

## Anterior fracture dislocation of the odontoid peg in ankylosing spondylitis as a cause for rhinolalia clausa: a case study

J M HILTON, P TASSONE, J HANIF, B BLAGNYS\*

### Abstract

We present an unusual cause of rhinolalia clausa secondary to an oropharyngeal mass. A 69-year-old male presented to the otorhinolaryngology clinic with a one year history of a 'plummy' voice. He had a longstanding history of severe ankylosing spondylitis. Examination revealed an obvious hyponasal voice and a smooth hard mass in the midline of the posterior nasopharyngeal and oropharyngeal walls. Subsequent computed tomography scans and lateral plain neck X-ray showed a fracture dislocation of the odontoid peg, secondary to ankylosing spondylitis, which had eroded through the body of the C1 vertebra to lie anteriorly, resulting in the aforementioned impression into the pharyngeal mucosa. The radiological images, the role of the nasal airways in phonation and the causes of hyponasal speech are discussed.

**Key words:** Speech Disorders; Odontoid Process; Nasal Obstruction; Nasopharynx; Ankylosing Spondylitis

### Introduction

Referral of patients with dysphonia is a common occurrence in otolaryngology practice and any patient over the age of 40 with persistent symptoms must have an examination of the larynx to exclude sinister lesions. The aetiology of dysphonia is usually secondary to disease in the larynx affecting voice production. However, quality of voice is also affected by the status of the nasal airway. Excessive nasal air escape, as sometimes occurs in patients with cleft palate (rhinolalia aperta), or significant nasal obstruction, for example in patients with nasal polyps (rhinolalia clausa), can change the quality of sound on phonation.<sup>1</sup>

We present an unusual case of dysphonia secondary to nasopharyngeal obstruction, as a result of C1/C2 vertebral subluxation/dislocation in a patient with ankylosing spondylitis.

### Case report

A 69-year-old male was referred to the department of otolaryngology with an altered voice. The patient, a keen singer, was concerned that his voice had become increasingly 'plummy' and nasal over the last year. More recently he had developed mild dysphagia to solids, but was still able to eat everything he desired. He complained of no otalgia, odynophagia, hearing loss, reflux, rhinorrhoea or nasal symptoms. There was no report of recent trauma to the head or neck. The patient's past medical history was unremarkable except for a long history of ankylosing spondylitis.

On examination the patient had an obvious hyponasal quality to the voice. Examination of the oropharynx revealed a diffuse mass in the midline of the oropharynx,

extending up to the nasopharynx. This was firm, but was not fluctuant, tender or pulsatile. The overlying pharyngeal mucosa was healthy. The patient displayed signs of quite advanced ankylosing spondylitis including loss of normal cervical lordosis. There were no palpable lumps in the neck. The cranial nerves were intact and there were no signs of any cervical neuropathy.

A plain lateral neck X-ray and neck computed tomography (CT) scan were organised and these revealed extensive fusion of cervical and upper thoracic vertebral bodies and posterior elements below the level of C2, in keeping with ankylosing spondylitis. C2 was displaced 11 mm forward in comparison to C1 and the odontoid peg had migrated through the anterior arch of C1 to lie anteriorly (Figure 1). It was this fracture dislocation of the odontoid peg at the atlanto-axial junction that caused the significant indentation into the oropharynx (Figure 2). There was also some forward slip of C1 with respect to the occiput and likely fusion at the atlanto-occipital joint. New bone growth superiorly encroached upon the spinal canal, but there was no evidence of cord compression. Overall the radiological findings were consistent with the chronic spinal changes of ankylosing spondylitis including an apparently old fracture dislocation of the base of the peg which was indenting the oropharynx. In view of these findings the patient was referred to orthopaedic spinal surgeons.

### Discussion

The production of normal speech requires controlled expiration through vibrating vocal folds. The sound waves produced are altered by movement of the tongue, lips and mouth. The quality of the sound is affected by

From the ENT Department and the \*Speech and Language Department, Norfolk and Norwich University Hospital, Norwich, UK. Presented in *précised* form as a poster presentation at the British Rhinology Society Meeting in 18 May 2006, Swansea, UK. Accepted for publication: 31 January 2007.



FIG. 1

Plain cervical spine X-ray showing odontoid peg anterior to the body of C1 protruding beneath the pharyngeal mucosa.

resonance through various air filled spaces. The thoracic cavity resonates sound in the subglottic region. The supra-glottic resonators include the vestibule of the larynx, the oro- and nasopharynx, the mouth and the nose. Nasal resonance adds a beauty, richness and projection to the voice, which is especially relevant to singers. When the nose is blocked, as in a common cold, these qualities are lost and the voice becomes 'plummy'.



FIG. 2

Sagittal CT showing ankylosis of the cervical vertebrae and fractured odontoid peg beneath pharyngeal mucosa.

Nasal resonance is also important for normal speech. The character of the consonants 'm', 'n' and 'ng', which appear frequently in English, rely upon nasal resonance, for example in the words 'man' and 'hum'. Sound waves that are caught in the oro- and nasopharynx by closed lips when the soft palate is slightly raised or lowered, allow for the formation of the prolonged nasal hum common to 'm', 'n' and 'ng'. Videofluoroscopy can be used to demonstrate this palatal movement and the approximation of the soft palate with the posterior nasal wall. However, as the cause of hyponasality was obvious in our case patient and his swallow was safe this was not performed.

If the nose is blocked 'm', 'n' and 'ng' consonants often become short and clipped, more like the plosives 'b', 'd' and 'g'.<sup>2</sup> This phenomenon results in the words 'man' and 'hum' sounding like 'ban' and 'hub'. Thus a patient with rhinolalia clausa, especially one who enjoys public speaking or singing, may be much handicapped by their dysphonia.

Nasal blockage is very common. Causes include adenoidal hyperplasia in children, rhinitis, polypoidosis, deviated nasal septum and, very rarely, neoplasia.<sup>3</sup> Anatomically the bodies of C1 and C2 sit just posteriorly to the oropharynx and inferior nasopharynx. In fact the oropharynx is a popular route for odontoid peg surgery<sup>4</sup> and vertebral injury is recorded as a cause of ruptured pharyngeal mucosal membrane.<sup>5</sup> Pathology of the upper cervical vertebrae can present as a mass causing pharyngeal intrusion. The literature reports several cases of spinal osteophytes presenting as dysphonia or dysphagia<sup>6</sup> due to the compression of laryngopharyngeal structures. One Italian paper records the association of dysphonia with an osteophyte secondary to ankylosing hyperostosis.<sup>7</sup> There is not, however, a recorded link between ankylosing spondylitis and hyponasal speech.

Ankylosing spondylitis is an autoimmune condition typified by chronic inflammatory disease of the tendinous insertions of skeletal muscle into bone. This results in fibrosis and ossification of the axial skeleton known as enthesopathy. There is also associated synovitis and cartilage destruction that can lead to severe arthritis. Eventually the patient's spine may become rigid with loss of normal curvatures and movement.

The disease belongs to a family of seronegative arthropathies including Reiter's disease, psoriatic arthropathy, arthropathy of chronic bowel disease and possibly Behcet's disease. Human leukocyte antigen-B27 is common to 90 per cent of patients and there is therefore a strong familial tendency to the disease. It is most common in western European males.

- This paper describes an unusual case of dysphonia secondary to nasopharyngeal obstruction, as a result of C1/C2 vertebral subluxation/dislocation in a patient with ankylosing spondylitis
- There is an important anatomical proximity of the nasopharynx to the upper cervical spine
- Cervical pathology must be considered in patients with nasal symptoms

The abnormal fusion of originally separate bones causes atypical loading and osteoporosis.<sup>8</sup> Therefore even mild trauma may result in serious bony injuries such as spinal fractures. Thoracolumbar or cervicothoracic junction fractures are common. Higher cervical spine fractures and atlanto-axial subluxation occur, but less frequently.

There is often a delayed diagnosis of these fractures.<sup>9</sup> An unstable fracture or dislocation of the odontoid peg can result in forward displacement of the body of C1 on C2. Considering the previously discussed anatomy it is possible that the body of C1 could therefore present as an oropharyngeal mass.

### Conclusion

Although high cervical fractures are rare they do occur with increased frequency in patients with ankylosing spondylitis in comparison to the general population. This case illustrates that any patient presenting to the ENT clinic with denasal speech or oropharyngeal mass with obvious signs of ankylosing spondylitis or other cervical spinal disorder should receive lateral cervical spine radiology to exclude spinal pathology, including fracture dislocation, as a cause for their otorhinolaryngeal disorder.

### References

- 1 Mathieson L. *Greene and Mathieson's The Voice and its Disorders*, 6th edn. London: Whurr, 2001;228–36
- 2 Dworkin JP, Marunich MT, Krouse JH. Velopharyngeal Dysfunction: Speech characteristics, variable etiologies, evaluation techniques and differential treatments. *Lang Speech Hear Serv Sch* 2004;**35**:333–52
- 3 Morrison M, Rammage L. *The Management of Voice Disorders*, 1st edn. London: Chapman and Hall Medical, 1994;137
- 4 Shaha AR, Johnson R, Miller J, Milhorat T. Transoral-transpharyngeal approach to the upper cervical vertebrae. *Am J Surg* 1993;**166**:336–40
- 5 Pollock RA, Purvis JM, Apple DF, Murray HH. Esophageal and hypopharyngeal injuries in patients with cervical spine trauma. *Ann Otol Rhinol Laryngol* 1981;**90**:323–7
- 6 Kissel P, Youmans JR. Posttraumatic anterior cervical osteophyte and dysphagia: surgical report and literature review. *J Spinal Disord* 1992;**5**:104–7
- 7 Salvinelli F, Marte C, Firrisi L, D'Ascanio L, Casale M, Lamanna F *et al.* Dysphonia and cervical hyperostosis: a case report [in French]. *Rev Laryngol Otol Rhinol (Bord)* 2003;**124**:191–3
- 8 Tait TJ, Barlow G, Iveson JMI. Cervical spine fractures in ankylosing spondylitis. A case of “autofracture”. *Br J Rheumatol* 1998;**37**:467
- 9 Peretti FAE, Sane JC, Dran G, Razafindratisiva C. Particular diagnostic features of recent “spine bone” fractures in patients with ankylosing spondylitis and hyperostosis. *J Bone Joint Surg Br* 2004;**86**:27

Address for correspondence:

J M Hilton,  
48 Pightle Way,  
Lyng, Norwich,  
NR9 5RL, UK.

Fax: 44 1603 287288

E-mail: jenniferhilton@supanet.com

---

Dr J M Hilton takes responsibility for the integrity of the content of the paper.

Competing interests: None declared

---