

# Late laryngo-tracheal cartilage necrosis with external fistula 44 years after radiotherapy

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## Abstract

Major late complications, following radiotherapy of head and neck carcinomas, such as laryngeal oedema, perichondritis and chondronecrosis usually occur between three and 12 months after treatment. However, the present case displayed necrosis of the laryngo-tracheal cartilage and ulceration of anterior neck skin with a tracheal fistula 44 years after irradiation. The reasons for the long interval between irradiation and late complications may be explained by long-standing hypovascularity and/or infection of the irradiated area. Histological study revealed chondronecrosis without inflammatory cells in the laryngo-tracheal cartilage and bacterial colonization of subcutaneous tissue. Necrotic tissue was removed and tracheostomy was performed. The fistula was almost completely closed using a delto-pectoral cutaneous flap and the clinical course of patient has been good. This paper demonstrates the possibility of laryngo-tracheal necrosis in cases that had received radiation as long ago as 44 years.

**Key words:** Laryngeal Cartilages; Necrosis; Radiotherapy

## Introduction

Radiotherapy is one of the mainstays of treatment for head and neck carcinoma. Immediate and late complications of radiotherapy include reaction of the skin, mucous membranes, bone, and cartilage. More severe changes involve necrosis of the above tissues, resulting in fistula formation. Local and general factors such as infection, tumour invasion, and immune status can affect tissue tolerance of radiation. Although the interval between radiotherapy and development of radionecrosis varies, most reported cases have developed between three and 12 months after irradiation.<sup>1</sup>

The present case report describes an unusual case demonstrating a fistula in the anterior neck with laryngo-tracheal necrosis 44 years after radiotherapy.

## Case report

A 77-year-old female presented with skin ulceration and a fistula in the anterior neck in 1999. She had undergone total thyroidectomy and radiotherapy for thyroid tumour in 1955. The histological diagnosis of that tumour was unknown. Despite the absence of tumour recurrence, skin ulceration without fistula developed in 1977 and surgical treatment with skin grafting was performed. In November 1999, the patient again noticed skin ulceration with a concomitant fistula in the anterior neck (Figure 1) and presented to our hospital in November 1999. The ulcer measured 10 mm in diameter and the skin around the fistula was moderately inflamed. An air leak was noticed with respiration, indicating a tracheo-cutaneous fistula. Magnetic resonance imaging (MRI) revealed a tracheo-cutaneous fistula (Figure 2). The lesion was surgically exposed and debridement of necrotic tissue around the fistula was performed in December 1999. The tracheo-

cutaneous fistula was partially closed two months later using a delto-pectoral (DP) flap, as complete closure included the risk of dyspnoea due to the existence of subglottic stenosis. Histopathological study of the necrotic tissue revealed a small number of chondrocytes in cartilage implying persistent chondronecrosis. Around this cartilage, the arterial wall was replaced with the fibrous tissue, with subsequent stenosis of the lumen. Specimens also displayed infiltration by inflammatory cells with bacterial

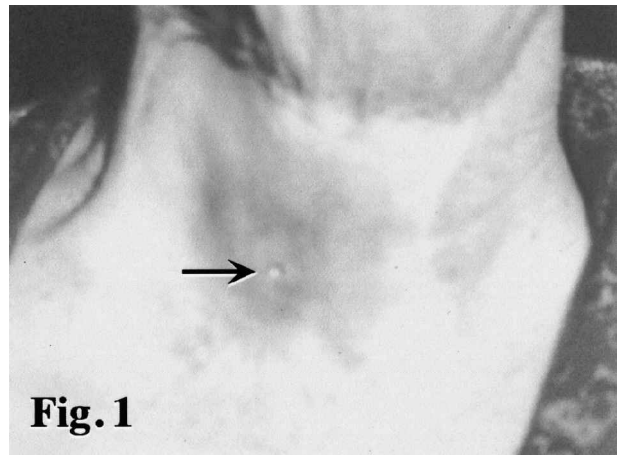


FIG. 1

Skin ulceration in the anterior neck region. Skin ulceration with fistula was recognized in the anterior neck region. The calibre of the ulcer was 5–10 mm, and the skin around the ulcer was slightly reddened. An air leak synchronized with respiration was noticed at the fistula, indicating a connection to the airway.

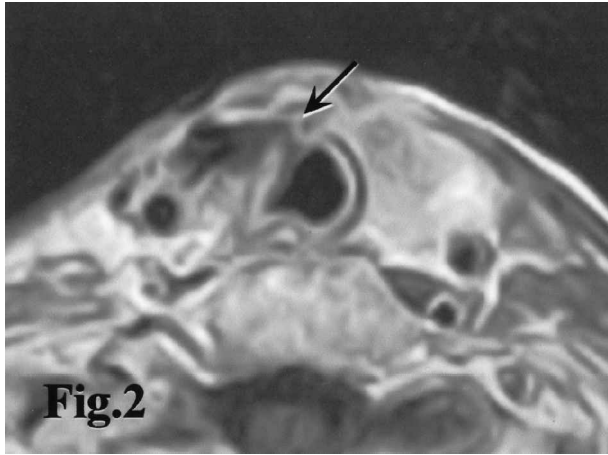


FIG. 2

Magnetic resonance imaging (MRI) of tracheo-cutaneous fistula. Partial loss of the tracheal wall links the subcutaneous air and endotracheal spaces. Skin ulceration was connected with the subcutaneous air space. These findings confirm a tracheocutaneous fistula.

colonization and the formation of inflammatory granulation in the subcutaneous tissue. No definite tumour cells were identified. The patient has experienced an unremarkable recovery in the two and a half years after treatment, with only the creation of a small tracheocutaneous fistula to assist respiration.

### Discussion

Major late complications following radiotherapy such as laryngeal oedema, perichondritis and chondronecrosis usually occur between three and 12 months after treatment.<sup>1</sup> However, the present case displayed necrosis of the laryngo-tracheal cartilage and a tracheo-cutaneous fistula with cutaneous ulceration 44 years after irradiation.

One reason for the occurrence of necrosis and a fistula such a long time after irradiation may be poor vascularity. Irradiation to the neck is known to result in obliteration of capillaries, reducing the blood supply to the larynx and trachea<sup>2</sup> and leading to chronic malnutrition of the area. These pathological changes are irreversible, lasting long after radiotherapy.<sup>2</sup> If decreases in blood flow remain more than 20 years after irradiation, necrosis and the formation of tracheal fistulae may result. In the present case, the specimen from the necrotic cartilage and surrounding tissues displayed few chondrocytes without inflammatory cells. Stenosis of capillaries caused by the fibrosis of vessel walls was also recognized around the cartilage.

In addition to malnutrition of the cartilage, infection may represent another important factor predisposing cartilage to chondronecrosis.<sup>3</sup> Chronic malnutrition may also reduce the resistance of cartilage to infection and trauma. In particular, irradiated airways such as the larynx and trachea offer increased opportunities for infection, as the function of the respiratory epithelia in removing secretions from the airways is disturbed by irradiation.<sup>2</sup> Sputum containing infectious agents therefore remains in the larynx and/or trachea and can result in infections in these areas. The bacterial colony observed in specimens from the present case demonstrated that this specimen had been exposed to a source of infection. When irradiated

cartilage with decreased resistance to infection is exposed to infectious sputum, chondronecrosis would occur, as seen in this case.

- This is a case report of delayed and recurrent necrosis with a tracheo-cutaneous fistula in a patient who had undergone radiotherapy for carcinoma of the thyroid 44 years previously
- The fistula was not associated with recurrent tumour
- The pathophysiology of chondronecrosis and treatment of such cases is discussed

In cases of laryngeal necrosis, countermeasures can be summarized as follows:<sup>4</sup> trial of antibiotics and corticosteroids; multiple deep biopsies if symptoms persist longer than six months for confirmation of tumour recurrence; and total debridement for cases in which perichondritis and cartilage necrosis continue despite aggressive medical management and multiple negative biopsies. In the present case, immediate debridement of necrotic tissues around the fistula, biopsies, and administration of antibiotics was conducted. Cleaning around the fistula was continued for two months. In addition, the tracheo-cutaneous fistula was covered with a DP flap obtained from a non-irradiated area with a sufficient blood supply. The existence of ischaemia and inflammation of the recipient site was a negative factor for failure of reconstruction using a pedicled cutaneous flap. Sufficient debridement and a pedicle flap from a non-irradiated region are therefore necessary for a positive outcome.

Radiation is widely employed against cancers of the head and neck and has proved effective in innumerable cases. However, the adverse effects can last for many years after treatment. Awareness of the possibility of chondronecrosis following irradiation is therefore important, as demonstrated in the presented case, even when radiotherapy has been performed a long time ago.

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