

Differential diagnosis of a carcinoma of the maxillary sinus that resembles a sparganum infection: A case report

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Abstract. Primary malignant tumors of the maxillary sinuses are rare. The present study reports the case of a maxillary sinus adenocarcinoma that was misdiagnosed as a frog sparganum infection, and discusses the differential diagnosis between the two diseases. The patient was ultimately diagnosed with a carcinoma of the left maxillary sinus that presented as a progressive mass in the left eye and the maxillary sinus. Eosinophilic infiltration was observed in the subcutaneous tissue, and the patient had experienced previous exposure to undercooked frog. Although an anti-sparganum ELISA was performed and the results were negative, a sparganosis infection was initially diagnosed. However, following the application of anti-sparganosis treatment, no improvements were observed. Histological examination of an orbital mass resection revealed an adenocarcinoma with bone metastases. To the best of our knowledge, the present study is the first to report a maxillary sinus carcinoma misdiagnosed as sparganosis. Therefore, the findings of the current study should be considered in the differential diagnosis between a carcinoma of the maxillary sinus and sparganosis. Avoidance of misdiagnosis at an early stage is crucial for effective diagnosis and treatment of sinonasal malignancies.

Introduction

Adenocarcinomas comprise 10-20% of all primary malignant neoplasms of the nasal cavity and paranasal sinuses (1). Occupational exposure to dust from hard wood or leather has been considered as one of the major causes of adenocarcinomas since the 1960s (2,3). Therefore, adenocarcinomas

of the nasal cavity and paranasal sinuses are generally considered to be occupational diseases, although spontaneous cases do occur (4,5). The majority of adenocarcinoma cases originating from the nasal cavity or sinuses (85%) are located in the ethmoid sinus and upper part of the nasal cavity (6). Approximately 10% of cases arise in the maxillary sinus, and these are not usually associated with wood dust exposure (7). The median age of onset is between 50 and 60 years, and a higher incidence of cases are observed in males subjects than in females (8). The present study reports the case of a maxillary sinus adenocarcinoma that was misdiagnosed as a frog sparganum infection, and discusses the differential diagnosis between the two diseases.

Case report

A 52-year-old male patient was admitted to a local hospital 20 months previously with complaints of left eye swelling and epiphora, without light fear or pain. The swelling gradually extended to the face and neck, without fever or pain, and remained for ~1 year. Written informed patient consent was obtained from the patient's family. The patient was initially treated with several courses of dacryocyst irrigation, anti-inflammatory treatment and traditional Chinese medicine for presumed dacryocystitis in two local hospitals. With the deterioration of symptoms, the onset of left-side eyelid swelling and the difficulty in opening the eyes, two endoscopy biopsies were performed, which revealed eosinophil infiltration without any other abnormalities. The patient was referred to the Collaborative Innovation Center for Diagnosis and Treatment of Infectious Diseases at the First Affiliated Hospital of Zhejiang University (Hangzhou, China), 20 months subsequent to the first presentation at the local hospital, due to severe left facial swelling. The general health of the patient was satisfactory and a gross physical examination revealed no evident abnormalities. The patient had a history of consuming undercooked frogs and snakes. The patient exhibited diffuse soft-tissue swelling with a firm surface on the left side of his face. The swelling extended to the left eye, which was associated with difficulty in eyelid opening and purulent secretions on the edge (Fig. 1A). Computed tomography (CT) and magnetic resonance imaging (MRI) examinations of the head revealed multiple lesions disseminating in the left

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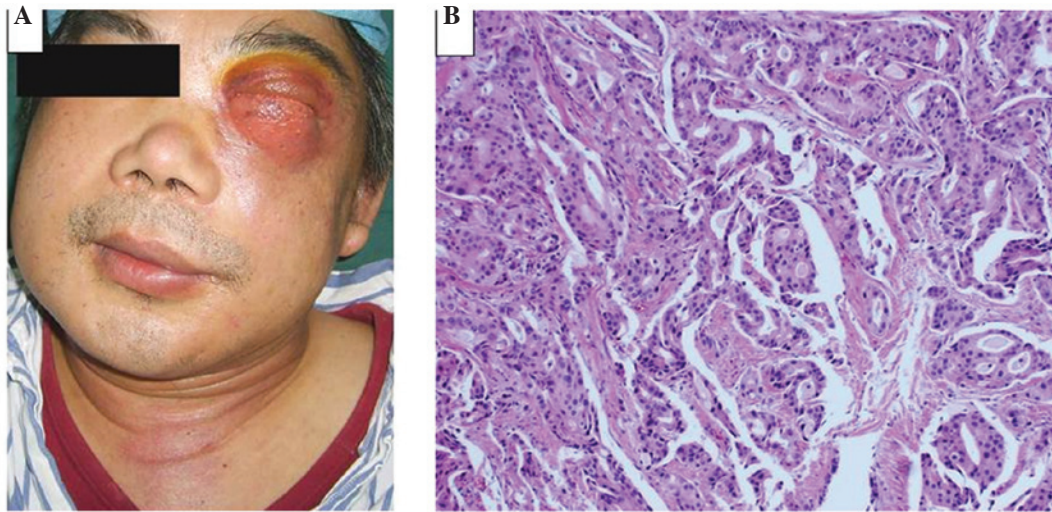


Figure 1. (A) Photograph showing the initial presentation on admission to the clinic, demonstrating the diffuse soft-tissue swelling on the left side of the face and neck extending to the left eye. (B) Photomicrograph showing the pathological examination of the adenocarcinoma. Atypical spindle cells with an invasive growth pattern, and tubular and trabecular elements, were observed in the stroma below the left canthus (hematoxylin and eosin staining; magnification, x200).

orbit, maxillary sinus and ethmoid sinus, as well as diffuse soft-tissue swelling of the left side of the face and neck. An ELISA for *Sparganum mansoni* was performed and the results were negative, using a *Sparganum mansoni* antibody detection kit (Shenzhen Combined Biotech, Co., Ltd.). However, a sparganosis infection was suspected based on the patient's history of consuming frogs and snakes and the detection of eosinophil infiltration in the biopsy. However, anti-sparganosis treatment with praziquantel and anti-inflammatory treatment, consisting of 0.5 g azithromycin, 0.4 g amikacin and 0.5 g levogloxacin for 1 week, produced no discernible improvement. The patient underwent a left orbital mass resection and biopsy, during which white, fish-like tissue and bone destruction was identified in the deep left inner canthus. Pathological examination of the biopsy specimens collected from the mass and bone revealed the presence of an adenocarcinoma (Fig. 1B) with bone metastases. Distant bone metastases were also observed in an emission CT scan of the skeleton. Therefore, a final diagnosis of a carcinoma in the left maxillary sinus with bone metastasis was established. The patient was treated with chemotherapy and palliative care. The chemotherapy regimen consisted of 75 mg/m² cisplatin on day 1 and 1,250 mg/m² on days 1 and 8, every 21 days, which was repeated four times. However, the patient succumbed to multiple organ failure six months following treatment.

Discussion

Primary malignant tumors of the maxillary sinuses are rarely observed in clinical practice. The presence of large air spaces within the sinus allows the asymptomatic expansion of sinus tumors. At the early stages, these tumors are often asymptomatic or mimicking inflammatory diseases, leading to a delay in diagnosis or misdiagnosis (9). Thus, a number of patients present with advanced-stage disease and local invasion to the surrounding tissues, including the orbit, dura, brain, pterygo-maxillary fossa and the cribriform plate (10). In the present study, the patient first complained of left eye swelling, which

was initially misdiagnosed as dacryocystitis and a sparganosis infection.

Sparganosis is a rare infectious disease that is caused by the plerocercoid larva of tapeworms belonging to the *Spirometra* species. The disease is most prevalent in Eastern Asia. Humans may acquire the infection by eating undercooked meat, particularly meat from snakes or frogs (11). The early stages of the disease are often asymptomatic; however, the sparganum typically causes a painful inflammatory reaction in the tissues surrounding the subcutaneous site with growth (12). The clinical manifestations are diverse, and the majority of infections present with masses in the soft tissues and sinus tract, including the sinonasal passage and eyes (13). A sparganum is typically accompanied by granulomatous inflammation and eosinophilic infiltration in the subcutaneous tissues, which is often identified with microscopic examination of the excised tissue. In addition, an anti-sparganum ELISA test can be indicative (14). In general, a preoperative diagnosis should be established based on the identification of exposure history and a painful, migratory, subcutaneous nodule. However, the final diagnosis should be confirmed by surgical removal of the worms or identification of the parasite in a tissue specimen.

In the case of the present study, the patient lived in a sparganum endemic area, where five cases of a sparganum infection were reported between 2000 and 2010 (15). The current patient presented with a progressive mass in the sinonasal passage, had an exposure history to undercooked frog, and eosinophilic infiltration was observed in the subcutaneous tissue. Although the anti-sparganum ELISA test was negative, a diagnosis of a sparganosis infection was initially proposed. However, treatment with anti-sparganosis drugs resulted in no improvements. To confirm a definite diagnosis, a resection of the left orbital mass was performed and histological examination was conducted. The results confirmed the diagnosis of an adenocarcinoma with bone metastases. Notably, a sparganosis parasite was not identified during the surgery or the pathological analysis of the specimens.

To the best of our knowledge, the present study was the first to report the case of a maxillary sinus carcinoma misdiagnosed as sparganosis. In the present case, two endoscopic biopsies failed to provide any evidence for malignancy, and the patient received several treatments for dacryocystitis and sparganosis infection. The duration between the initial visit to the local hospital and the definite diagnosis was almost two years, during which period multiple invasive examinations and treatments that caused physical injuries and an economical burden to the patient were performed. Previous studies have reported that the diagnosis of sinus malignancies is difficult due to the air-filled nature and deep position of the structures involved (9,16). The outcomes of the present case strongly indicated that the avoidance of misdiagnosis at an early stage is crucial for the effective treatment of this type of cancer. The following clinical manifestations are strong indications for a carcinoma of the maxillary sinus: Patients aged >40 years with a slowly progressing unilateral nasal obstruction associated with epistaxis, pain below the eye, hyposmia, facial numbness, facial swelling, skin sensory loss or difficulty in opening the mouth. Since the differentiation between a carcinoma and sparganosis is based on histological examinations, an adequate tissue biopsy is a necessary requirement. Maxillary sinus exploratory surgery should be considered to eliminate the possibility of the disease. In addition, CT scanning is helpful to delineate the extent of the cancer, although differentiating between tumors and edematous mucosa can be difficult with CT scanning. However, MRI may be applied to aid the differential diagnosis.

In conclusion, the consideration of a carcinoma is important since carcinomas and sparganosis in the maxillary sinus are extremely rare diseases. Due to the anatomical nature of the maxillary sinus, misdiagnosis and delayed diagnosis are common. Therefore, a careful biopsy and pathological examination are required for early diagnosis and treatment.

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