in these patients, and may be present even in early vocal fold dysfunction appears to be highly prevalent et al., polysomnography, followed by emergent tracheostomy signs of obstructive sleep apnoea in these patients has supported recommendations that symptoms or stages of the disease and/or without symptoms of Shy-Drager syndrome date back to 1967 (Bannister et al., 1978). Interestingly, Isozaki and colleagues (1994) have recently provided some intriguing information concerning the mechanism of laryngeal obstruction in these patients. Using a novel catheter electrode array, they demonstrated both abductor paresis (posterior cricoarytenoid muscle) as well as persistence of adductor tone (thyroarytenoid muscle) during inspiration in some Shy-Drager patients with vocal fold paralysis, raising the possibility that laryngeal obstruction in this disorder may in part be dyskinetic as well as paralytic.

References

Dear Sir,
I read with interest the excellent case report entitled ‘Abductor vocal fold palsy in the Shy-Drager syndrome presenting with snoring and sleep apnoea’ by McBrien et al. (1996). The authors rightly point out the risks associated with anaesthesia in these patients, but do not mention the reports of sudden death during sleep in Shy-Drager patients with vocal fold dysfunction and obstructive sleep apnoea. Sleep apnoea from laryngeal obstruction in Shy-Drager syndrome carries a far worse prognosis than common idiopathic sleep apnoea, and deserves further discussion.

Reports of sleep-related stridor in Shy-Drager syndrome date back to 1967 (Bannister et al. 1967). Lehrman et al. (1978) noted the association between Shy-Drager syndrome, vocal fold paresis, and obstructive sleep apnoea; although tracheostomy was planned in their patient, sudden death during sleep occurred before definitive treatment of upper airway obstruction could take place. Since 1978, several additional case reports and series have supported the concept that obstructive sleep apnoea related to vocal fold abductor paralysis can be lethal in Shy-Drager syndrome (Briskin et al., 1978; Williams et al., 1979; Kavey et al., 1989; Munschauer et al., 1990). The compelling nature of these reports has supported recommendations that symptoms or signs of obstructive sleep apnoea in these patients should be immediately evaluated by overnight polysomnography, followed by emergent tracheostomy if obstruction is demonstrated (Kavey et al., 1989; Munschauer et al., 1990). In addition, diurnal vocal fold dysfunction appears to be highly prevalent in these patients, and may be present even in early stages of the disease and/or without symptoms of stridor or snoring (Williams et al., 1979). It may well be prudent to suggest that all Shy-Drager patients undergo routine laryngological examination (Williams et al., 1979), followed by overnight polysomnography if vocal fold paresis is noted. Maximal inspiratory/expiratory flow-volume loops should also be useful in screening these patients for upper airway obstruction.

It is not certain why patients with Shy-Drager syndrome and obstructive sleep apnoea, in contrast to those with idiopathic obstructive sleep apnoea, are so prone to sudden death during sleep. Autonomic cardiovascular instability from their disease may be responsible, or it may be a consequence of the laryngeal nature of the obstruction. While tracheostomy has been most frequently performed for definitive treatment of the upper airway obstruction, laryngofissure and fold lateralization has also been reported (Kenyon et al., 1984). Unfortunately, sudden death during sleep still occurs in some Shy-Drager patients even after relief of upper airway obstruction, presumably from the central apnoeas also noted to occur in this disease, or perhaps from cardiovascular instability (Briskin et al., 1978; Bannister et al., 1981).

Incidentally, Isozaki and colleagues (1994) have recently provided some intriguing information concerning the mechanism of laryngeal obstruction in these patients. Using a novel catheter electrode array, they demonstrated both abductor paresis (posterior cricoarytenoid muscle) as well as persistence of adductor tone (thyroarytenoid muscle) during inspiration in some Shy-Drager patients with vocal fold paralysis, raising the possibility that laryngeal obstruction in this disorder may in part be dyskinetic as well as paralytic.

L. K. Brown, M.D., F.A.C.P., F.C.C.P., Professor of Clinical Medicine, University of Arizona College of Medicine, 350 West Thomas Road, Phoenix, Arizona 85013, USA.

References

Abductor vocal fold palsy in the Shy-Drager syndrome presenting with snoring and sleep apnoea

(Roentgen Evidence of Normality), and his study of this work provides a comprehensive view of the current state of knowledge in the field. The results of his investigations have been published in numerous articles and reviews, as well as in his book 'The Nature and Significance of Normal Roentgen Evidence'.

The study was conducted using a novel catheter electrode array, which demonstrated both abductor paresis (posterior cricoarytenoid muscle) as well as persistence of adductor tone (thyroarytenoid muscle) during inspiration in some Shy-Drager patients with vocal fold paralysis, raising the possibility that laryngeal obstruction in this disorder may in part be dyskinetic as well as paralytic.

L. K. Brown, M.D., F.A.C.P., F.C.C.P., Professor of Clinical Medicine, University of Arizona College of Medicine, 350 West Thomas Road, Phoenix, Arizona 85013, USA.

References
