CASE REPORT

A rash and a rare cause of unilateral diaphragmatic paralysis

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Accepted 15 February 2017

SUMMARY

A 61-year-old man who was an ex-heavy smoker presented to our ambulatory care centre with a 4-week history of dyspnoea on mild exertion. 2 weeks prior to his symptoms, he had developed right-sided cervical herpes zoster for which he was prescribed oral acyclovir by his general practitioner. On examination, a rash over the right C4–5 dermatomes was noted and dullness on percussion of the right mid and lower zones with markedly reduced air entry. His chest radiograph showed a raised right hemi-diaphragm with associated right middle and lower lobe collapse. Further investigation with CT and bronchoscopy did not identify a cause and showed no evidence of underlying malignancy or endobronchial obstruction. An ultrasound 'sniff test' was performed to confirm diaphragmatic paralysis. We present a rare case of cervical herpes-induced diaphragmatic paralysis, and summarise our approach and the current understanding of this interesting condition.

BACKGROUND

This is an important case that highlights a rare complication of a common condition. The association between unilateral diaphragmatic paralysis with cervical herpes zoster (shingles) is not well known and therefore not made often. Beyond educating readers on this topic, we also remind them of the importance of investigating for more common and more serious causes of a presentation before diagnosing rare conditions or sequelae.

CASE PRESENTATION

A 61-year-old man who was an ex-heavy smoker presented to our ambulatory care centre with a 4-week history of dyspnoea on mild exertion. Two weeks prior to his symptoms, he had developed right-sided cervical herpes zoster for which he was prescribed oral acyclovir by his general practitioner. The patient initially attributed his symptoms to his antiviral medication; however, his symptoms persisted after completing his course. He also reported ~5 kg of weight loss in the preceding month. He denied a cough, haemoptysis, chest pain or a fever. There was no history of recent travel.

On inspection, a partially healed cervical herpetic rash over the right C4–5 dermatomes was evident (figure 1). On chest examination, there was dullness on percussion of the right mid and lower zones with markedly reduced air entry. There was no significant cervical lymphadenopathy or any evidence of ipsilateral Horner's syndrome.

Figure 1 Photograph of the patient showing a partially healed cervical herpetic rash over the right C4–5 dermatomes.

INVESTIGATIONS

His blood tests which included a full blood count, urea and electrolytes, C reactive protein, liver function tests and coagulation profile were all within normal parameters; however, his chest radiograph (figure 2) identified a raised right hemi-diaphragm with associated right middle and lower lobe collapse.

A CT scan of his chest was performed in view of his background of heavy smoking and recent weight loss. This showed no evidence of underlying malignancy or endobronchial mass and any mediastinal or hilar lymphadenopathy. It confirmed the right lower and middle lobe collapse and concluded that it appeared to be secondary to an unexplained elevated right hemi-diaphragm.

Our next diagnostic investigation was an ultrasound sniff test, which confirmed that there was right diaphragmatic paralysis.

It was deemed appropriate to further investigate the patient by performing bronchoscopy with trans-bronchial right lower lobe biopsies. This also did not identify evidence of endobronchial disease and the biopsies were negative with no cancerous cells identified.

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To cite: Ashkir ZM, Tsaknis G. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-218440

Figure 2 Chest radiograph showing an elevated right hemi-diaphragm and right lower and middle lobe collapse.

OUTCOME AND FOLLOW-UP

At 6 months follow-up, the patient continued to experience shortness of breath on moderate exertion and still needed regular amitriptyline for postherpetic neuralgia.

DISCUSSION

Unilateral diaphragmatic paralysis is usually an incidental finding on chest radiographs. It is most commonly caused by phrenic nerve injury from cardiothoracic and cervical spine surgery, neck trauma and compression from cervical spondylosis or mass lesions.¹ Inflammatory, infective and neuromuscular causes are rare and are more likely to cause bilateral diaphragmatic paralysis.

Cervical herpes zoster infection is a rare but recognised cause of unilateral diaphragmatic paralysis, and one, which has not been studied in depth.

The intense inflammatory process and subsequent neurological injury associated with herpes zoster reactivation can extend beyond the dorsal root ganglion into the anterior spinal cord and affect the anterior horn cells and/or motor nerve roots.² At the C3–5 level, this can lead to phrenic nerve palsy and result in unilateral diaphragmatic paresis or paralysis.

The only published literature review (of 29 cases) performed by Oike *et al*³ identified older age and previous history of respiratory disease as risk factors to developing unilateral diaphragmatic paralysis secondary to cervical herpes zoster.

Diaphragmatic paralysis appears to occur at least 2 weeks after eruption of the characteristic herpes zoster rash, which is always ipsilateral.

When considering a diagnosis of diaphragmatic paralysis secondary to cervical herpes zoster, initial investigation should aim to rule out more common causes of atelectasis and unilateral diaphragmatic paralysis. A thorough history and physical examination is therefore essential.

Particular consideration not to miss malignancy is important, especially in patients (such as ours) who have concerning risk factors and/or symptomatology. CT, MRI and invasive modalities such as bronchoscopy and CT-guided lymph node biopsy may therefore be warranted in some cases.

To confirm diaphragmatic paralysis, ultrasound or fluoroscopic ‘sniff test’ can be used to demonstrate this in real time; alternatively, phrenic nerve conduction and diaphragm electromyography studies can be used.

Beyond the distinctive herpes zoster erythematous rash, gadolinium-enhanced MRI can be used to identify cervical herpes zoster as the underlying cause. This may show contrast enhancement of the anterolateral spinal cord at the level of C3–5 in keeping with herpes zoster myelitis⁴ or localised leptomeningitis.

Unfortunately, in the vast majority of reported cases of herpes zoster-induced diaphragmatic paralysis, there was no resolution.² Although in some cases, patients received of antiviral therapy, there is not enough evidence to suggest any significant benefit in terms of recovery.

The mainstay of management is therefore conservative and supportive therapy (like for any other cause of diaphragmatic paralysis), including physical rehabilitation, optimising treatment of any concomitant respiratory disease and serial pulmonary function tests to monitor for restrictive deficits.

Learning points

- ▶ Cervical herpes zoster can lead to permanent unilateral diaphragmatic paralysis.
- ▶ It is a rare cause of diaphragmatic paralysis and as such should be considered a diagnosis of exclusion. More common causes should be excluded, particularly malignancy in patients with risk factors.
- ▶ Management of cervical herpes zoster infection-induced diaphragmatic paralysis is mostly conservative.

Contributors ZMA contributed to all aspects of this article. GT made substantial contributions to conception and revision of this article.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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