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Case report

A pediatric case of acute neck swelling due to bilateral submandibular sialadenitis following influenza A infection

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ABSTRACT

A 12-year-old boy presented with acute neck swelling diagnosed as submandibular sialadenitis secondary to influenza; it resolved quickly with supportive care. Recognition of this as a clinical entity during the influenza season may help guide management.

Introduction

Acute neck swelling is occasionally seen in pediatric clinical practice. It has a wide range of severity, from life-threating bacterial infections (such as Ludwig angina or deep neck abscess) to self-limited conditions (such as viral sialadenitis). In addition, non-infectious etiologies (including auto-immune disorders and sialolithiasis) are known causes [1]. Life threatening conditions often require pre-emptive intubation, aggressive antimicrobial therapy, and occasionally surgical intervention. Therefore, it is important to understand the etiology of acute neck swelling in the setting of the pediatric emergency department (ED) in order to determine optimal management.

Mumps and other viruses are a major cause of sialadenitis [2]. Influenza A (H3N2) is reported as a cause of parotitis [3], however, there are few reports of submandibular sialadenitis due to influenza virus [4,5] and the information regarding its clinical course is still limited. We report a pediatric case with rapid neck swelling that was ultimately diagnosed as submandibular sialadenitis due to influenza A (H3 subtype).

Case report

A 12-year-old boy presented to the pediatric ED with a history of bilateral lower jaw pain. Five days before admission, his disease was diagnosed as influenza A infection by the rapid influenza diagnostic test and was treated with inhaled laninamivir octanoate hydrate. He soon became afebrile; however, lower jaw pain developed one day before admission. He visited our pediatric ED because of rapid swelling and pain in the submandibular area. He was not toxic-appearing and had a body temperature of 36.5 °C, heart rate of 75 beats/minute, and respiratory rate of 24/minute (on room air).

Physical examination revealed soft bilateral swelling and tenderness of the submandibular area (Fig. 1, Panel A). The orifice of the submandibular duct was also swollen and red. Cervical lymph nodes were not palpable and no respiratory distress was observed. Complete blood count and chemistry was normal with white blood cell count 4610/μL (45.8% neutrophils, 41.1% lymphocytes), amylase 87 U/L, and C-reactive protein 0.11 mg/dL. A neck CT scan with contrast revealed bilateral enlarged submandibular glands with homogeneous contrast enhancement (Fig. 2); however, the airway was patent and no abscess was detected.

He was admitted to our hospital for careful observation for the potential risk of progressive airway obstruction. Neck swelling improved spontaneously without antibacterial therapy (Fig. 1,

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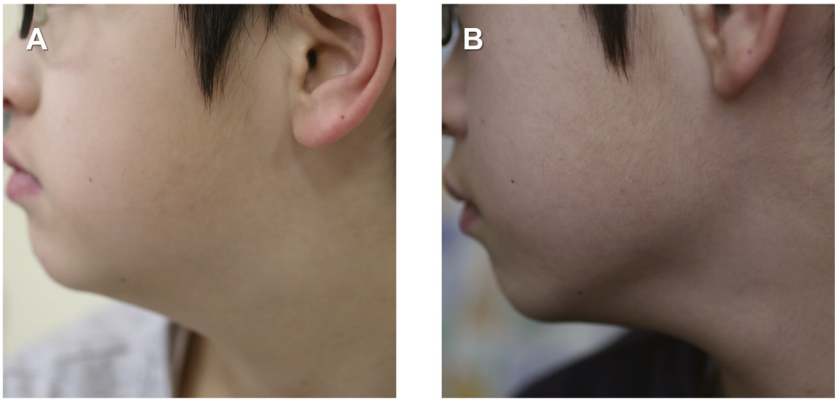

Fig. 1. Lateral view of the patient's face at admission (panel A) and at discharge (panel B). The submandibular area was swollen but soft, and his general appearance was fair in panel A. The submandibular area improved without complications, as shown in panel B.

Fig. 2. Contrast-enhanced CT image at admission. Bilateral submandibular glands were enlarged and enhanced homogeneously (arrows).

Table 1
Three cases of submandibular sialadenitis associated with influenza virus.

	Age (years)	Sex	Influenza type	Laboratory test	Imaging	Onset to swelling (days)	Time to improvement	Other symptoms	Mumps vaccination	Treatment	Underlying disease	Prognosis
Case 1 (Current case)	12	Male	A (H3 subtype)	RIDT/PCR	CECT	6	10 days	No	Once	No	No	Good
Case 2 (Ref. 4)	7	Male	A	RIDT	CECT X-ray US	3	Several days	Cough, rhinorrhea, fever	No data	No	No	Good
Case 3 (Ref. 5)	45	Male	A	RIDT	CT	11	Several days	General fatigue	No data	No	No	Good

CECT, contrast-enhanced CT; PCR, polymerase chain reaction; RIDT, rapid influenza diagnostic test; US, ultrasound.

Panel B) and he was discharged 5 days after admission without any complications. The saliva was tested by polymerase chain reaction (PCR). A saliva swab around the submandibular gland was collected and nucleic acid was extracted using the QIAamp DNA Mini Kit (QIAGEN, Hilden, Germany) according to the manufacturer's instructions. Influenza and mumps viruses were tested by multiplex real-time RT-PCR using the "FTD Flu differentiation" and "FTD Mumps" kits (Fast-Track Diagnostics, Luxembourg), respectively. Influenza A (H3 subtype) was positive and mumps virus was negative. Based on this data, the diagnosis was confirmed as submandibular sialadenitis following influenza A (H3).

Discussion

Many conditions cause acute neck swelling in children and some of these are potentially life-threatening [2]. Therefore, it is important to exclude the possibility of emergency airway obstructive condition due to bacterial infection (including Ludwig angina and deep neck abscess) for these patients. Ludwig angina is a bacterial infection involving the sublingual, submaxillary, and submandibular spaces [2]. It generally presents as a rapidly progressive, bilateral, submandibular, sinewy, "board-like" swelling that often requires tracheal intubation for airway control [2]. In contrast, the current case showed rapid neck swelling without any signs indicating severe bacterial infection (high fever, trismus, limited neck motion, or respiratory distress). Capturing the differences among these symptoms along with enhanced CT is sometimes helpful in differentiating lethal and benign underlying causes and pathology of the patient with rapid neck swelling.

Infection may cause parotitis or mumps-like illness in children [6]. The period prevalence of parotitis among patients infected with influenza virus differs according to location and year. Barrabeig et al. [7] reported the etiology of the suspected mumps cases in Spain from 2007 to 2011; EB virus was shown to be the major pathogen (24.8%), whereas influenza virus was detected in only 1% of cases. Elbadawi et al. [8] reported that 53% of non-mumps parotitis cases were associated with influenza virus during the 2014–2015 winter season in the United Status. Thompson et al. [3] also reported that 15% of children with a diagnosis of non-mumps viral parotitis in England were infected with influenza A (H3N2) virus during the 2014–2015 influenza season.

The prevalence of submandibular gland sialadenitis (not parotitis) associated with influenza virus is still unclear and only a few reports are available [4,5]. We summarized the details of cases of submandibular sialadenitis, including our case, in Table 1. Duration from the onset of influenza infection to neck swelling was 3–11 days and swelling spontaneously improved within several days. Reported cases of influenza-associated parotitis followed a similar clinical course [9,10].

Conclusions

Influenza infection may cause rapid neck swelling due to submandibular sialadenitis. Proper diagnosis may prevent unnecessary intubation and administration of antibiotics. ED clinicians should consider submandibular sialadenitis secondary to influenza infection in the differential diagnosis of rapid neck swelling during the influenza season.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflict of interest statement

The authors have indicated they have no financial relationships relevant to this article to disclose.

We would like to declare the contribution of each authors as follows. Mei Ikenori and Kensuke Shoji conceptualized and drafted the manuscript, Toshihiro Matsui, Akira Ishiguro critically reviewed and edited the manuscript, Naoko Kono preformed the PCR analysis and reviewed the methods, Isao Miyairi supervised the manuscript and is responsible for funding acquisition.

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