Laryngospasm in hypoparathyroidism

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The symptoms of transient hypoparathyroidism and of typical permanent hypoparathyroidism following thyroidectomy are usually well recognized, but sometimes they may be mild and are then frequently missed or misinterpreted. Unfortunately, dangerous and irreversible manifestations may develop insidiously. The case reported in this paper presented with laryngospasm eighteen years after total thyroidectomy and also showed other features of a permanent hypoparathyroid state.

Case report

A 70-year-old woman presented as an emergency with severe stridor accompanied by a 'crowing' noise, extreme distress, breathlessness and cyanosis. Each attack would last about ten minutes, occurred five to six times during the day, rarely nocturnally; there were no obvious precipitating factors. She was asymptomatic between attacks until a few days previously, since when there was always some degree of stridor. The patient was very deaf and confused so that history taking was difficult. It was elicited that she had had a thyroidectomy eighteen years previously and this had been followed by radiotherapy.

Examination

General. The hair was dry and coarse, and the nails thick and brittle, with evidence of moniliasis. There was exfoliative dermatitis, especially of the extremities.

Neck. There was a lower cervical scar consistent with a thyroidectomy. The atrophic and telangiectatic appearance of the skin was consistent with the effects of radiotherapy.

Eyes. Showed changes consistent with the removal of cataracts.

Indirect laryngoscopy. Showed normal-looking true cords fixed in the midline position. There was no spasm of the false cords or ary-epiglottic folds. The inspiratory stridor appeared to be aggravated by the hyperventilation associated with indirect laryngoscopy.

Initial management. It was considered that the patient's dangerous state demanded an immediate tracheostomy. This was carried out under local anaesthesia.

The patient's sister was contacted and she was able to supplement the history; notes from the radiotherapy department were made available. Eighteen years previously she had had a total thyroidectomy for adeno-carcinoma of the thyroid, followed two weeks later by a course of radical radiotherapy to the neck. She had been followed-up regularly in the Radiotherapy Department

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and had been on thyroid replacement therapy (60 mg. thyroid extract t.d.s.) since operation. Seven years previously she had developed her first symptoms, these being irritability and constant tiredness. Five years previously her vision, over a period of a few weeks, had deteriorated. She had the right cataract removed and two years later, the left. At this time no comment was made on her general state and no serum calcium estimation was done. Two-and-a-half years previously, other more noticeable symptoms appeared; these being stridor with breathlessness, the attacks increasing in frequency; parasthesiae and numbness of the fingers; twitching of the arms and legs at night, and always accompanying her attacks of breathlessness during the day; and increasing mental confusion. Further examination elicited immediate strongly positive Trousseau and Chvostek signs. There was generalized increase in muscle tone with brisk reflexes.



FIG. 1.
This shows a thyroidectomy scar, irradiation changes in the cervical skin and an ocular appearance consistent with cataract removal.

Investigations. Full blood count, electrolytes, urea, alkaline phosphatase, plasma proteins, serum cholesterol—all normal.

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Protein-bound Iodine—4·6 μg./100 ml. Serum calcium—4·8 mg.%. Serum phosphate—9·5 mg.%. The ECG demonstrated a prolonged QT interval.

The skull X-rays showed no evidence of intra-cranial calcification.

Diagnosis. Permanent partial hypoparathyroidism.

Further management. She was given intravenous calcium gluconate (6 doses of 20 ml. each of a 10% solution) for the first forty-eight hours, and Calciferol 150,000 units daily, accompanied by calcium supplements with milk drinks. With this régime her serum calcium eventually climbed to normal in two weeks. Her larynx recovered within a few days, with full abduction on inspiration. She was successfully decannulated. Her general health improved tremendously, her new state of well-being contrasting markedly with her previous condition. In a month her skin was again normal. She is being followed-up regularly. Her maintenance therapy consists of Thyroxine 0·2 mg. daily, Calciferol 50,000 units daily, Calcium Sandox tabs three—q.i.d. Her serum calcium is monitored weekly.



FIG. 2.
This shows exfoliative dermatitis and thick nails.

Comments and discussion

The parathyroid glands are difficult to identify at operation and to preserve them a meticulous dissection in a dry field is required. Even if they are preserved, the necessary ligation of the thyroid arteries, particularly the inferior one, may interrupt their blood supply (Wade 1960). Hypoparathyroidism must be anticipated and deliberately sought for after total thyroidectomy for cancer of the thyroid (35% of Freeman's series, 1970). Here, as with total laryngopharyngectomy with thyroidectomy for pharyngeal carcinoma, the hazard to the parathyroid glands is marked. In the latter operation, if a bilateral radical

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neck dissection with clearance of the superior mediastinum has been performed, removal of all parathyroid tissue is almost inevitable.

In acute hypoparathyroidism the onset of symptoms is usually within the first forty-eight hours, these being numbness and parasthesia around the mouth and in the hands and feet and also muscle irritability with carpo-pedal spasm. The situation is managed with intravenous injections of 10% calcium gluconate repeated as often as necessary (Fourman, 1960). The oral administration of calcium salts and vitamin D is also commenced, and continued on a maintenance level, together with a low phosphate diet. In the early stages of treatment repeated estimations of serum calcium must be made in order to stabilize the patient and to establish whether recovery of parathyroid function is occurring. Initially the dosage should be high, about 15 Gm. of calcium salt and 400,000 units of vitamin D every day, and then re-adjusted to a maintenance dose. Absence of symptoms is no guarantee of a stable state.

Complete spontaneous recovery of remaining or damaged parathyroid tissue is not the rule post-operatively after thyroidectomy, and acclimatization can occur to the low serum calcium level. The symptoms may be mild and the clinician may have a false impression of the true metabolic state. This is a dangerous situation and serious complications can develop. Our patient showed all the classical features of permanent partial hypoparathyroidism which had been latent for many years.

When she developed cataracts, the aetiology was still not suspected. Cataracts develop in 50% of untreated cases (Holtz, 1934), usually within two years if the serum calcium is below 7·5 mg.% (Lachmann, 1941). The time interval of over twelve years between thyroidectomy and the appearance of cataracts must be unusually and deceptively long. Our patient's extremely low serum calcium level—4·8 mg.%—represents an almost complete hypoparathyroidism which is in keeping with her severe and protracted symptoms over the previous one-and-a-half years.

Laryngospasm is a known complication of hypoparathyroidism. Initially we failed to realize the significance of this symptom in our patient. The low ionized serum calcium level increases the reflex excitability at the neuromuscular junctions of the recurrent laryngeal nerves. Alkalosis produced by hyperventilation enhances this effect. This was demonstrated during indirect laryngoscopy in our patient. The Jacksons (1937) described adductor spasm of the glottis, and also total sphincteric closure of the larynx, in children with laryngismus stridulus due to tetany. They noted snapping together of the vocal cords and arytenoids and, in some instances, the spasm was almost constantly present, with only a few moments interval between the prolonged spasmodic closures. Our patient's larynx showed similar features. The Jacksons also observed that increased stimulation of the larynx occurred at night due to the collection of pharyngeal secretions. This explained the occurrence of laryngospasm nocturnally. This only occasionally happened to our patient. They also described a patient who became unconscious from asphyxia, and another who required emergency tracheostomy. When death occurs in hypoparathyroidism it can be due to cachexia or broncho-pneumonia following repeated convulsions, or laryngospasm (Lachmann, 1941). A review of the literature demonstrates the rarity of the occurrence of this symptom as a presenting manifestation of hypo-

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parathyroidism in adults. It was more commonly seen in ricketty children and referred to as laryngismus stridulus in the paediatric literature, but more details about incidence are vague (Wilson, 1955).

Management of the permanent hypoparathyroid state depends on its severity. Mild deficiency states can be controlled by calcium salts alone. Treatment should always aim at administering as little vitamin D as possible (Wade, 1960). Most cases require stabilization with large doses of calcium salt and small doses of vitamin D. These patients must be kept under observation for the rest of their lives, for control of their serum calcium levels, in order to prevent over- or under-dosage.

Our patient illustrates how insidious the onset of hypoparathyroidism can be. An increased awareness of this condition is necessary so that it can be recognized before it has run its full course. A successful outcome has been achieved in our patient but at the cost of many years of ill health, the development of defective vision and the real risk of asphyxia.

Summary

A case of laryngospasm complicating long-standing hypoparathyroidism is reported. The management of this particular case is outlined. The presentation and management of transient, and in particular permanent, partial hypoparathyroidism following total thyroidectomy is briefly discussed. Chronic hypoparathyroidism can produce unusual clinical patterns which are easily missed, and laryngospasm is a rare manifestation of this condition.

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