

ORIGINAL RESEARCH

Dysphonia as an Indicator of Impending Respiratory Arrest in Patients with C1-C2 Fractures

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ABSTRACT

Introduction: Patients with cervical spine injury in the absence of spinal cord injury are at risk for developing airway compromise secondary to retropharyngeal swelling or hematoma formation. In particular, C1 (atlas) and C2 (axis) fractures have been associated with up to a 4.9% incidence of developing airway compromise, even in the absence of spinal cord injury. Our objective was to describe a physical sign associated with respiratory distress in a case series of upper cervical fractures.

Methods: The study was a retrospective chart review. Neurologically intact patients with cervical spine injuries who developed respiratory distress requiring intubation were identified, and their electronic medical records, cervical CT scans, and cervical MRI scans were reviewed. The clinical course of patients with respiratory distress and the descriptive statistics of the patient population were analyzed.

Results: Five male patients were identified with the mean age of 64.8 years (± 18.85). All presented within 24 hrs of onset of injury with mean ISS score of 19 (± 8.8) and had prevertebral swelling between 12.1 and 23.1 mm. All developed dysphonia prior to respiratory distress, and two patients subsequently died.

Discussion: Older patients with C1 fractures may be at high risk for developing airway compromise. If acute dysphonia develops, intubation should be considered for definitive airway management.

Keywords: Dysphonia; Cervical fracture; Retropharyngeal hematoma; Respiratory distress.

INTRODUCTION

Acute respiratory failure in the absence of spinal cord injury is a known but rare

complication of cervical spine fractures. Retropharyngeal hematomas that become large enough to obstruct the airway in cervical trauma settings have been reported to be a cause of respiratory failure in this trauma population (1). Airway compromise is a life-threatening medical emergency, and attending

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physicians should be aware of this possibility in cervical fracture patients regardless of the severity of the trauma (2). Additionally, there may be a subset of cervical fractures in which the incidence of airway compromise is higher. One recent study reported that the incidence of respiratory distress is 4.9% in isolated C1 and C2 fracture patients (3). The underlying mechanism is a mechanical obstruction resulting in soft tissue impingement of the airway secondary to retropharyngeal swelling or hematoma (1-5). Many early signs of respiratory compromise may be non-specific, which can make diagnosis in the acute trauma situation challenging. Recognizing prevertebral soft tissue swelling or retropharyngeal hematoma on radiographic imaging prior to impending acute respiratory failure may help the modern multidisciplinary trauma team prepare for airway issues in the patient with cervical fractures (4).

Clinical predictors and risk factors in cervical trauma patients developing airway compromise have been previously assessed, and the results have indicated that those with a level of injury above C5, fracture displacement, preexisting cervical degenerative changes, significant prevertebral swelling, complete spinal cord injury, male, advanced age, and Injury Severity Scale (ISS) greater than 16 were at higher risk of airway compromise (3,6). Two other studies stated that patients with anticoagulation therapy, coagulation disorders, vascular lesions, associated head injury above C3, and a Glasgow Coma Scale score of less than 8 require more frequent emergent airway management (7, 8).

Dysphonia has been reported to be an important clinical predictor of early airway compromise from multiple etiologies (9). To our knowledge, there has not been a

study investigating dysphonia as a possible early predictor of impending acute respiratory arrest in patients with cervical trauma. Clinical experience at our institution suggests that dysphonia was frequently present in isolated C1-C2 fracture patients who went on to experience respiratory distress. This case series examines the incidence of respiratory failure in this population and documents a possible association between the presence of dysphonia and subsequent respiratory distress in neurologically intact patients with fractures at C1-C2.

MATERIALS & METHODS

A descriptive retrospective cohort study was performed using a comprehensive electronic medical record search of patients with C1-C2 fractures at Scott and White Memorial Hospital (SWMH) (Level I trauma center) in Temple, Texas from years 2005 to 2010. In addition, using the SWMH trauma registry database, the total number of trauma patients and cervical fracture patients over the same period were searched for comparison purposes. Exclusion criteria included associated spinal cord injury, altered mental status on arrival, previous vocal cord injuries, penetrating injury to the neck or chest, previous cervical fractures, long bone fractures, co-morbidities that directly affect respiratory function, and heavily sedated patients.

The study group was identified from electronic medical records using International Classification of Diseases (9th Revision) codes for C1-C2 fractures, and the electronic medical records of all identified patients were reviewed. Patients who were not intubated on arrival, had isolated C1 or C2 fracture, and required

emergent intubation for acute respiratory distress or failure were selected for further chart and radiographic review. The medical records of these patients and respective radiographic imaging studies were extracted to gather further information including age, sex, mechanism of injury, type of fracture, cervical spine involvement, presence and size of prevertebral swelling, clinical presentations and descriptors used to assess dysphonia or respiratory difficulty, associated spinal injuries or other systemic injuries, intubation requirements, number of days on ventilator, total number of hospitalization days, ISS score, treatment methods, clinical condition at discharge, and the condition of the patient at last contact.

Cervical spine fractures in these patients were confirmed by MRI or CT imaging; prevertebral swellings in the cervical region were measured using available digital tools included with the Centricity Enterprise Web V3.0 viewer.

The criterion for airway compromise in this study was classified as acute oxygen desaturation below 80% after a C1-C2 fracture that required immediate intervention or intubation to preserve the airway, in the absence of any other diagnosis that could have induced respiratory failure.

RESULTS

From 2005 to 2010, the total number of trauma patients seen at SWMH was 12,265. Of these patients, 469 had C1-C7 fractures (3.8%) and 208 had C1-C2 fractures (1.7%). A total of 5 patients met the inclusion criteria defined in the Materials & Methods section. This number represented 0.04% of all trauma patients and 1.9% of all C1-C2 fracture patients.

All 5 patients had C1 anterior arch fractures, and 4 of these patients also had C2 fractures, which were all confirmed by radiographic studies and reports. The mean age of this group was 64.8 (± 18.85) years, and they were all male. All of the patients presented within 24 hours of onset of injury. A prevertebral swelling or a retropharyngeal hematoma was present in all patients on MRI and CT of cervical spine (Figures 1 and 2), and the mean prevertebral swelling was 15.35 (± 4.472) mm. Four of five injuries were due to falls, and one was due to motor vehicle accident. The mean ISS score was 19 (± 8.8). No record of anticoagulation therapy was found for any of these patients. Further information of the study group is presented in Table 1.

All of the patients presented with dysphonia or hoarseness either upon admission or shortly thereafter (24-48 hrs). Some of the descriptive terms used by physicians were: dysphonia, dysphagia, progressive stridor, breathing difficulty, and voice changes. The senior author (C.D.C.) was consulted for the cervical fractures, examined the patients, and confirmed the presence of dysphonia in all 5 patients. Acute respiratory failure developed in all patients shortly after admission, and, upon signs of airway compromise, 4 patients were intubated immediately. A 90-year-old male (Patient 1) with multiple medical illnesses included in the study declined surgical intervention and further supportive care such as intubation. On hospital day 5, this patient suffered a respiratory arrest and died.

Of the 4 who were intubated, the mean number of days on ventilator was 3.5 (± 2.38) days. All of these intubated patients underwent surgical stabilization, and all but one survived. After undergoing posterior

Table 1. Patient demographic and injury characteristics.

Pt	Age	Sex	Injury Mechanism	Fracture Type	Prevertebral Swelling [mm]	Dysphonia/ Hoarseness Assessed?	Descriptor	Intubated Days	Hospital Days	ISS	Timing of Events	Type of Treatment	Outcome/ Condition at Discharge
1	90	M	Fall	C1-C2	12.12	Yes	Dysphonia, Dysphagia	N/A	5	9	<24h	Palliative care C-collar	Death
2	50	M	Fall	C1-C3 C6-C7	12.83	Yes	Dysphonia, Dysphagia, Progressive stridor	5	8	27	<24h	Surgery: Anterior+Posterior stabilization	Stable
3	54	M	MVA	C1, C3	23.14	Yes	Dysphonia, Breathing difficulty	1	8	16	<24h	Surgery: Posterior stabilization, Laminectomy	Stable
4	80	M	Fall	C1-C2	13.88	Yes	Dysphonia, Breathing difficulty, Voice change	6	14	29	<24h	Surgery: Posterior stabilization	Death
5	50	M	Fall	C1-C2	14.78	Yes	Dysphonia	2	6	13	<24h	Surgery: Evacuation of retropharyngeal hematoma	Stable

Pt, patient number; ISS, Injury Severity Scale; MVA, Motor vehicle accident.

stabilization, the 80-year-old male (Patient 4) was extubated. However, 2 days after extubation, the patient developed a second event of acute respiratory distress and was emergently intubated. He subsequently had a cardiac arrest, suffered anoxic brain injury, and died.

Patient 2 underwent posterior fixation and was extubated without any incident. This patient complained of mild dysphagia at a follow-up appointment, which subsequently resolved. Patient 3 had a large hematoma of 23.14 mm in size, and, after posterior stabilization, his condition improved and he was discharged to a rehabilitation facility in stable condition. Patient 5 also had a large hematoma (14.78 mm), which was subsequently evacuated, and he made a full recovery (Table 1, Figures 1 and 2).

DISCUSSION

The retropharyngeal space is classically described as a space bounded by the pharynx and the cervical spine. The anterior



Figure 1. CT scan of cervical spine (Patient 4) showing an increase in the prevertebral soft tissue measurements possibly representing hematoma or swelling.

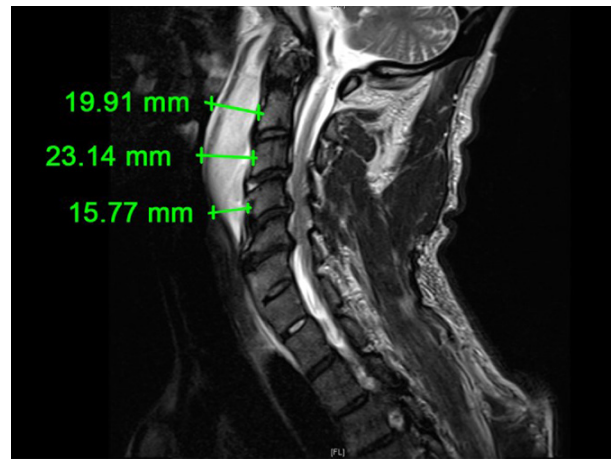


Figure 2. MRI of cervical spine from the same patient in Figure 1 (Patient 4) confirming the presence of a large retropharyngeal hematoma that measures 23.1 mm anterior to C3.

wall is bordered by the buccopharyngeal fascia, the posterior wall by the prevertebral fascia, and the lateral wall by the connective tissue of carotid sheath (10). The normal measurement of this space is between 1 and 7 mm, and a conventionally accepted rough estimation is made by noting that the retropharyngeal soft tissue space is normally less than one-half of the width of the cervical vertebra on plain radiography (10-12). The mechanism of acute respiratory failure secondary to cervical trauma is thought to involve retropharyngeal swelling or hematoma that can cause mechanical obstruction and eventually airway compromise (10-13). Smith et al. (13) reported that this is a well-recognized cause of upper airway obstruction. While the exact mechanism of the swelling is unclear at this point, there are several potential sources of bleeding that may contribute to this phenomenon: the actual fracture of the vertebrae, a tear of the anterior longitudinal ligament, tearing of the longus colli muscle, or ruptures of the vascular structures themselves (2,10).

A detailed review of the literature regarding the anatomy of the upper neck

revealed several potential venous and arterial sources that may be responsible for developing hematomas (14-17). The fractures of the anterior arch of C1-C2 can leak venous blood from the bone or the intervertebral plexus themselves, causing venous blood to drain into the retropharyngeal space.

Arterial blood flow to the anterior suboccipital region primarily consists of branches from the ascending pharyngeal artery, which has multiple anastomotic connections involving the muscular branches of the vertebral artery, the ascending cervical branch of the inferior thyroid artery, and the occipital arteries. Bleeding in the suboccipital region may come from spinal branches of the vertebral arteries that supply the vertebral bodies, muscular branches of the vertebral artery, or branches of the ascending cervical branch of the inferior thyroid artery that supply the deep muscles of the neck. Fractures or tearing of any of these structures or their associated arteries can lead to hematoma formation in the retropharyngeal space (16).

The primary source of arterial blood to this region comes from the ascending pharyngeal artery, which lies on the posterior aspect of the pharynx and gives off numerous vessels that supply the longus capitis and longus colli muscles, the sympathetic trunk, the hypoglossal and vagus nerves, and a number of the deep cervical lymph nodes (16). The detailed anatomy of the ascending pharyngeal artery and its relationship to the suboccipital vertebra were described in an angiographic study that demonstrated its vulnerability to injuries in this region (17). After a short common trunk, the artery divides into an anterior and posterior trunk. The anterior trunk supplies the pharyngeal constrictor muscles and the pharyngeal submucosal spaces.

Rupture of these branches may result in bleeding directly into the retropharyngeal space. The posterior trunk of the ascending pharyngeal artery makes two important contributions to the pathology observed in upper cervical fractures. First, the posterior descending branch contributes to the odontoid arch system, anastomosing with branches from the vertebral artery. These branches may be disrupted in the fracture involving C1 and C2, with resultant bleeding directly into the retropharyngeal space. The other important contribution is from the hypoglossal and jugular branches of the neuromeningeal trunk. These arterial branches supply the vasa vasorum of cranial nerves IX-XII (5,6). An insult to these small vessels can lead to ischemia of the involved cranial nerves (primarily the vagus), which can contribute to the primary clinical findings of dysphonia, dysphagia, or other signs of respiratory distress.

In the setting of fractures at C1 or C2, however, bleeding bone likely plays a major role in hematoma formation, as in other fractures. Unlike the airway more distally, there are no cartilaginous supporting structures in the airway in this region that might restrict swelling or help tamponade bleeding. This may make the retropharyngeal space particularly susceptible to formation of an expanding hematoma and swelling. Multiple studies have indicated that neurologic impairment is not the cause of respiratory failure in these reported cases (2,10,12,13).

Airway compromise secondary to cervical spine surgery or manipulation has been well documented. The incidence of respiratory compromise secondary to traction and manipulation for cervical fractures is reported to be as high as 10%, with higher risk seen in posteriorly displaced fractures (12,18).

Table 2. Terms used to describe signs or symptoms of acute respiratory distress.

Dysphonia	A general term used to describe an abnormal voice highlighted by altered quality, pitch, loudness, or effort (9,21). Dysphonia and hoarseness are often used synonymously, but dysphonia is a diagnostic terms, whereas hoarseness is a symptom of voice change and is usually characterized by a coarse or rough quality (9).
Dysphagia	The symptom of difficulty in swallowing. It develops owing to functional or structural problems in control of the esophagus, oral cavity, pharynx, or gastric cardia. Acute dysphagia can lead to airway obstruction and death secondary to asphyxiation (22).
Stridor	An abnormal, high-pitched sound created by a blockage in the throat or the voice box during inspiration. It is commonly described as “musical breathing”(23).
Dyspnea	Also known as breathlessness, defined as the symptom of breathing discomfort and is synonymous with “shortness of breath.” This experience is caused by excessive stimulation of the respiratory center, which results from the activation of different receptors of the upper and lower airway, lung tissues, or chest wall, as well as activation of central and peripheral chemoreceptors (24).

Harrop et al. (12) reported that this complication is most likely due to development of a retropharyngeal edema, and the immobilization of the cervical spine with concurrent flexion resulting in compression of trachea in these patients. In addition, it has also been reported that 6% of patients who underwent anterior cervical spine surgery experienced respiratory failure, and this complication was most likely due to extensive tissue trauma (18-20).

There are several different clinical terms used by physicians to describe breathing difficulties or signs of respiratory distress (9,21-24). For clarification, we have attempted to distinguish between dysphonia and these other terms as depicted in Table 2.

Dysphonia has been associated with impending airway compromise (7,9,21). When dysphonia occurs, there may be little time to intervene and treat the impending airway compromise. A challenging aspect

of managing these patients is that they may not initially seem to need urgent airway management. These patients may present with vital signs and respiratory status that are within normal limits. However, the status of these patients can deteriorate quickly and unpredictably, as it was seen in our patients. Patients who present with dysphonia may also have a spectrum of clinical findings that may indicate respiratory distress, such as difficulty speaking, increased phonation effort, weak voice, raspiness, or breathiness (3,9). However, changes in vocal quality are infrequently assessed in the clinical setting and have not been found to predict acute respiratory failure in cervical trauma patients. Our review of current articles in the fields of orthopaedics, spine surgery, neurosurgery, and emergency medicine disclose rare mention of this phenomenon. While some of these articles described stridor, noisy breathing, and/or apnea in patients with imminent airway compromise, the current

initial trauma assessment does not typically include documenting changes in vocal quality or acute onset of hoarseness (2,5).

Kuhn et al. (5) and Mitchell et al. (10) reported cases of airway compromise secondary to retropharyngeal hematoma following cervical spine injury. Some of the descriptors used in these case studies were stridor, difficulty with oropharyngeal secretions, dysphagia, and difficulty breathing. Only one of the patients in the study was documented to have hoarseness (1). Unfortunately, the presence or absence of hoarseness was not assessed in all of these patients, and it would have been interesting to know if those patients also presented with changes in vocal quality as well.

The common characteristics of the 5 studied patients at our institution over a 5-year-period were consistent with the patients in previously reported cases (2,3,5,6,20). They were all older males with mean age of 64.8 years, and had retropharyngeal hematomas measuring greater than 12 mm. The mechanisms of the injury (4 falls and 1 MVA) were also consistent with the previous findings. Three of the patients had ISS score greater than 16, and 3 of them made full recoveries. All of these patients initially presented with normal respiration status and normal vital signs; but, they developed dysphonia within 24-48 hours and quickly deteriorated to acute respiratory compromise. The patients also had other clinical findings, such as stridor, difficulty breathing, and dysphagia, which are also consistent with the characteristics in other previous reports. Two patients (ages 80 and 90) died as a result of this complication. One elected to receive palliative care due to multiple comorbidities and a poor surgical prognosis. The other patient suffered a second respiratory failure after surgery and

subsequent cardiac arrest resulting in anoxic brain injury. Three of the 4 patients who were immediately intubated and received surgical care had no airway or dysphagia issues at follow-up of greater than 1 year.

These cases were potentially catastrophic, and careful observation in these patients who showed signs of changes in their vocal quality and ensuing onset of respiratory distress was crucial to the management. Cleiman et al. (25) reported on a patient who did not seem to need emergent airway management but soon presented with airway compromise. They also emphasized the importance of acknowledging the possibility of airway compromise before clinical deterioration. While there are other signs that may indicate impending respiratory distress, dysphonia is one key to recognizing early signs of airway compromise. In our experience, the recognition of this prodrome can allow a definitive airway to be established in a controlled, prepared fashion prior to a frank respiratory collapse.

Current management of cervical trauma patients includes initial assessment of vital signs, patient's airway, oxygenation status via pulse oximetry on room air and oxygen, and cervical immobilization. Owing to high proportion of patients in the current series who had a prodromal period of dysphonia prior to respiratory distress, characterization of the vocal quality in a patient with cervical trauma appears to be an important additional part of the physical exam for acute vocal changes in a trauma setting. Schwartz et al. (9) recommended listening to the voice in a critical and objective manner for important diagnostic information and visualizing the larynx with bedside laryngoscopy for early signs of airway obstruction. In contrast, we do not find attempts at visualizing the larynx solely for the purpose

of diagnosis as necessary if a patient has dysphonia and CT evidence of a potential source of swelling and airway compromise. In the setting of upper cervical trauma, laryngoscopy without a definitive airway can be difficult, and there are potential risks to doing this in the setting of an unstable cervical spine. Our practice is to intubate the patient urgently with appropriate equipment and anesthesia support to address a difficult airway in a patient who requires strict maintenance of cervical spine precautions.

This was a retrospective cohort study, and there were some limitations. All retrospective studies face challenges such as the accuracy of the medical record and missing or undocumented data. Assessing the possibly subtle changes in the voice of a patient may be difficult for physicians who lack specific training. In addition, the subjective nature of assessing changes in the voice of a patient may be subject to bias and may vary from one physician to another. However, in our experience, the patient or a family member usually notices and describes the change in voice if asked, and they can often help with the question of whether the voice has changed significantly since the accident. Another limitation may have resulted from our inclusion or exclusion criteria. There may have been patients who developed dysphonia and airway obstruction secondary to C1 or C2 fracture, and who were excluded due to other associated injuries that could potentially lead to respiratory compromise, specifically those with neurological injury, which may have multifactorial causes of respiratory failure. Furthermore, since dysphonia is infrequently assessed in acute trauma settings, there may have been patients whose dysphonia was either not noticed or undocumented. Despite these limitations, the present study suggests that

dysphonia can occur prior to respiratory distress in the setting of upper cervical fractures and further prospective studies may be warranted.

CONCLUSIONS

Patients with upper cervical fractures may initially present with normal respiratory findings. However, such patients can quickly deteriorate and require emergent intubation for respiratory distress. A high index of suspicion for impending airway compromise with upper cervical fractures should be maintained, especially those with acute prevertebral swelling or fluid on cervical CT or MRI. The elderly may be at particular risk for respiratory arrest and death from these injuries, which often occur after relatively minor falls from a standing height. Because of this, monitoring in the ICU should be considered for this subset of patients, even if other ICU criteria are not met. If dysphonia develops—even in an otherwise stable patient—intubation should be considered urgently in order to establish a definitive airway.

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