Case Report

Long-term resolution of delayed onset hypoglossal nerve palsy following occipital condyle fracture: Case report and review of the literature

ABSTRACT

The authors present the case of a patient that demonstrates resolution of delayed onset hypoglossal nerve palsy (HNP) subsequent to occipital condyle fracture following a motor vehicle accident. Decompression of the hypoglossal nerve and craniocervical fixation led to satisfactory long-term (>5 years) outcome. There is a scarcity of literature in recognizing HNPs following trauma and a lack of pathophysiological understanding to both a delayed presentation and to resolution versus persistence. This is the first report demonstrating long-term resolution of hypoglossal nerve injury following trauma to the craniocervical junction.

Keywords: Cranial nerve palsy, craniocervical fracture, hypoglossal, nerve compression, neuropraxia, occipital condyle fracture, tongue anesthesia

INTRODUCTION

A growing number of reports have illustrated hypoglossal nerve injury after occipital condyle fractures (OCFs).[1-9] Despite rare recognition, treatment options remain indicated on the severity of OCF.[10,11] Only one case in the literature has suggested improvement in the subacute presentation of hypoglossal nerve palsy (HNP).[7] Unfortunately, all other studies have demonstrated persistence HNP varying from 3.5 months to 2 years following OCF [Table 1].

The exact mechanism by which OCFs contribute to HNP and in the delayed presentation is unclear. Thus far, case reports have demonstrated an association between OCFs and hypoglossal nerve injury [Table 1], but no case has demonstrated causality. Here, we report on a patient in whom an avulsed OCF was identified with a delayed presentation of HNP, and after removal and stabilization, resolution of neuropraxia was demonstrated on long-term follow-up (>5 years).

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CASE REPORT

Here, we report on a 20-year-old unrestrained female passenger involved in a motor vehicle accident (MVA) who presented to an outside hospital. Neurological examination revealed no focal deficits on the initial presentation or during inpatient course. She was diagnosed with right hip, pelvis, and OCF and underwent right hip open reduction and internal

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fixation as well as placed in a hard collar. At 6 weeks, she presented to our institution with a new complaint of rightward tongue deviation. Repeat computed tomography (CT) imaging revealed an Anderson and Montesano (AM) Type III OCF with a large bone fragment resting in the epidural space at the foramen magnum-C1 junction without change from the initial CT imaging study after MVA [Figure 1]. The patient was recommended for surgery and subsequently underwent a limited suboccipital craniectomy with C1 laminectomy, removal of bone fragment in C1 ventrolateral epidural space [Figures 2 and 3] with lysis of arachnoid adhesions, and occiput to C1 fusion [Figure 4]. Postoperatively, her right tongue deviation appeared to improve on postoperative day (POD) 1 and completely resolved by POD 2. On both short- and long-term follow-up, she remained without evidence of tongue deviation at 2 weeks and 5 years, respectively.

DISCUSSION

This is the first case in which a causal relationship has been reported between an OCF and neuropraxia of the hypoglossal nerve. We illustrate delayed presentation of unilateral tongue deviation after AM Type III OCF and importantly after surgical decompression with fixation, the palsy resolved. Evidence here, after removal of the avulsed OCF, suggests compression along the deep course of the hypoglossal nerve may contribute to delayed neuropraxia.

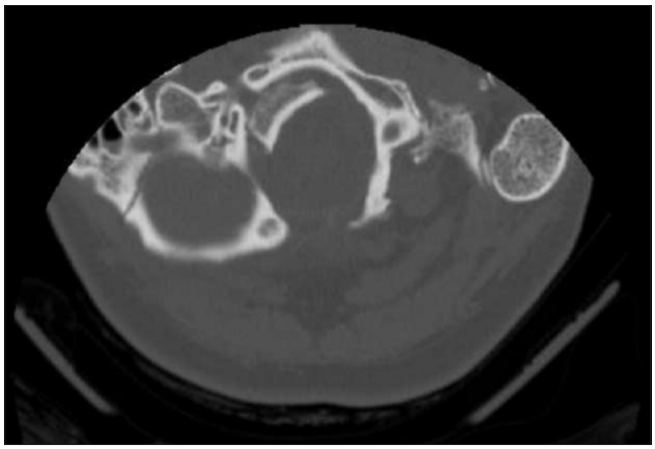

Figure 1: Computed tomography imaging-based evidence of a displaced right occipital condyle fragment in the epidural space adjacent to the hypoglossal canal

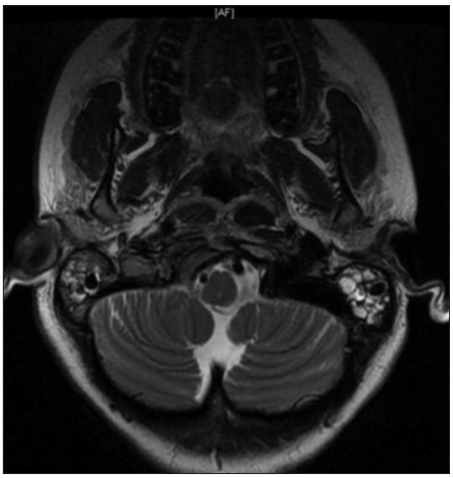

Figure 2: Magnetic resonance imaging-based evidence of a displaced right occipital condyle fragment in the epidural space adjacent to the hypoglossal canal

Table 1: Cases of delayed onset hypoglossal nerve palsy after occipital condyle fracture

Prior literature first author	Age/sex	Mechanism injury	CN12 palsy/onset	LOC	Time until onset	Fx class	CCJ misalignment	Tx	Follow-up period	CN12 palsy outcome
Muthukumar	32 male	Motorcycle	Yes/delayed	Yes	3 weeks	N/A	No	Rigid cervical collar for 12 weeks	18 months	Unchanged
Demisch	45 female	MVA	Yes/delayed	Yes	9 weeks	N/A	Yes	Rigid cervical collar	12 months	Unchanged
Orbay	37 male	MVA	Yes/delayed	No	12 weeks	N/A	No	Soft cervical collar	15 months	Unchanged
Hicks	21 male	Motorcycle fall	Yes/delayed	Yes	6 days	Anderson and Montesano Type II	N/A	None	2 years	Unchanged
Rué	19 male	MVA	Yes/delayed	No	2 weeks	N/A	No	Cervical brace unspecified	Remote unspecified	Unchanged
Wasserberg	39 male	MVA	Yes/delayed	Yes	3 weeks	Anderson and Montesano Type III	Yes	None	N/A	Unchanged
Legros	44 male	MVA	Yes/delayed	No	2 days	Anderson and Montesano Type II	No	Cervical brace for 6 weeks	3.5 months	Resolved
Noble	33 male	MVA	Yes/delayed	N/A	N/A (S)	Anderson and Montesano Type I	N/A	None	Unreported	N/A
Bridgman	32 male	Fall down stairs 12 feet	Yes/delayed	No	Few days	Anderson and Montesano Type III	No	Rigid cervical collar	1 year	unchanged
Current Study	20 female	MVA	Yes/delayed	No	6 weeks	Anderson and Montesano Type III	Yes	Decompression, bone fragment removal, O-C1 fusion	5 years	Resolved

MVA - Motor vehicle accident; LOC - Loss of consciousness; CCJ - Craniocervical junction; N/A - Not available; S – Significant; CN - Cranial nerve

Delayed cranial nerve palsies after OCFs are a rare complication, with an incidence of <1%, and unilateral HNP the most commonly reported.[1-9,12,13] Nine cases of delayed onset isolated HNP after OCFs have been previously reported [Table 1].

Two popular classification systems for OCFs help guide treatment options [Table 2].[14] The AM system classifies the fracture based on various unrelated factors that allow overlap between types and the Tuli *et al*. system on the location of the fragment and radiographic evidence of craniocervical joint (CCJ) instability.[10-12] AM Type I and II OCFs are treated with rigid cervical orthosis with symptomatic improvements and complete union after 6–12 weeks in the majority of patients.[8,12,14,15] AM Type III OCFs are treated with halo fixation if unstable on flexion-extension imaging or rigid cervical collar if stable.[12,13,15] Despite the current classification systems, their clinical utility is limited since the treatment of the various types is similar.

Table 2: Classifications of occipital condyle fractures

	Treatment
Anderson and Montesano[10]	
Type I - comminuted fracture	Rigid cervical collar
Type II - extension of basilar skull fracture	Rigid cervical collar
Type II - avulsed condyle with compromise of alar ligament	Rigid cervical collar or halo fixation
Tuli *et al*.[11]	
Type I - no displaced fragment	Rigid cervical collar
Type IIa - displaced fragment without evidence of radiographic instability	Rigid cervical collar
Type IIb - displaced fragment with evidence of radiographic instability	Halo fixation
Harding-Smith	-
Type I - horizontal fracture with axial decompression	-
Type II - oblique with displaced bone fragment crossing both jugular foramen and hypoglossal canal	-

The exact etiology of the delayed onset HNP after OCF is unknown. The hypothesized mechanisms of delayed onset include callus formation during normal healing applying traction to the hypoglossal nerve as it exits the canal, bone fragment movement impinging on the hypoglossal nerve near the canal, and secondary edema.[1,4,16] Furthermore, airway management has been reported to cause delayed HNP. When considering injury during airway securement, nearly, all HNPs presented in acute or subacute fashion (POD 1–3) and majority resolved within 6 months without intervention, unlike the cases reviewed in this paper.[17,18] Only one of the ten cases of delayed HNP involved intubation in the course of their management,[2] and all presented at least 1 week or later with symptoms persisting for months or greater despite conservative management.

The etiology of our patient's HNP may be related to fracture instability [Figure 5] though no gross movement was identified on repetitive imaging. It is unlikely due to airway management given no recent history of intubation before symptom onset. The patient denied any event, traumatic or infectious, preceding the onset of her HNP.

CCJ misalignment and neural element compression are the predominant indications for surgical intervention despite the classification of fractures,[8,12,13,19,20] although conservative management has been reported for such instances.[21] In our case, the patient presented 6 weeks after her MVA with an acute onset of tongue deviation. The patient was started on steroids and taken to the OR <72 h from onset

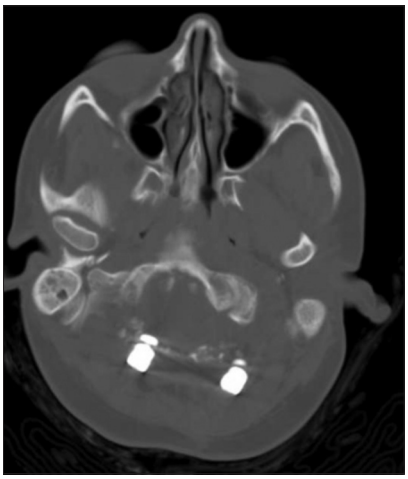

Figure 3: Postoperative computed tomography scan showing removal of condylar fragment

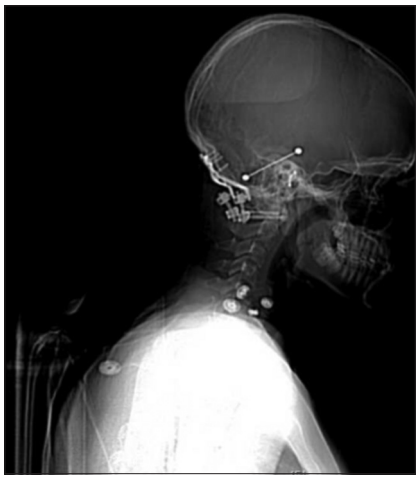

Figure 4: Postoperative X-ray demonstrating occipital-cervical fixation from occiput to C1

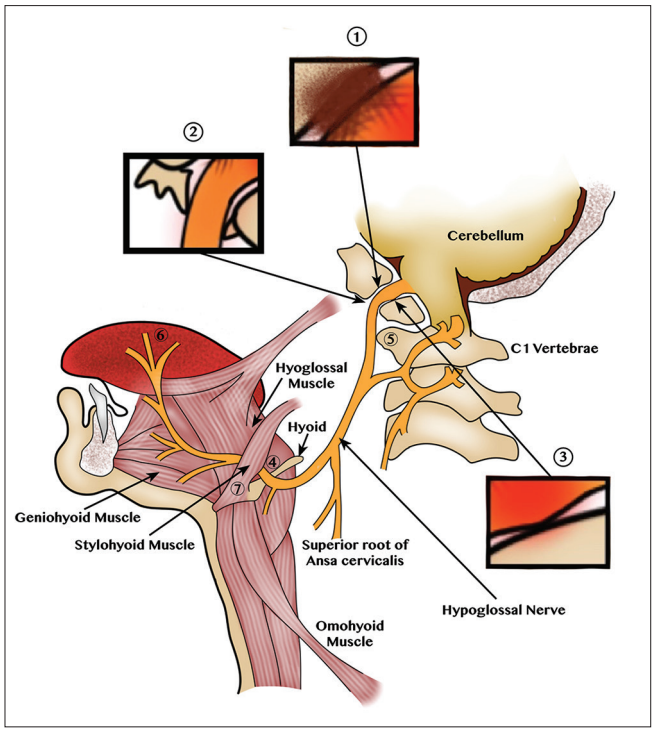

Figure 5: Illustration of CN12 injury from reported etiologies. Regions of CN12 injury after occipital condyle fracture. (1) Condylar callus with CN12 traction through the canal. (2) Displaced fragment impinges CN12 on canal exit. (3) Edema-induced CN12 compression during canal exit. Regions of CN12 injury during airway management. (4) Superficial CN12 course with resultant impingement. (5) CN12 stretch at the C1 transverse process. (6) Laryngoscope pressure on the tongue with resultant distal CN12 injury. (7) CN12 impingement from a calcified stylohyoid ligament modified from Shah *et al*.[17]

of symptoms. Due to the acute onset of symptoms and imaging evidence, the patient underwent decompression and posterior stabilization. In the setting of a nonhealed AM Type III condylar fracture, it was presumed that there was instability of the CCJ, and therefore fusion and decompression of the hypoglossal nerve was indicated. The hypoglossal canal was adequately decompressed posteriorly allowing for simultaneous hypoglossal decompression and occiput to C1 fusion without the need for a second stage procedure.

We present a case of HNP resolution after surgical decompression and internal fixation following traumatic OCF. There are currently no well-established treatment guidelines. Most OCFs can be managed conservatively with collar or halo immobilization. Decisions for surgical intervention are based on stability and neurological compression with a correlative neurological examination. Our patient's HNP resolved after decompression, and she continues to remain asymptomatic. Evidence here demonstrates for the first time causality in hypoglossal neuropraxia and OCFs.

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Conflicts of interest

There are no conflicts of interest.

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