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Acquired Torticollis as the Initial and Only Finding in Nasopharyngeal Carcinoma: A Case Report

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ABSTRACT

Torticollis or involuntarily twisted neck is considered to be a sign rather than a condition. Some of the main causes of torticollis are trauma, medication side-effects, infectious and inflammatory processes, and head and neck tumors.
A 26-year-old female patient presented with acute acquired torticollis for four months, and the conditions had complicated due to constitutional symptoms, such as weight loss, sweating, and decreased appetite, eventually leading to trismus. Neck CT-scan showed bilateral lymph node enlargements, soft tissue stranding, right-sided asymmetry of the fossa of Rosenmüller (pharyngeal recess), and a heterogeneous enhancing mass on the nasopharynx roof with left extension and bilateral pressure on the Eustachian tube. The biopsy of the mass indicated the infiltration of atypical epithelial cells with marked nuclear atypia in small solid nests within the lymphoid tissue of the nasopharynx, which corresponded to nasopharyngeal carcinoma. However, the patient had no risk factors for nasopharyngeal carcinoma. This study highlighted the importance of a complete work-up for the underlying tumors in the head and neck in the patients presenting with the only finding of torticollis.

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Introduction

Torticollis or involuntarily twisted neck is considered to be a sign rather than a condition, which has been reported in various conditions (1,2). Torticollis is the result of some conditions that cause the spasm of the cervical muscles, particularly sternocleidomastoid and trapezius muscle shortening, as well as the irritation of the cervical nerves, all of which lead to the abnormal position of the neck and one-sided turning of the head (3,4).

Torticollis is relatively common in childhood and could be acquired or congenital, with the latter being more prevalent. This clinical finding has also been reported in adults. Several benign and aggressive conditions have been implicated in the pathogenesis of acquired torticollis, including trauma (blunt trauma or malposition of the neck), infections (e.g., retropharyngeal abscess) (5), neurogenic conditions (e.g., myasthenia gravis) (6), ocular conditions, and tumors.

Among the tumors that may cause torticollis, even as the first and sole presentation of the underlying malignancy, posterior fossa tumors and spinal cord tumors have been highlighted in the

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literature (7-9). In a study regarding the prevalence of torticollis in children, 22% of pediatric patients were reported to have central nervous system (CNS) tumors as the first presentation before the emergence of other neurological signs and symptoms (10). Moreover, the CNS tumors affecting the other adjacent anatomic locations could also cause torticollis albeit very rarely. For instance, nasopharyngeal carcinoma mimicking as peritonsillar abscess has been reported to cause torticollis, as well as dyspnea and fever (11).

There have been few reports on the unusual tumors that could cause torticollis, while researchers have realized that the awareness of these rare tumors could contribute to the management of patients with torticollis effectively.

The present study aimed to describe the case of a young adult patient with torticollis and discuss the diagnostic difficulties.

Case report

A 26-year-old woman with a four-month history of neck pain, neck muscle stiffness, and right-sided twisted neck was referred. The condition had started one week after a tooth extraction procedure. The patient reported no trauma in the head and neck trauma or other anatomic areas beforehand. In addition, she had no recent history of other medical conditions, especially oropharyngeal infections, and used no medications.

Upon the initiation of the symptoms, the patient was visited by an otolaryngologist, and neck CT-scan (without contrast) and magnetic resonance imaging (MRI; without gadolinium enhancement) were requested, indicating no abnormalities. Afterwards, the patient was referred to a neurologist to receive botulinum injection for the treatment of cervical dystonia. However, due to nausea, frequent postprandial vomiting, significant weight loss (12 kg), and notable dysphagia to solid foods, the mentioned process could not be performed. Furthermore, the increased serum level of erythrocyte sedimentation rate (ESR; 72 mm/h) was observed on the laboratory tests. Following that, she was referred to a gastroenterologist. Esophagogastroduodenoscopy was carried out, which was unremarkable. Abdomino-pelvic ultrasound was also performed in another center, which was normal. During this period, more constitutional symptoms emerged, including weight loss (12 kg), sweating, anorexia, and fever (without chills). During the last month, the patient also complained of gradually-onset, decreased hearing (more severe on the left side), fatigue, and cough, while she reported no dyspnea.

In order to investigate the neck pain, muscle spasm, and increased ESR levels and due to the suspicion of ankylosing spondylitis, the patient was referred to the rheumatology clinic. On the physical examination, the patient was alert and cachectic. Her vital signs were also within the normal limits, with the exception of the oral temperature of 38.2ºC. She had fixed, right-sided torticollis and was unable to extend her neck. In addition, her neck movements were painful in all the directions. Upon palpation, the neck showed tenderness. Sternocleidomastoid muscle stiffness and spasm were observed, while the patient was negative in terms of Kernig's and Brudzinski's signs. Enlarged lymph nodes were also palpated in the submandibular region, which were prominent on the right side. Lymphadenopathy was also palpated at the anterior portion of the sternocleidomastoid muscle on the right side of the neck. Furthermore, the patient had tenderness on the percussion of the cervical vertebrae, as well as difficulty in mouth opening (trismus). Jugular venous pressure and thyroid were normal, while she had pharyngeal erythema with no bulging.

The examination of the cranial nerves indicated no abnormalities, with the exception of CN VIII, which showed decreased hearing. Conjunctiva and sclera were normal, and there was no evidence of papilledema on ophthalmoscopy. The examination of the chest, abdomen, and extremities was normal, with the exception of digital clubbing. Neck radiography was normal without syndesmophite. Whole body bone scan was unremarkable.

The laboratory test results showed normal complete blood count, with the exception of the high neutrophil percentage, elevated ESR on several occasions (74 mm/h; normal: <20 mm/h), elevated quantitative C-reactive protein (CRP; 22.87 mg/l; normal: <6 mg/l), and elevated lactate dehydrogenase (527 U/l). The other tests were negative, including HLA-B27, Wright and 2ME, anti-nuclear antibody, anti-cyclic citrullinated peptide, and rheumatoid factor. Purified protein derivative was estimated at one millimeter after 48 hours, and the other routine tests were normal, such as calcium, phosphorus, hepatic function tests (alanine aminotransferase, aspartate aminotransferase, and alkaline phosphatase), and thyroid stimulating hormone. In addition, serum protein electrophoresis was indicative of hypergammaglobulinemia.

On neurology consultation, brain imaging with gadolinium-enhanced MRI and magnetic resonance venography (MRV) were performed. The brain MRI and MRV (without thrombosis) were normal. On the other hand, MRV incidentally showed a midline lesion with deviation to the right side with heterogeneous enhancement at

the nasopharynx. Restriction was also observed on the Durbin-Watson test, which extended to the apex of the petrous, suggesting inflammation. Moreover, bilateral mastoiditis was detected on MRI and MRV.

Spiral neck CT-scan showed several bilateral lymph node enlargements, soft tissue stranding, right-sided asymmetry of the Rosenmüller fossa (pharyngeal recess), and a heterogeneous enhancing mass on the nasopharynx roof with left extension and bilateral pressure on the Eustachian tube (Figure 1). In addition, temporal CT-scan showed a slight increase in the mucosal thickness of the left maxillary and sphenoid sinuses with bilateral middle ear effusion and mastoiditis. The IV contrast CT-scan of the abdomen, pelvis, and lung was normal.

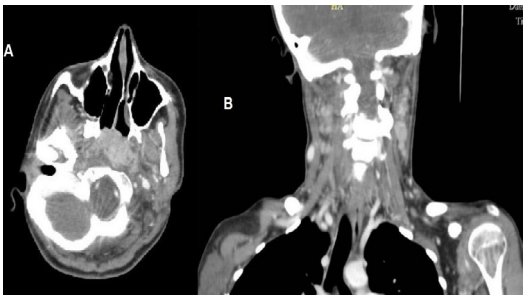

Figure 1. A) Axial View of Neck CT-scan with IV Contrast Showing a Large Nasopharyngeal Enhancing Mass; B) Coronal View of Neck CT-scan with IV Contrast Showing a Nasopharyngeal Mass with Several Enlarged Lymph Nodes.

Based on the findings of the neck CT-scan and presumptive diagnosis of a nasopharyngeal tumor, the patient was referred to the head and neck surgery department for tumor resection or biopsy. Considering the difficulty in mouth opening, awake fiber-optic intubation was initially planned. However, due to the severe nasal septal deviation, using a fiber-optic scope was not feasible. Therefore, the patient underwent general anesthesia with orotracheal intubation. Glottis, subglottis, and supraglottis were normal, and no lesions were observed in the vocal cords. Esophagoscopy (30 cm) was normal as well.

On nasal endoscopy, a nasopharyngeal mass was observed with a normal mucosal surface and extension to the masseter muscle. The surgical operation was uneventful. The nasopharyngeal mass biopsy and histopathologic examination indicated the infiltration of atypical epithelial cells with marked nuclear atypia in small, solid nests within the lymphoid tissue of the nasopharynx, which was compatible with poorly differentiated nasopharyngeal carcinoma. Considering the extension to the masseter muscle, T2, and N2, the patient was diagnosed with nasopharyngeal carcinoma (stage III) (Figure 2). The biopsy of the neck mass (fine needle biopsy) was unsatisfactory. The patient was referred for radiation oncology services and further tumor treatment.

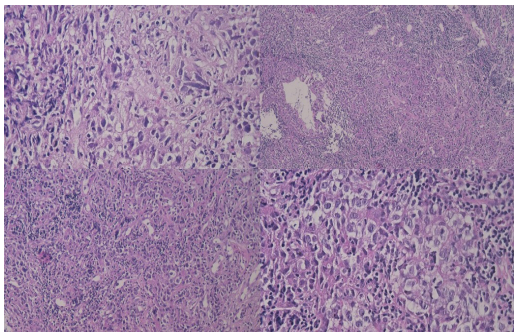

Figure 2. Histopathologic Examination of Nasopharyngeal Mass Biopsy Showing Infiltration of Atypical Epithelial Cells with Marked Nuclear Atypia in Small, Solid Nests within Lymphoid Tissue of Nasopharynx (H&E staining).

Discussion

Acquired torticollis is a sign that should be investigated thoroughly not only under neurogenic conditions, but also in the presence of other possible underlying causes, including tumors. We have presented the case of young patient with nasopharyngeal adenocarcinoma with the earliest and only symptom of acute, acquired torticollis. Although the patient developed some constitutional symptoms (e.g., fatigue, significant, unintentional weight loss, sweating, and trismus) within the following 2-3 months, the first symptom associated with her underlying malignancy was considered to be acquired torticollis. It is likely that the involvement of the neck muscles caused torticollis in the patient.

In the present case, the constitutional symptoms initially implied a systemic disease. As a result, a full work-up for infectious and inflammatory diseases was performed via imaging and laboratory tests. The neck CT-scan revealed a nasopharyngeal mass, and the patient presented peculiarly as she had no established risk factors for nasopharyngeal carcinoma, such as male gender, peak incidence in the sixth decade of life, Epstein-Barr virus infection, alcohol consumption and smoking habits, and geographical region (12).

Evidence is scarce regarding the association of torticollis with nasopharyngeal carcinoma. One of the report was on an eight-year-old male patient presenting with neck swelling and palpable enlarged lymph nodes. The patient was feverish, and the pharyngeal examination demonstrated the displacement of the right tonsil, which led to the presumptive diagnosis of peritonsillar abscess

considering the high fever of the patient. However, similar to the present case, CT-scan indicated a tumor, and biopsy confirmed the diagnosis of nasopharyngeal carcinoma (11).

The present case report emphasizes on the meticulous examination of the neck region, including the nasopharynx, in the patients with torticollis in order to determine the underlying cause. In another case report, nasopharyngeal tuberculosis was also observed in a 32-year-old female patient with neck pain and headaches for two years (13). In the mentioned study, CT-scan showed an enhanced tumor with the subsequent destruction of the adjacent bony structures.

Brain metastases has rarely been reported in the literature (14). Therefore, brain imaging was performed similar to the present study and was unremarkable. This modality is essential to the management of such patients. In this regard, fever is a non-specific finding, which often implies infectious causes. However, as observed in the present case and other similar pediatric patients with nasopharyngeal carcinoma (11), fever could occur under such circumstances

Conclusion

In case of acute, acquired torticollis and no history of infections or trauma, other less likely diagnoses in the particular malignancies of the head and neck region should be considered. This condition is more expected in the presence of constitutional symptoms and elevated serum levels of inflammatory markers (e.g., ESR and CRP). Imaging via contrast-enhancing CT-scan was the modality of choice, which enabled the diagnosis of nasopharyngeal mass.

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None.

Conflict of Interest

The authors declare no conflict of interest.

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