Pilomatricoma is the second most common benign dermal-subcutaneous tumor of the head and neck region, after epidermoid cyst, originating from the outer sheath cells of the hair follicle. It can be easily treated with surgical excision. However, it is a relatively unknown skin lesion to maxillofacial surgeons. Therefore, we report a case of pilomatricoma occurring inferolateral area of the parotid gland in a 54-year-old man; with a review of the relevant literature.

Key words: Pilomatricoma, parotid gland

I. INTRODUCTION

Pilomatricoma is a skin lesion that originates from the outer root sheath cells of the hair follicle. It is the second most frequent lesion of the head and neck region, after epidermoid cyst, and it occurs with a prevalence of approximately 1% among all benign skin lesions. Pilomatricoma may occur at different ages, but most commonly occurs before 20 years of age. Clinically it is found in the form of an indolent mass (≤3 cm in diameter) in the epidermis, and is characterized by its firmness and gradual proliferation. Pilomatricoma can be preoperatively diagnosed using Doppler sonography, computed tomography, or fine-needle aspiration (FNA). It is usually treated by surgical excision including the border of lesion, and its recurrence rate is low.

Below, we present a case report of pilomatricoma incidentally discovered in the inferolateral area of the parotid gland; the following discussion includes a review of the relevant literature.

II. CASE REPORT

A 54-year-old male patient visited the emergency room of Kyungpook National University Hospital because part of
a broken blade was embedded in the right facial area during gardening. The patient had no specific medical history, and complained of pain in the right cheek owing to the embedded broken blade. Panorama and computed tomography were performed to ascertain the precise location of the foreign body in the right cheek, and whether the adjacent tissue was infiltrated and damaged by the blade. Computed tomography (CT) revealed a foreign body located at the height of the occlusal plane in the interior region of the right cheek (Fig. 1A, B); CT simultaneously showed a radiopaque, elliptical lesion (2.3 cm × 1.0 × 0.8 cm) with a well-defined margin separated from surrounding tissues in the inferolateral area of the parotid gland (Fig. 1C, D). The patient had not been aware of this lesion. It was diagnosed clinically as a calcified lymph node and prepared for surgery.

Under general anesthesia, the broken blade was removed through an intraoral incision, and the lesion in the inferolateral area of the parotid gland was approached extraorally and removed as well (Fig. 2A, B). The biopsy result of this lesion showed marked ghost cells with ectopic bone formation around them and some multinucleated giant cells. The lesion was then diagnosed as a pilomatricoma (Fig. 3A, B). There were no recurrence or further pathologic
findings during the follow-up period, and the surgical site was repaired favorably.

III. DISCUSSION

Pilomatricoma is a primarily benign lesion that occurs in the epidermis. In most cases, pilomatricoma is found on the head and neck, back, or upper limb, and rarely on the lower limb. It can occur at all ages, but tends to be prevalent in younger individuals and is rare in middle-aged and older individuals. The pathogenic mechanism is known to be associated with mutation of the CTNNB1 gene, which encodes beta-catenin, but the precise cause of this disease remains to be determined. Clinically it appears as a hard, painless, round or elliptical lesion of lesser than 3 cm in diameter on the surface layer of the skin.

For pilomatricoma of the head and neck region, clinically it should be differentially diagnosed from epidermoid or dermoid cyst, parotid tumor, calcified lymph node, and hemangioma. Especially when pilomatricoma occurs around the parotid gland, as in this case, differential diagnosis from parotid gland tumor and calcified lymph node is necessary. Additional tools used in differential diagnosis include noninvasive methods such as Doppler sonography, radiography, and computed tomography, beside invasive methods such as FNA and biopsy. However, if the cell type constituting the pilomatricoma is predominant or if ghost cells or multinucleated giant cells are deficient, pilomatricoma may sometimes be misdiagnosed as a malignant tumor by the FNA test. In particular, the presence of high cellularity and a high nuclear/cytoplasmic ratio is liable to lead to a misdiagnosis of squamous cell carcinoma. Therefore, diagnosis should be performed very carefully.

Maria et al. reported the histologic features of pilomatricoma as the presence of basaloid cells, ghost cells, and multinucleated giant cells. Intra- and extracellular matrix calcification is present in 69–85% of cases. The older the lesion, the more likely is the observation of histologic features such as ghost cells, multinucleated giant cells, and calcification. In some cases, however, the cell matrix contains chronic inflammatory cells infiltrated with foreign body giant cells, and an enhanced level of intracellular mitosis is evident; these are the microscopic characteristics of malignant pilomatricoma.

If a benign pilomatricoma is completely removed by surgical excision, recurrence is rare. Where a recurrence rate of 2–6% has been reported, in most cases the lesion had not been removed in its entirety. Recently, however, there are reports of malignant transformation. Unlike a benign tumor, when pilomatricoma develops into a malignant lesion it tends to be locally very aggressive and recur tendency is very high. This malignant transformation is extremely rare, but is more frequent in men. The prognosis in such cases is very poor.

Although pilomatricoma occurs mainly in the maxillofacial region, it is a skin lesion relatively unknown to maxillofacial surgeons and is thus difficult to differentiate from other diseases. This case report, involving a trauma patient, was found incidentally and the lesion was ultimately diagnosed as a pilomatricoma by biopsy following surgical excision. Because pilomatricoma is relatively simple to treat by surgical excision and rarely recurs, it should be accurately diagnosed by FNA or biopsy so that appropriate treatment can be provided.

REFERENCES


