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Case Report

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Medico-Legal Pitfall in Adenoid Cystic Carcinoma of The External Auditory Canal

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Abstract

A roadway is a structure designed for the movement of individuals and vehicles, functioning as a public route, a street within a community, or an unclassified passage where traffic occurs. In its most comprehensive definition, a roadway may be regarded as any surface that facilitates travel or movement and links two locations. A roadway constitutes a designated area of land that is either prepared for or utilized by some form of traffic that connects two endpoints.

Keywords: Adenoideocystic Carcinoma, Tumors of The External Auditory Meatus, Medical Liability

1. Introduction

Among the head and neck cancers, the adenoideocystic carcinoma (AC) recurs as a rare variant that usually affects the parotids and its primary origin from the ceruminous glands of the ear auditory canal (EAC) is considered extremely rare. In fact, the full medical literature reports only very few anecdotal cases or reviews with no controlled studies on clinical management as well as therapeutic responses and survival .The EAC cancers generally occur with protean clinical presentation and symptoms while their morphologies overlap between benign and malignant tumours and particularly AC, represents 0.0001% of all neoplasms of the cervico-facial district although it is the most frequent among tumours originating from the ceruminous glands [1-3].

Due to the extreme rarity of AC of the ceruminous glands (ACCG), even in the latest AIOM (Italian Association of Oncology) 2019 guidelines there are no survival data and specific indications for treatment. For this reason, the approach to diagnosis, therapy and follow-up must necessarily be related to the general lines of head and neck tumours for which integrated therapeutic protocols are available and primarily include surgical excision followed by chemo-radiotherapy as well as immunotherapy in relation to the clinical stage of the disease.

Several progress in staging has recently grown in relation to the presence of a few viruses including HPV in particular in squamous neoplasms of the oral cavity and to the expression by the cancer cells of the epithelial growth factor receptor (EGFR), namely a cancer proliferation inducer molecule.

The surgical radicalization followed by radiotherapy is the gold standard treatment for these tumours up to their T2-N0 stage, whereas in advanced stages chemotherapy including platinum derivatives combined with anti-EGFR monoclonal antibodies and radiation treatment is functional in inducing the tumour remission. According to international studies, the combined chemo-radio

treatment of patients within the III-IV, M0 stage after the surgical resection leads to a 5-year survival in approximately 58% of patients [4-7].

ACCG is considered a slow-growing carcinoma and leads to gradually occupy the EAC resulting in hearing loss as first symptom though other common disorders including ear pain, otorrhea, tinnitus and dizziness can also occur. In a review of the ACCG from literature 1, it is stated that, if removed at stage I-II N0 M0 and properly treated with chemo-radio, the survival is variable and depends on various factors. According to Craig et all 1, the survival over 10 years occurred in more than 50% of patients considered in the study which, however, included a cohort of 17 cases.

However, although slow in its growing, ACCG shows marked infiltrative and metastatic properties whose clinical effects occur even several years after the tumour ablation. The organs frequently affected are beyond the structures adjacent to the excision site and may include the central nervous system, lungs, liver, kidneys and skeleton for which the positive surgical margins for tumour on the operative report, bone infiltration and invasion perineural are considered to be unfavourable prognostic criteria [8,9].

From the medico-legal point of view, this neoplasm presents some criticalities represented by the difficulty of making a diagnosis due to the rarity of the tumour thus rendering quite difficult the histotype classification and for the fact that the patient often underestimates the symptoms affecting the ear thus delaying the visit to the otrorinolaryngologist specialist. This case report aims to clarify what potential cases of malpractice can be related to this type of cancer.

2. Case Report

This is the case of Mrs. A.D. who in April 2010 was admitted to a hospital in southern Italy for left hearing loss since about 4 years. A ceruminous plug was extracted and both antibiotics and cortisone were administered. The patient was discharged with a diagnosis of perichondritis of the left auricle. In September 2010, the patient underwent a biopsy of the lesion whose histological examination indicated demonstrated the presence of fragments of basal cell carcinoma with cutaneous aspects features of Morphea associated whose and with involvement of the resection margins were site of neoplasm. MRI performed to stage the neoplasm described the pathological thickening of the skin, subcutaneous and mucous layers of the external auditory canal (EAC) which was impregnated with contrast medium without other signs of involvement of nearby and intracranial structures as well as organs.

In January 2011, a histopathological revision of the biopsy was made performed and she underwent surgery after Thea diagnosis of benign eccrine EAC neoplasm was made. For which the patient underwent surgery of the lesion which occupied the anterior pretragal and perianular area with initial erosion of the bone duct on the back. During surgery, an extemporaneous histological examination was performed and described the diagnosis and of AC was clarified achieve clarified . Due to the complexity of the surgery caused by the extension of the tumour, the patient was advised to necessity of the petrosectomy to be performed at a Swiss otolaryngology centre where the histological evaluation of the removed lesion confirmed the extemporaneous intraoperative diagnosis of AC of the, with trabecular growth characteristics, onset from the ceruminous glands with EAC arising from ceruminous glands, with tubulo-trabecular and solid patterns of growth, perineural and infiltration, and with involvement of susurrounding tissue infiltration and, involvement of the surgical resection margins. and perineural infiltration.

In February 2011 the patient underwent In the Swiss hospital, to resection of the left auricle, left subtotal petrosectomy, left parotidectomy, and resection of the left mandibular joint capsule. The histological examination of the surgical specimen confirmed the histological diagnosis of AC in fragments residual from the previous surgery, and revealed tumour infiltration only in the soft tissues without further tumour presence in the other resected structures. Demonstrated the presence of residual carcinoma infiltrating skin and soft tissues.

In April 2011 she began radiotherapy treatment for adjuvant purposes.

In July 2012, the total body PET examination highlighted "... Multiple millimetre bilateral pulmonary nodular formations not metabolically characterized due to their small size ...".

In November 2012, following a follow-up CT scan, the patient was diagnosed with volumetric enlargement of some pulmonary nodular lesions in the right lower lobe which were interpreted as metastatic repeats. The patient was then submitted to first line chemotherapy (Cisplatin + 5FU + Erbitux) until May 2013 and then to second line protocol from February to April 2014 to counteract the progression of lung metastases whereas In November 2019 she underwent hypo fractionated radiotherapy on lung metastatic lesions refractory to chemotherapies. The procedure involved only the most voluminous lesions at risk of bronchial compression.

By September 2020, the patient underwent further radiotherapy regimens to slow the progression of lung lesions and several lesions were reducing as effect of radiation therapy applications, whereas other nodular lesions showed a tendency to increase in their size. Besides the lung lesions, a metastatic nodule was detected on the VI segment of the liver that was ablated by stereotaxis radio-treatment in March 2021.

3. Discussion

The patient presented a definitely aggressive variant of AC as effect of both perineural and bone invasion described in the surgical specimen following the first excisional surgery in January 2011. Furthermore, in the natural evolution of the neoplasm, despite the

chemo-radiotherapy treatment, the metastatic disease progressed with high aggressiveness in relation to the discovery of the voluminous pulmonary nodules detected by PET in July 2012 as well as the liver metastasis in the examination TC of March 2021. Therefore, based on the time longer than 10 years since the original symptoms and accordingly to the natural history of the tumour, the unfavourable prognostic factors including perineural, bone and soft tissue invasion, were predictors of the metastatic progression that occurred in the following years.

In our case the diagnostic delay (2008-2011) may have influenced the evolution of the tumor. In fact, due to the slow progression of ACCG neoplasm, if in 2008 an MRI examination of the left EAC would have been performed, the tumor diagnosis would have been easier and more prompt. It should be emphasized that the symptoms presented by the patient, since 2006, characterized by hearing loss and otalgia, should have required in addition to the attending physician also the otolaryngologist for a necessary diagnostic investigation. Furthermore, according with Prasad et all patients with long standing otalgia, decreased hearing and ear canal mass should be early considered for deep incisional biopsy along with imaging since early prompt surgery combined with adjuvant radiotherapy may help to prevent distant metastasis. It must be considered, according with Markou that the rarity of the disease and the relative inexperience combined with the nonspecific symptomatology and the often-vague imaging results render the diagnosis especially difficult. It must be reported that the lack of specific clinical and radiological signs makes the diagnosis of ceruminous gland tumors challenging. In his study CT scans were performed in a few cases, but no tumor suspicion was addressed by the radiologist while, moreover, the difficulty of achieving an accurate pathological diagnosis when inadequate specimens are provided has been repeatedly emphasized [10-13].

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