Case series

Inflammatory bowel disease: an increased risk factor for recurrent laryngeal nerve palsy in thyroid surgery

Malattie infiammatorie intestinali: un fattore di rischio per la paralisi ricorrenziale nella chirurgia della tiroide

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SUMMARY

Transient or permanent recurrent laryngeal nerve palsy is a well known complication in thyroid surgery with reported incidences of 5-8% and 1-3%, respectively [1]. Diplegia has an incidence of 0.4% [2]. Inflammatory bowel disease (IBD) is an important cause of peripheral neurosensitivity, particularly autonomic neuropathy, which can lead to transient or permanent laryngeal nerve palsy when neural structures are involved during surgery. Several mechanisms have been implicated in the physiopathology of these neurological disorders, but the actual mechanism is still unknown. Herein we report on two patients with IBD presenting with transient bilateral recurrent laryngeal nerve palsy after total thyroidectomy without any evident mechanical or traumatic manoeuvres on apparently preserved nerves.

KEY WORDS: Thyroid surgery • Laryngeal nerve palsy • Inflammatory bowel disease

RIASSUNTO

La paralisi ricorrenziale transitoria o permanente è una possibile complicanza ben conosciuta della chirurgia tiroidea. I dati di Letteratura indicano un'incidenza variabile rispettivamente tra il 5-8% e l'1-3%[1]. La percentuale riportata di diplegia laringea è dello 0.4%[2]. Le malattie infiammatorie intestinali (IBD) rappresentano una causa importante di aumentata sensibilità nervosa periferica, in particolare del sistema nervoso autonomo, che potrebbe spiegare una paralisi transitoria o permanente di strutture nervose coinvolte durante le manovre chirurgiche. Numerosi meccanismi sono stati ipotizzati ma la reale fisiopatologia di tali disfunzioni resta al momento sconosciuta. In questo articolo sono riportati i casi clinici di due pazienti affetti da IBD che hanno presentato una paralisi ricorrenziale bilaterale transitoria dopo essere stati sottoposti a tiroidectomia totale. I nervi ricorrenti una volta identificati sono stati seguiti dalla loro emergenza mediastinica fino all'ingresso nello spazio crico-tiroideo, evitando accuratamente qualsiasi manovra traumatica e soprattutto ogni causticazione nelle immediate vicinanze delle strutture nervose.

PAROLE CHIAVE: *Chirurgia della tiroide • Paralisi ricorrenziale • Malattie infiammatorie intestinali*

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Introduction

Inflammatory bowel disease (IBD) describes a group of chronic, recurrent intestinal disorders mainly represented by Crohn's disease (CD) and ulcerative colitis (UC). Concerning the extraintestinal manifestations of IBD, peripheral neuropathy is one of the most frequently reported neurological complications [3]. Autonomic neuropathy (AN) is common in patients with IBD. In CD there is mainly sympathetic dysfunction, while in UC vagal dysfunction is more often observed [4]. Malabsorption due to folic acid or vitamin B12 deficiency [5 6] or metronidazole therapy may be causal [7], although the real mechanisms of this neuropathy remain unknown. Increased peripheral neurological sensitivity must be kept in mind during surgery when nervous structures may be involved.

Recurrent laryngeal nerve palsy (RLNP) represents the most serious complication in thyroid surgery [8], resulting in a negative impact on the quality of life. Permanent RLNP is reported to occur in 1-3% of thyroid surgeries, while temporary palsy is seen in 5-8% of cases [1]; laryngeal diplegia has been reported in 0.4% of thyroid surgeries [2].

We describe two patients with a clinical history for IBD presenting a transient bilateral recurrent laryngeal palsy after total thyroidectomy, although neural integrity was absolutely preserved during surgery and no traumatic manoeuvres were made.

Case series

During the period from January 2002 to December 2010 at the ENT Unit of University of Siena, 482 total thyroidectomies were performed by the same surgeon (GC). We report

two subjects with a history of IBD who underwent total thyroidectomy presenting with postoperative bilateral transitory RLNP, despite the integrity of the nerves which were carefully identified and preserved during surgery. Before intervention, all patients were submitted to endocrinologic evaluation at the Department of Endocrinology, University of Siena, with ultrasound and a sonographically guided fine-needle biopsy of suspected hypoechoic thyroid nodules.

Case 1

A 48-year-old man was admitted to the ENT Unit with a diagnosis of micro- and macrofollicular goitre and scheduled to undergo surgery. The clinical history showed a diagnosis of Crohn's disease treated with repeated partial ileocecal resection at the ages of 34, 40 and 42 years. The patient was also in medical therapy with azathioprine and mesalamine. The ENT examination was negative except for the thyroid region, and vocal cord motility was normal. The patient underwent total thyroidectomy. Both laryngeal nerves were carefully identified and preserved. In the immediate postoperative period, laryngeal diplegia with important dyspnea appeared, and an emergency tracheotomy was performed. Intravenous steroids (methylprednisone) and neurotrophic therapy (cyanocobalamin) was immediately started and continued for 2 weeks and 1 month, respectively, after discharge. Complete recovery of laryngeal motility was observed 10 days after surgery.

Case 2

A 73-year-old woman presented with a multinodular goitre showing right posterior tracheal deviation and mediastinal involvement. Clinical history showed a diagnosis of mild chronic gastritis, hiatal hernia and infection with H. pylori treated by eradication therapy. The patient was also submitted to subtotal colectomy for chronic UC. The ENT examination was normal except for the neck, and vocal cord motility was preserved.

The patient underwent total thyroidectomy with a conventional procedure: laryngeal nerves were identified and preserved, avoiding any traumatic manoeuvres or cauterization near the nerve. In the immediate post-operative period diplegia with dyspnoea appeared. Intravenous steroid therapy was administered and tracheostomy was performed. Complete recovery of recurrent laryngeal nerve function was achieved at 22 days after surgery.

Discussion

Disorders of the thyroid gland constitute the second most common endocrine disease following diabetes mellitus [9]. In most cases, total thyroidectomy is the surgical procedure of choice, displacing other more conservative procedures as subtotal or near-total thyroidectomy. RLNP is the most serious complication in thyroid surgery. The incidence of RLNP is variable, and percentages ranging from 0-4% have been reported in the literature [10]. An Italian multicentric study on 14,934 patients documented an incidence of 2% for transient palsy, 1% for permanent palsy and 0.4% for diplegia [2]. Permanent RLNP is reported to occur in 1-3% of all thyroid surgeries, while temporary recurrent laryngeal nerve injury is seen in 5-8% of cases [11]. Recently, in a systematic review on RLNP after thyroidectomy by Jeannon et al. [1], the mean incidence of temporary and permanent RLNP after thyroid surgery was 9.8% and 2.3%, respectively, although wide variations have been reported. As noted by the Authors, the varying rates of RLNP may be dependent upon the method of assessment. Nerve lesion may be the result of accidental sectioning, thermal insult, excessive isolation of the nerve, stretch, oedema, or haematoma [10]. In secondary or extended surgery, thyroid carcinoma, Graves' disease or inexperience of the surgeon [10 12]. Anatomical nerve variants, and particularly the relationship with the inferior thyroid artery or the presence of a non-recurrent inferior laryngeal nerve, must be carefully taken into account [8]. In our series of 482 consecutive total thyroidectomies, we observed transient or permanent unilateral nerve palsy in 2.1% and 1.0% of cases, respectively. Two patients affected by IBD showed bilateral laryngeal paralysis due to transient bilateral dysfunction of the recurrent laryngeal nerves (0.41%).

A broad spectrum of manifestations, including dermatologic (erythema nodosum, pyoderma gangrenosum), ocular (episcleritis, uveitis), skeletal (peripheral arthropathy, ankylosing spondylitis, sacroiliitis), vascular (thromboembolic disease, vasculitis, arteritis) and hepatobiliary disorders (fatty liver, chronic active hepatitis, cirrhosis, primary sclerosing cholangitis, cholelithiasis, cholangiocarcinoma) have been described to be associated with IBD [13-15]. Peripheral neuropathies are the most frequently reported extraintestinal manifestations [1]. In many cases, vitamin B12 deficiency and folic acid malabsorption [3 4] were considered, but the pathophysiology of these disorders is not yet fully known. Metronidazole, due to its ability to promote free radical formation, seems to cause damages to nerve fibres [16]. Regarding the various side effects of mesalamine, peripheral neuropathy (< 1/10,000) is a very rare occurrence.

Polyneuropathy, such as paresthesias, muscle weakness and muscle pain of lower limbs in patients have also been described in patients with IBD without any other predisposing factors, suggesting a possible autoimmune mechanism [17]. Altered response to non-invasive tests based on the heart reactions to deep breathing (E/I ratio) and to tilt (acceleration and brake indices) has been reported in patients with CD showing autonomic nervous system malfunction in these patients, which does not seem to be related to inflammation, malabsorption or treatment with immunosuppressive agents [18]. A hyperreflexia of the autonomic nervous system has been associated with inflammation in patients with systemic IBD, although it is unclear if this hyperreflexia arises from CNS dysfunction or is it a response to inflammation related to IBD [19].

To our knowledge, cases of RLNP in patients with IBD have not been described. However, the case of a 69-year-old man affected by UC who developed bilateral sensorineural hearing loss with altered ABR and bilateral facial palsy improved with steroid therapy has been reported, which was apparently related to autoimmune mechanisms [20].
In our two patients presenting bilateral RLNP after total thyroidectomy, any traumatic manoeuvre on the recurrent laryngeal nerves was carried avoided, and the anatomical integrity of both nerves was carefully preserved. The functional recovery of nerve motility was complete in both patients, although at different times.

Conclusions

Injury to the recurrent laryngeal nerve is a major complication in thyroid surgery, and its incidence is strongly linked to anatomical anomalies and the surgeon's experience. RLNP has a low incidence in experienced hands and can be avoided by proper recognition and isolation of anatomical structures.
Nonetheless, risk of injury to the recurrent laryngeal nerve cannot be completely eliminated despite perfect knowledge of anatomy and vast surgical experience if there are secondary conditions that enhance the nervous sensitivity such as IBD. In all patients, before thyroid surgery it is extremely important to obtain a thorough clinical history paying attention to all diseases which may be associated with increased nerve sensitivity. This is particularly important in patients with IBD or with risk factors for this disease even if still not diagnosed.
Careful ENT examination is necessary with flexible endoscopy to exclude pre-existing unrecognized impaired laryngeal motility. It is also very important to carry out a flexible endoscopic examination to evaluate laryngeal motility on awakening in the immediate post-operative phase, especially in patients with a history of IBD.

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