

## CASE NOTE

# Malakoplakia of the neck in a patient with cancer of the colon

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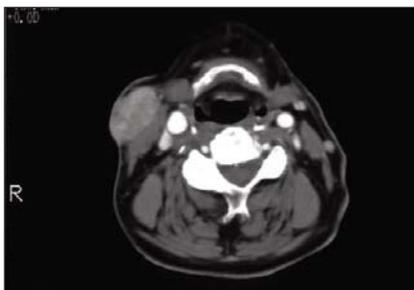
**M**alakoplakia is a rare chronic inflammatory disease that occurs most commonly in the genitourinary tract, especially the urinary bladder. Reported extragenitourinary sites include the brain, thyroid gland, middle ear, female genital tract,<sup>1</sup> pulmonary system, central nervous system, bone, lymph nodes and soft tissue. Most patients have associated conditions characterized by some degree of immunosuppression, as seen in solid-organ transplants, autoimmune diseases requiring steroid use, chemotherapy, chronic systemic diseases, alcohol abuse and poorly controlled diabetes. Malakoplakia of the soft tissue of the neck is rare; only 2 cases have been reported in the literature.<sup>2</sup> We present a case of malakoplakia of the neck in a patient with a history of intestinal adenocarcinoma. To our knowledge, this is the first report of the condition occurring outside the gastrointestinal tract and with lymph-node involvement in association with colon cancer.

Malakoplakia should be considered in the differential diagnosis of a neck mass in the appropriate clinical setting.

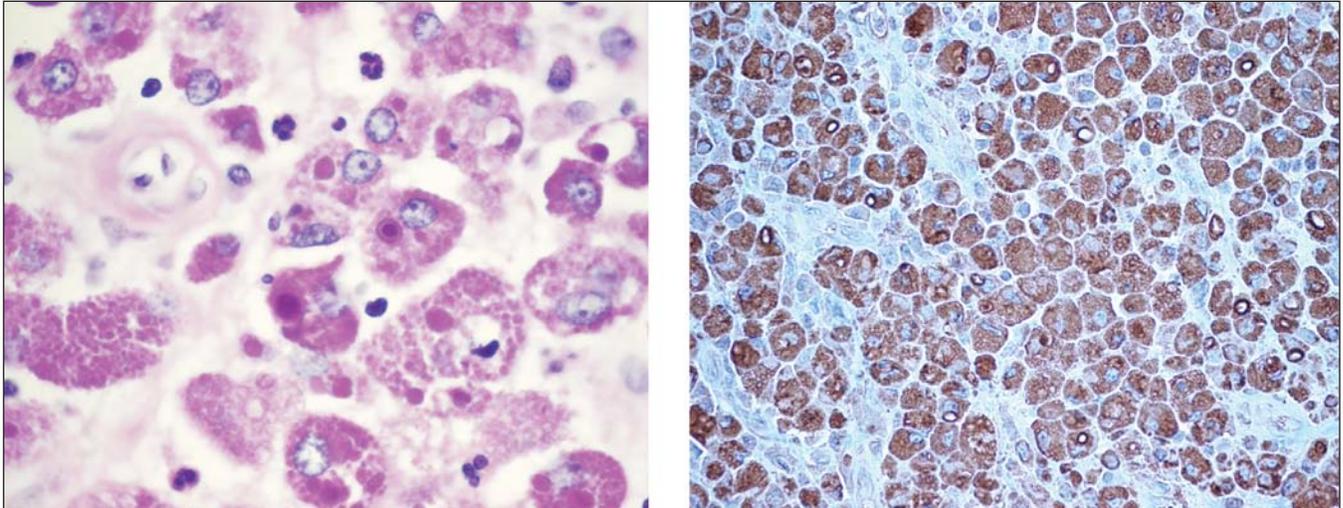
**CASE REPORT**

A 78-year-old man presented with a 2-month history of a progressively enlarging, painful, subcutaneous right neck mass that eroded the overlying skin. The mass was firm, measuring 5 × 4 × 1 cm, and was located at the right anterior triangle of the neck. It was mobile horizontally but not vertically. A computed tomography (CT) scan revealed a heterogeneous soft-tissue mass situated in the anterior triangle on the right side contiguous with both the caudal margin of the parotid gland and the anteromedial margin of the sternocleidomastoid muscle (Fig. 1). On surgical excision, the mass was noted to arise deep to the anterior border of the sternocleidomastoid muscle. It was firmly adherent to the sternocleidomastoid muscle and to the underlying jugular vein. There were also attachments to the tail of the parotid gland. There were no pressure manifestations.

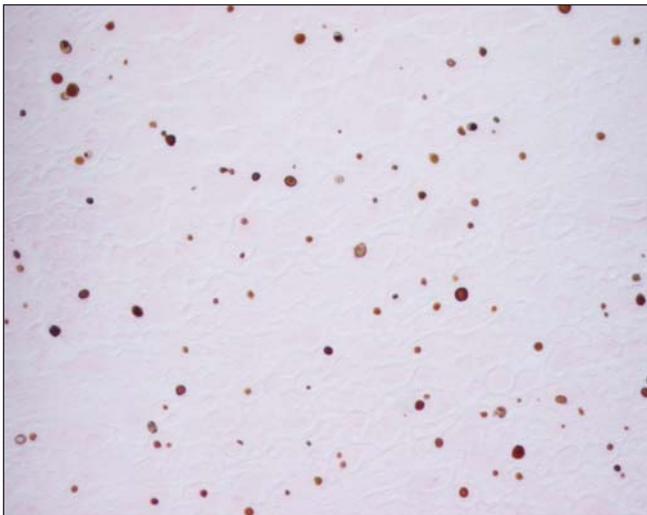
Fine-needle aspiration biopsy was not diagnostic, so the mass was excised accompanied by neck dissection. The patient had a history of large bowel carcinoma, which had been excised 6 years earlier, and chronic obstructive pulmonary disease (COPD). He had smoked cigarettes for 60 years. On microscopic examination of the excised specimen (Fig. 2), sections showed a diffuse infiltrate of histiocytes, most of which contained cytoplasmic inclusions, which were periodic acid-Schiff positive, diastase resistant and von Kossa positive (constant with von Hansemann cells). Diagnostic Michaelis-Gutman bodies (Fig. 3) were also identified. Eighteen of 27 lymph nodes examined showed involvement by the same process. The patient received antibiotics for 6 months. A year after, he died of complications of COPD.



**Fig. 1.** Computed tomography scan showing a heterogeneous soft-tissue mass situated in the right anterior triangle of the neck.



**Fig. 2.** Left: Specimen stained with period acid-Schiff (PAS) after diastase pretreatment, showing PAS diastase-resistant intracytoplasmic inclusions (original magnification  $\times 400$ ). Right: Histiocytes are CD68-positive (PAS stain, original magnification  $\times 100$ ).



**Fig. 3.** Specimen shows calcium-ion deposition (Michaelis-Gutman bodies) in malakoplakia (von Kossa stain, original magnification  $\times 100$ ).

## DISCUSSION

Malakoplakia has now been described in both sexes with a slight female predilection. Most cases occur in patients with a depressed immune system. The etiology of malakoplakia has not been fully elucidated. It is thought to result from inadequate intracellular killing of phagocytosed bacteria by monocytes and macrophages, leading to the accumulation of calcium and iron on bacterial glycolipid within cellular phagolysosomes. It is postulated that the inadequate killing of bacteria is related to low levels of intracellular cyclic guanine monoamine phosphates that result in poor microtubular function and lysosomal killing activity.<sup>3</sup> Gram-negative bacteria are most commonly seen in malakoplakia with *Escherichia coli* found in more than two-thirds of the lesions.

Malakoplakia of the neck is extremely rare. The association between malakoplakia and colon cancer is documented in the literature. In all previously reported cases, however, it was an incidental finding confined to the area adjacent to the carcinoma.<sup>4</sup> Local alteration of the gut flora was postulated as a mechanism for the development of malakoplakia in these cases. Our case raises the possibility that the mechanism involved is more than just a local phenomenon. Regional lymph-node involvement by malakoplakia has been reported.<sup>5</sup> The possibility still exists that in our case the malakoplakia and carcinoma was just a coincidence, although it is more likely that both lesions are related, since malakoplakia is known to occur in immunosuppressed patients.

Our case demonstrates the protean nature of the presentation of malakoplakia, where it can mimic malignant disease, especially when the lesion is ulcerated and is accompanied by lymph-node involvement. Histologic examination is necessary to establish the correct diagnosis before a more radical excision is attempted.

**Competing interests:** None declared.

## References

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