Case Report

Late Onset Spinal Cord Infection after Dilatation of the Cervical Oesophagus in Patients Treated with Total Laryngectomy and Radiotherapy

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Abstract

Objective: Total laryngectomy is often performed as salvage therapy after loco-regional radiotherapy for larynx carcinomas. A well known long-term postoperative complication is stenosis of the proximal oesophagus. A better passage can be obtained by dilatation of the stenosis by using the Savary-Gilliard polivinyl-chloride bougies.

Methods/Results: We present a series of 3 patients with spinal cord injury after dilatation for stenosis in the upper oesophagus. All patients were treated with dilatation using the Savary-Gilliard polivinyl-chloride bougies.

Conclusions: This procedure which might be performed regularly in some patients might cause unexpected morbidity.

INTRODUCTION

Total laryngectomy (TLE) after high dose loco-regional radiotherapy for larynx carcinoma has a twice as high complication rate [1] compared to laryngectomy as primary treatment.

Acute complications mainly involve occurrence of fistulas or delayed or insufficient wound healing. After salvage TLE, postoperative fistulas can occur up to 39% compared to 4% in primary TLE [2]. Long term complications are strictures or stenosis of the neopharynx or upper oesophagus causing dysphagia. Many surgical reconstruction procedures have been described to treat stenosis of the neopharynx. Besides reconstruction procedures, dilatation of the neopharynx or upper oesophagus is a valuable method to treat strictures and stenosis. Several methods for this procedure have been described with relatively few complications reported [3,4].

In our clinic the Savary-Gilliard polyvinyl-chloride bougies (SGPC) are used. This method involved passage of the bougie through the neopharynx and upper oesophagus with an increasing diameter. This can be done under local or general anaesthesia. In local anaesthesia the patient sits upright on a treatment chair and the SGPC is introduced after administration of lubricant. In general anaesthesia the patient is placed in a supine position and the SPCG is introduced orally after administration with lubricant. Most patients choose to have the procedure done under general anaesthesia. This procedure is often repeated and regarded as a safe and effective method to treat even very tight oesophageal strictures [5].

We describe three patients treated with salvage TLE after radiotherapy with subsequent stenosis of the neopharynx which was dilated using SGPC. These patients presented with severe neurological symptoms which, initially, was not related to a rupture of the neopharynx/cervical oesophagus.

CASE REPORTS

Case 1

A 79 year old male was treated with local radiotherapy in 1972 for larynx carcinoma. In 1980 he developed an advanced stage recurrence for which salvage TLE was performed. In 1999 he developed a second primary tumour of the hypopharynx, cT2N0M0, which was treated with loco-regional radiotherapy. Subsequently he developed a stenosis of the neopharynx and underwent several dilatation procedures using SGPC without subsequent complications. In 2001, four months after the latest
dilatation procedure, he was admitted to our hospital with severe pain in the neck radiating to the right arm. The neck was flexed anteriorly. Retroflexion was not possible. There was mild loss of strength in the right hand with intact sensibility. Magnetic resonance image (MRI) scanning showed a large pathological process around C5-C6 with extension to the parapharyngeal space anteriorly and posteriorly. There was no leukocytosis, the Erythrocyte Sedimentation Rate (ESR) was elevated to 71 mm/h. Metastasis of his previously treated laryngeal carcinoma was suspected though Fine Needle Aspiration Cytology (FNAC) could not confirm this. As the MRI showed destruction of the cortical bone from the caudal part of C5 and superior part of C6, spondylodiscitis was suspected. The patient was treated with long-term antibiotics and he recovered well. 3 months later a barium swallowing examination was performed showing a little fistula ending blind at the level of the stenosis. He died because of pulmonal metastasis of his previously treated laryngeal carcinoma.

Case 2

A 60 year old male was admitted to the neurology department with hemiparesis of the left side. A week prior to this he developed pain in the neck with progressive loss of strength in both arms and the left leg. In 1995 he was diagnosed with glottic carcinoma of the larynx and was treated with loco-regional radiotherapy. Two years later he developed a recurrence and a TLE was performed as a salvage procedure. Three months prior to his admittance to the Neurology department a dilatation of the neopharynx was performed because of dysphagia. Regional metastasis of his previously treated laryngeal carcinoma was suspected and a MRI was performed which showed a destruction of vertebra C3 with extension in C2 and C4 with bulging of tumour on the myelum and surrounding oedema. Because of eminent paresis acute radiotherapy on the affected area was started assuming that myelum compression was caused by metastatic disease. However, no recurrence of disease could be diagnosed and radiotherapy was stopped after several attempts with FNAC did not show malignant cells. Treatment with antibiotic was started and MRI was repeated after 4 weeks. It showed spondylodiscitis C3-C4 with development of an abscess anterior for the spinal column as well as in the muscles of the neck. An intraspinal empyema was seen in the cervical part of the spinal column with extension to the thoracic part. He developed sepsis and died subsequently.

Case 3

A 63 year old male was admitted to our hospital. In 2002 he was diagnosed with cT3N0M0 transglottic laryngeal carcinoma and treated with loco-regional radiotherapy. He developed a recurrence in 2003 and a TLE was performed as salvage procedure. Subsequently he developed a stenosis of the neopharynx and several times a dilatation procedure was performed without complications. Four months before admittance he had undergone a dilatation procedure in another hospital. After three months he had started to complain of pain in the neck. A radiographic evaluation of swallowing (EVS) revealed a perforation in the proximal part of the oesophagus with leakage of contrast to the ventral side of the spinal column. A feeding tube was placed and the patient got instructions not to eat or drink.

Three months later he developed a severe partial tetraparesis. Against advice he had continued oral feeding. EVS showed increase of the leakage (Figure 1) MRI showed an epidural abscess C2 to C4 with compression of the myelum and oedema (Figure 2). A laminectomy had to be performed and he recovered relatively well but with residual symptoms (Figure 3). Feeding was commenced through a percutaneous gastrostomy and after revalidation-period of almost a year a pharyngectomy was performed excising the ruptured area and reconstructing the defect with a free forearm flap. He died after 4 months follow-up of non cancer related causes.

DISCUSSION

In this study we presented three cases suffering from a rupture of the neopharynx/cervical oesophagus months after dilatation with SPGC. All patients presented with severe neurological symptoms which could be interpreted as loco-regional recurrence or metastasis with invasion of the cervical spine. As to our knowledge this presentation of oesophagus rupture in this case: Figure 1 Video-fluroscopy showing a defect in the neopharynx with leakage of contrast liquid pre-vertebral. Figure 2 T2 and T1 weighted MRI scan showing a pre-vertebral abscess with compression of the myelum and myelum edema.
specific category of patients months after dilatation has not been described before.

Stenosis of the neopharynx is a major complication after salvage TLE in patients earlier treated with loco-regional radiotherapy. Sweeney et al. describe in their study a percentage of 19% [6] in a large cohort of patients treated with salvage TLE. In the study of Vu et al. with a smaller cohort of patients a percentage of 50% stenosis was found [7]. All of these patients required dilatation of the neopharynx.

SPGC dilation procedures have shown to be an effective and safe method to treat stenosis of the oesophagus. Piotet et al. found a complication and mortality rate of 0.18 and 0.09% for benign and 4.58 and 0.81% for malignant aetiologies, respectively [4]. In the study of Fry et al. where 248 patients underwent a total of 365 oesophageal dilatations with 212 dilatations with SPGC, the overall incidence of perforation based on the number of procedures was 2.2% (8 perforations/365 procedures) and 3.2% (8/248 patients) based on the number of patients. The perforation rate was 1.9% for SPGC dilatation, 0% for balloon dilatation and 5-4% for pneumatic balloon dilatation in achalasia. Indications for dilatation were very variable [8].

In general, symptoms from a cervical oesophagus perforation might present less severe than perforation of the thoracic part of the oesophagus. Pain is a main symptom which might manifest spontaneous but also on swallowing. Other symptoms might be dysphagia, painful cervical contracture, retrosternal pain, dysphonia and dyspnoe [9]. Subcutaneous emphysema is commonly found on physical examination and appears radiographically in 95% of patients [10].

The overall mortality in oesophageal perforations is around 20%. A delay in diagnosis is of great prognostic value for patient outcome: a delay longer than 24 hours after occurrence doubles the mortality to 40-50% [10,11]. In a recent study from Rueth et al [12] management of oesophageal cervical perforations in patients operated with anterior cervical spine management has been described. All patients presented with a neck abscess and subsequent oesophagocutaneous fistula. Only one patient (out of a total of six) developed an extensive paraspinal abscess extending to the midthoracic spine. No neurological symptoms were described. All patients had osteomyelitis as per definition but only one had vertebral body destruction.

In our series of patients symptoms which might be expected to evolve within 24 hours actually developed after a prolonged time period of at least three months after SPGC dilatation of the neopharynx/cervical oesophagus. No invasive cervical surgery was performed. Neck pain was the common symptom though severe neurological symptoms were present without accompanying dysphagia, cervical contracture, and dyspnoe or pharyngo cutaneous fistulas.

In general SPGC is considered a safe and effective method of treatment of benign strictures of the cervical oesophagus. This series of cases illustrates that extra care should be taken in treatment of benign cervical oesophageal strictures in patients treated with radiotherapy and salvage total laryngectomy. Symptoms leading to the diagnosis oesophagus rupture might arise months after the procedure is performed and can be disguised by neurological symptoms.

REFERENCES

Kross et al. (2016)
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