caused by *C psittaci*.

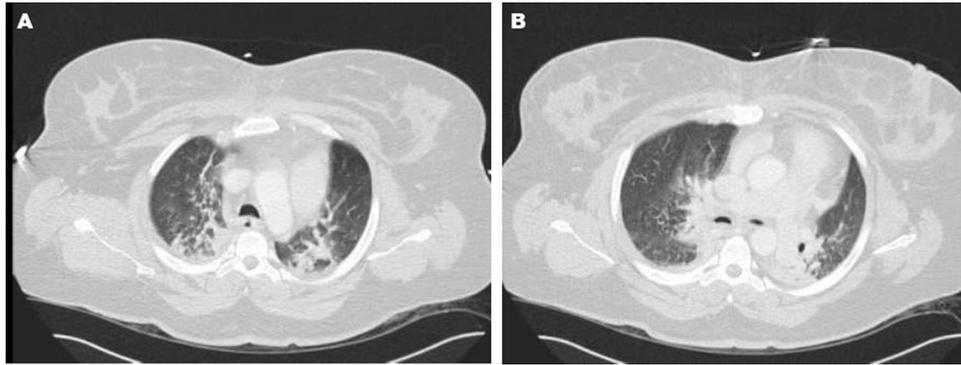
**CASE PRESENTATION**

A 34-year-old, previously healthy woman and non-smoker, presented with a left-sided thyroid nodule and two palpable left-sided level V lymph nodes. She was clinically euthyroid with a thyroid stimulating hormone of 1.90

(normal range 0.30–5.50 mU/l), and a free thyroxine 14.1 (9.0–25.0 pmol/l). Fine needle aspiration of the thyroid nodule established a microfollicular pattern and histology of a lymph node biopsy found extensive infiltration with a metastatic papillary thyroid carcinoma. Subsequent CT delineated the left-sided thyroid nodule, a large left-sided level V lymph node and a level III lymph node. Small volume lymph nodes were also noted in the central compartment. Based on these findings it was decided that the patient would most benefit from a total thyroidectomy and bilateral modified radical neck dissection with level II to VII clearance.

On admission, 12 h prior to surgery, the patient denied any new symptoms. Full clinical examination was equally unremarkable and vital signs were all within normal limits. Preoperative routine blood tests showed normal renal and liver function, a C-reactive protein of less than 5 mg/l and a mild lymphocytosis of 4.3 (1.4–4.0×10<sup>9</sup>/l). In theatre, the patient had an uneventful anaesthetic induction and endotracheal intubation. Total thyroidectomy with modified radical neck dissection was performed successfully with no immediate directly associated complications – recurrent laryngeal nerve intact bilaterally, satisfactory haemostasis achieved, parathyroid glands preserved and no tracheal resection as there was no tracheal invasion.

Postoperatively on extubating the patient and on removing positive end-expiratory pressure (PEEP) there was acute marked oxygen desaturation requiring immediate re-intubation. Emergency bronchoscopy revealed collapse of greater than 50% of the lumen of the lower trachea and large bronchi indicative of tracheobronchomalacia. Subsequent attempts at reducing the PEEP were



**Figure 1** CT chest and neck study during expiration. Note the greater than 50% collapse of both the trachea (a) and the bronchi (b).

also associated with immediate lower airway collapse and compromise. Chest radiograph after re-intubation showed no initial evidence of intrapulmonary disease. The patient was kept intubated on an intensive care ward subsequently. A chest film 4 days later revealed opacification of the left lower zone suspicious for consolidation. These radiographic observations were accompanied by fever and respiratory compromise with increasing neutrophilia noted from 8.97 (2.20–6.30  $10 \times 9/l$ ) at day 4 postoperation to 14.4 at day 7. Based on signs of infection and an early sputum culture isolating both *Staphylococcus aureus* and group A streptococcus (both sensitive to penicillin) the patient was commenced on intravenous co-amoxiclav. Despite penicillin the patients inflammatory markers continued to increase and further attempts to extubate and reduce PEEP were again associated with immediate respiratory compromise secondary to tracheal and bronchial collapse. As the airway compromise was persisting beyond 10 days postoperatively, the patient underwent tracheostomy.

Further elucidation of the patient's condition via a dynamic CT neck and chest study showed prominent collapse of the right and left main bronchi distal to the tip of the tracheostomy tube (figure 1). Also noted on CT was patchy consolidation in both lungs, greatest in the left lower lobe, and evidence of pulmonary hypertension with prominence of the main pulmonary artery. As no organisms were identified with repeat blood and sputum cultures an atypical pneumonia serology screen was performed and the patient was started on macrolide therapy to cover causes of atypical pneumonia. Complement fixation found a significant increase in the chlamydia antibody titre and subsequent micro-immunofluorescent antibody assay found a specific, over four fold, increase in IgG to the *C psittaci* strain of 1:512 (<1:64).

#### TREATMENT

The patient was commenced on the macrolide antibiotic erythromycin intravenously in high standard doses.

#### OUTCOME AND FOLLOW-UP

On instigation of erythromycin there was a rapid improvement in the patient's clinical condition with normalisation of white cell count and radiographic resolution of the lower

zone consolidations. Repeat dynamic CT neck and chest study, 14 days after the first CT scan and after the course of erythromycin, showed normal airways on both inspiratory and expiratory phase. Clinical improvement was concluded with successful weaning of ventilatory support and eventual tracheostomy decannulation.

Post recovery, on retrospective focused questioning of the patient, it was discovered that she kept geese and that 2 weeks prior to surgery, she had undertaken cleaning of her goose enclosure. The patient also reported that 2 days before admission she had developed a sore throat, which from fear of delaying her surgery the patient had chosen not to disclose.

#### DISCUSSION

This case illustrates an unusual complication of elective surgery and an unfamiliar presentation of *C psittaci* infection. It is our supposition that the patient contracted the infection through inhalation of aerosolised organism from cleaning her bird coop.<sup>8</sup> With subsequent intubation and ventilation, the psittacosis was likely spread throughout the respiratory tract. We propose that the *C psittaci* infection, disseminated to the trachea and bronchus, then led to dispersed inflammatory infiltration, oedema and localised necrosis which in turn undermined the integral strength of the tracheal wall. This inflammatory associated weakening could have been further compounded by the trauma of intubation, local surgery and perhaps oxygen toxicity. These combined factors ultimately resulting in the diffuse tracheobronchomalacia are seen on extubation. Underlying this suggested pathophysiology is therefore both a rare clinical picture of *C psittaci* infection and a poorly characterised cause of tracheobronchomalacia (based on a journal search performed February 2011 see appendix 1).

Tracheobronchomalacia (TBM) is a disease of the central airway in which weakness of the tracheal and bronchial walls develop due to damage to the supporting cartilage, predisposing to airway collapse on exhalation.<sup>9–11</sup> In adults, common acquired causes include prolonged intubation, severe emphysematous disease, chronic inflammation and recurrent respiratory infections.<sup>12–14</sup> Refuting the argument that intubation alone may have caused the above cases TBM, is the short duration of intubation (3–4 h) and that there was observed diffuse tracheal and

bronchial weakness, whereas typically, intubation associated TBM is characterised by a segmental weakness from localised pressure necrosis from the cuff.<sup>12–14</sup> Another possible confounding aetiology is that there are several reported cases of TBM secondary to goitres. These cases are however, generally associated with large compressive goitres and/or tracheal invasion, neither of which were a feature of this case.<sup>15–19</sup> Furthermore in a recent review of 1969 patients with large retrosternal goitres, there were few reported cases of postoperative tracheal collapse.<sup>20</sup> We also put forth that goitre associated malacia would be localised to the area of the trachea which was in direct apposition to the thyroid, unlike the weakness noted in this case which extended into the bronchi. Related to this is the argument that the process of thyroidectomy in itself could lead to local inflammation and malacia. The generalised nature of the TBM again refutes this as the sole cause in this case, additionally a retrospective review of thyroidectomy suggested minimal risk of TBM with modern thyroid surgery.<sup>21</sup> In contrast, supporting a primary infectious aetiology, is the observation that recurrent lower airways infection, and therapy-resistant, irreversible, and/or atypical asthma, as well as regional inflammatory diseases (oesophageal or aortic) are well-characterised factors associated with a diffuse airway malacia.<sup>22–23</sup> With the evidence that TBM is rare in adults without chronic respiratory conditions it could also be argued that this patient may have had an unusual genetic predisposition to TBM. We feel however that this is unlikely in a patient who had no significant respiratory medical history and that the unusual combination of infection, intubation and surgery is enough to rationalise the atypical presentation.

Looking at *C psittaci* and extending into the Chlamydia genus there are no previous documented cases of these infections leading to TBM (based on a journal search performed February 2011 see appendix 1). Nevertheless we feel that the patient had a concomitant psittacosis, which complicated her surgery. Tallying with this is the observed approximate 10 days incubation period between exposure and infection, and the radiographic findings of lower zone consolidation both of which are consistent with recent psittacosis.<sup>4–24</sup> Moreover the serological evidence, at 15 days into infection, of an observed over fourfold increase in IgG antibodies to *C psittaci* on micro-immunofluorescent antibody assay is consistent with a probable diagnosis of psittacosis, as classified by the CDC.<sup>25</sup> Finally, supporting an infectious aetiology is the dramatic clinical improvement and resolution of the airway collapse post macrolide therapy, a well-recognised treatment of psittacosis.<sup>26–27</sup>

In closing, although this case highlights a life-threatening complication of surgery secondary to psittacosis, specific routine preoperative screening for *C psittaci* is arguably not justifiable due to the supposed rarity of this presentation and possible resource limitations. Nevertheless this case heavily highlights the essential need for a detailed clinical examination and history prior to surgery. It is our belief that if, in this patient's case, the pre-existent respiratory tract infection had been diagnosed and empirically treated, with an appropriate delay in surgery, tracheobronchomalacia and the associated high morbidity would have been avoided.

### Learning points

- ▶ On admission for surgery a thorough clinical examination and history is essential in all cases to reduce the risk of major complications.
- ▶ If a preoperative concomitant infection is diagnosed then surgery should be delayed or at the very least related complications expectantly managed.
- ▶ *C psittaci* is widely thought to be an under diagnosed infection, clinicians should be aware of it as it is often readily treatable.
- ▶ Even minor respiratory tract infections can be life-threatening in the context of major surgery.
- ▶ In cases of tracheobronchomalacia an infectious aetiology, such as psittacosis, should be considered.

**Competing interests** None.

**Patient consent** Obtained.

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## APPENDIX

1. On the 10th of June 2001 a journal search for the terms – tracheobronchomalacia/ tracheomalacia/ tracheitis and chlamydia, tracheobronchomalacia/ tracheomalacia/ tracheitis and chlamydia, tracheobronchomalacia/ tracheomalacia/ tracheitis and psittacosis – in Pubmed, Cochrane library, EMBASE, MEDLINE, BMJ Case reports returned 0 relevant results.

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