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Occipital condyle fracture—A rare but severe injury in cranial fracture patients

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ABSTRACT

We clarified occurrence, severity, and associated injuries of occipital condyle fractures (OCFs) in a cranial fracture population. Retrospective data of cranial fracture patients were analyzed. The outcome variable was presence of OCF in cranial fracture patients. Predictor variables were type of associated injury, Glasgow Coma Scale (GCS) value under 6, and death during hospital care. In addition, occurrence of OCF was assessed according to cranial fracture subtypes. Explanatory variables were age, sex, injury mechanism, involvement of alcohol, and high-energy injury. Treatment and outcome of OCFs were analyzed. Of 637 cranial fracture patients, 19 (3.0%) sustained an OCF, eight of whom had no other cranial fractures. In the multivariate adjusted model, increased risk for OCF was detected in patients with cervical injuries (OR 18.66, 95% CI 5.52, 63.12; $p < 0.001$) and facial fractures (OR 5.99, 95% CI 1.01, 35.45; $p = 0.049$). Patients with fractures not extending to the skull base were less likely to have OCF (OR 0.01, 95% CI 0.001, 0.25; $p = 0.004$), and fractures localized solely to the base of the skull offered a protective effect for OCF (OR 0.19, 95% CI 0.06, 0.58; $p = 0.003$). All OCFs were treated non-operatively with a cervical collar without complications. OCF patients typically sustain other severe injuries, particularly cervical injuries and facial fractures. Careful screening for associated injuries is therefore crucial when examining a patient with OCF. The classification scheme of Mueller et al. seems to be useful in guiding the treatment of OCFs, at least type 1 and 2 fractures.

1. Introduction

Occipital condyle fracture (OCF) is a relatively rare but potentially severe injury. The first description of OCF was published by Bell in 1817 on the basis of a postmortem examination (Bell, 1817). As computed tomography (CT) imaging has become more common, OCFs have been increasingly diagnosed, suggesting that their incidence has been underestimated. Many studies have now revealed that conventional radiography is not capable of detecting OCFs, whereas CT detects virtually all cases (Hanson et al., 2002; Aulino et al., 2005; Theodore et al., 2013). The incidence of OCFs is reported to be 0.1%–0.4% in level 1 trauma center patients (Hanson et al., 2002; Malham et al., 2009; Maserati et al., 2009; West et al., 2018). Postmortem CT imaging of fatal injury cases revealed that 22.6% of these patients had an OCF (Borowska-Solonynko et al., 2019). Most of these were caused by road traffic accidents (70%), and OCFs were present particularly in fatal high-energy injuries.

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Current knowledge of OCFs is based mainly on case reports and case series, and no consensus exists on how OCFs should be treated. Most OCFs are treated non-operatively, but the time and type of immobilization and the indications for operative treatment remain points of contention. The treatment options range from no immobilization to immobilization with a semi-rigid or rigid collar to halo fixation and surgical intervention with decompressive surgery and/or occipitocervical fusion. Many classifications have been developed in an attempt to guide the treatment of these fractures based on different features of OCFs, e.g. Anderson and Montesano's, Tuli's and Mueller's classifications (Anderson and Montesano, 1988; Tuli et al., 1997; Mueller et al. 2012).

Because of their relation to high-energy trauma, OCFs rarely occur as the only injury, and the mortality of OCF patients is reported to be as high as 4.3%–17.1% (Hanson et al., 2002; Aulino et al., 2005; Malham et al., 2009; Maserati et al., 2009; Mueller et al., 2012; West et al., 2018). This high mortality rate is explained by other injuries (Mueller et al. 2012). Cervical spine injuries are associated with 20%–59% of OCF patients (Hanson et al., 2002; Aulino et al., 2005; Malham et al., 2009; Maserati et al., 2009; West et al., 2018). Concomitant traumatic brain injuries (TBIs) have been reported in 30%–75% of OCF patients, and Hanson et al. noted that the main determinant of outcome of OCF patients was the presence and severity of concomitant TBI (Hanson et al., 2002; Aulino et al., 2005; Malham et al., 2009; Maserati et al., 2009; Krüger et al., 2013; West et al., 2018). OCFs have also been typically associated with lower cranial nerve palsies due to the proximity of the occipital condyles to the hypoglossal canal and the jugular foramen, which contain cranial nerves XII and IX-XI, respectively (Capuano et al., 2004).

The aim of the study was to compare patient and injury related variables between patients with and without OCF in cranial fracture population. We also present the treatment and outcome of OCF patients.

2. Material and Methods

2.1. Study design

The retrospective study was conducted to evaluate all patients with cranial fractures admitted to the tertiary trauma center of Helsinki University Hospital, Helsinki, Finland, between 2013 and 2018. The data of emergency patients with International Classification of Diseases Tenth Revision (ICD-10) codes for fracture of skull and facial bones (subsection S02) were retrieved from the electronic medical record system. Data were examined in detail by reviewing the patients' files, and detailed classifications for study variables were collected manually.

2.2. Inclusion criteria

All patients with radiologically confirmed cranial fracture were included in the study.

2.3. Study variables

The outcome variable was presence of OCF in cranial fracture patients.

Predictor variables included type of associated injury, Glasgow Coma Scale (GCS) value under 6, and death during hospital care. Recorded associated injuries were any cervical injury (i.e. cervical spine injury between levels C1 and C7), blunt cerebrovascular injury, TBI, thoracic injury (i.e. radiologically confirmed thoracic injury), and facial fracture.

Occurrence of OCF was assessed according to four cranial fracture subtypes: isolated OCFs, skull base fractures, other skull fractures, and combined skull fractures (i.e. combination of skull base and other skull fractures).

Explanatory variables were age, sex, injury mechanism, involvement of alcohol, and high-energy injury. Injury was classified as high energy in road- and traffic-related injuries and in falls from over 3 m (Evans et al., 2010).

Treatment and outcome of OCFs were analyzed further. Fractures were classified according to the classification of (Mueller et al. 2012).

2.4. Radiological evaluation

Maxillofacial surgeon J.S. with special expertise in head and neck traumatology reviewed all initial radiological reports and radiological images of skull base fracture patients, and confirmed the diagnosis of OCF. Classification of occipital condyle fractures was performed by orthopedic trauma surgeon T.B. and author H.L. Any inconsistencies were settled by consensus with other authors.

2.5. Statistical analysis

Fisher exact tests were performed for all categorical variables owing to the small number of patients with OCF. Absolute numbers and proportions for the categorical variables were also reported. The continuous variable of age was reported as median and interquartile range, as it was not normally distributed. Firth logistic regression analysis was also done for both univariate and multivariable analyses. Firth logistic regression can be used when there are small numbers in a dataset or in the outcome of interest, and it produces penalized log likelihood estimates by reducing the bias in the maximum log likelihood estimates. Variables retained in the multivariable model were based on a p-value of less than or equal to 0.1. The fit of the final model was tested using Firthfit and found to have a good fit. Estimates were reported as odds ratios (ORs), with statistical significance being indicated at 0.05 and reported alongside 95% confidence intervals (CIs). Data analysis was conducted using Stata version 16 (StataCorp, College Station, TX, USA).

2.6. Ethical considerations

The internal review board of the Head and Neck Center, Helsinki University Hospital (HUS/54/2019) approved the study protocol. Patient consent was not required because of the retrospective nature of the study. The guidelines of the Declaration of Helsinki were followed in this study.

3. Results

A total of 637 cranial fracture patients were included in the data analysis. Of these patients, 19 (3.0%) had sustained an OCF (Table 1). Of the 19 OCF patients, eight had fractures localized to the occipital condyle without additional cranial fractures (Table 2). None of the patients with cranial fractures solely in other parts of the skull had sustained an OCF ($p < 0.001$). Regarding the predictor variables, cervical spine injuries, thoracic injuries, and facial fractures all proved to significantly increase OCF risk. Mortality rate in patients with an OCF was three out of 19 (15.8%), whereas the corresponding rate in other cranial fracture patients was 10.0%.

In univariate analysis (Table 3), the highest risk factors associated with OCF were cervical injuries (OR 35.88, 95% CI 13.30, 96.75; $p < 0.001$), facial fractures (OR 14.69, 95% CI 2.76, 78.15; $p = 0.002$), and bicycle crash injuries (OR 5.66, 95% CI 2.12, 15.10; $p = 0.001$). Patients with cranial fractures located in other than the skull base

Table 1
Patients' descriptive statistics by presence of occipital condyle fracture (OCF).

Variable	Occipital condyle fracture - N (%)	No occipital condyle fracture - N (%)	Fisher's p-value
All patients	19 (2.98)	618 (97.02)	
Age, median (IQR)	53.9 (33.5, 66.2)	46.4 (24.9, 64.5)	0.480
Sex			
Male	14 (2.91)	467 (97.09)	0.791
Female	5 (3.21)	151 (96.79)	
Mechanism of injury			
Ground-level fall			
No	16 (3.58)	431 (96.42)	0.211
Yes	3 (1.58)	187 (98.42)	
Assault			
No	19 (3.20)	574 (96.80)	0.634
Yes	0 (0.00)	44 (100.00)	
Motor vehicle injury			
No	16 (2.93)	531 (97.07)	0.742
Yes	3 (3.33)	87 (96.67)	
Bicycle injury			
No	13 (2.23)	570 (97.77)	0.003
Yes	6 (11.11)	48 (88.89)	
Fall from height			
No	18 (3.49)	498 (96.51)	0.147
Yes	1 (0.83)	120 (99.17)	
Fall from stairs			
No	15 (2.60)	562 (97.40)	0.094
Yes	4 (6.67)	56 (93.33)	
Other/Unknown			
No	17 (3.04)	542 (96.96)	1.000
Yes	2 (2.56)	76 (97.44)	
High-energy injury			
No	14 (3.29)	411 (96.71)	0.626
Yes	5 (2.36)	207 (97.64)	
Alcohol involved			
No	12 (2.96)	394 (97.04)	1.000
Yes	7 (3.03)	224 (96.97)	

IQR, interquartile range.

were 96% less likely to be associated with OCF (OR 0.04, 95% CI 0.002, 0.67; $p = 0.025$), and patients with TBIs were 61% less likely to be associated with OCF (OR 0.39, 95% CI 0.16, 0.96; $p = 0.039$).

In the adjusted model (Table 4), patients with cervical injuries were at least 18 times more likely to have OCF (OR 18.66, 95% CI 5.52, 63.12; $p < 0.001$). Patients with facial fractures also had an increased risk of OCF (OR 5.99, 95% CI 1.01, 35.45; $p = 0.049$). On the other hand, patients with fractures not extending to the skull base were 99% less likely to have OCF (OR 0.01, 95% CI 0.001, 0.25; $p = 0.004$), and patients with fractures localized solely to the base of the skull were 81% less likely to have OCF (OR 0.19, 95% CI 0.06, 0.58; $p = 0.003$).

OCFs were classified according to the classification of Mueller et al. (Mueller et al. 2012). There were 17 type 1 and two type 2 OCFs. Our patient population did not include any type 3 fractures with atlanto-occipital dislocation (AOD). None of the patients required surgical stabilization. Of the 19 patients, 16 were treated with a cervical collar. Two patients died before deciding on the final treatment, and for one patient no specific treatment for OCF was found in the medical records. Treatment with cervical collar ranged from 6 weeks to 12 weeks, with 12 weeks being the most common length. Results of treatment were available for 11 patients, all of whom had no or mild symptoms (mild pain or stiffness in neck) at follow-up. Follow-up visits were organized until cervical collar treatment terminated (range 6–12 weeks, median 12 weeks). None of the patients presented with deficits of the lower cranial nerves (CN IX-XII). The classification, treatment, and outcome of OCFs are summarized in Table 5.

4. Discussion

During a 6-year period 19 OCFs were diagnosed in our tertiary trauma center. Helsinki University Hospital's trauma unit centralizes the treatment of severe blunt injuries, and it is one of the largest trauma centers in Northern Europe, with a catchment area of roughly 2 million people. Thus, incidence of OCFs in our area was approximately 0.16 per 100 000 persons per year. Byström et al. reported a fairly similar incidence, 0.4 per 100 000 persons per year, in 2017 (Byström et al., 2017).

Our results highlight the risk for a concomitant cervical injury in the OCF patient population. Slightly more than half of the patients (10 out of 19) in this study sustained cervical injuries, which is in accordance with previous results, as 20%–59% of OCFs have been reported to be associated with a concomitant cervical injury (Hanson et al., 2002; Aulino et al., 2005; Malham et al., 2009; Maserati et al., 2009; West et al., 2018). This finding is not unexpected, since occipital condyles are the parts of the skull that articulate with the cervical spine and are sometimes referred to as C0 vertebrae. Acting as an appendage of the cervical spine, it is plausible that the injury mechanisms and forces that cause cervical injuries might also result in OCF.

Previously, 30%–75% of OCFs have been estimated to be associated with TBIs (Hanson et al., 2002; Aulino et al., 2005; Malham

Table 2
Patients' associated injuries, cranial fracture type, and mortality by presence of occipital condyle fractures (OCF).

Variable	Occipital condyle fracture - N (%)	No occipital condyle fracture - N (%)	Fisher's p-value
Associated injuries			
Glasgow Coma Scale <6			
No	17 (3.03)	544 (96.97)	1.000
Yes	2 (2.63)	74 (97.37)	
Cervical injury			
No	9 (1.48)	600 (98.52)	<0.001
Yes	10 (35.71)	18 (64.29)	
Traumatic brain injury			
No	9 (5.29)	161 (94.71)	0.061
Yes	10 (2.14)	457 (97.86)	
Blunt cerebrovascular injury			
No	18 (2.90)	602 (97.10)	0.406
Yes	1 (5.88)	16 (94.12)	
Thoracic injury			
No	11 (2.14)	503 (97.86)	0.017
Yes	8 (6.50)	115 (93.50)	
Facial fracture			
No	1 (0.30)	336 (99.70)	<0.001
Yes	18 (6.00)	282 (94.00)	
Cranial fractures			
Isolated OCF			
No	11 (1.75)	618 (98.25)	<0.001
Yes	8 (100.00)	0 (0.00)	
Base of skull			
No	11 (3.42)	311 (96.58)	0.643
Yes	8 (2.54)	307 (97.46)	
Other part of skull			
No	19 (4.80)	377 (95.20)	<0.001
Yes	0 (0.00)	241 (100.00)	
Combined skull fracture			
No	16 (2.84)	548 (97.16)	0.470
Yes	3 (4.11)	70 (95.89)	
Death during hospital stay			
No	16 (2.80)	556 (97.20)	0.430
Yes	3 (4.62)	62 (95.38)	

Table 3
Univariate Firth logistic regression.

Variable	Unadjusted OR	95% CI	p-Value
Age	1.01	0.99, 1.04	0.150
Sex (reference: male)	1.17	0.43, 3.18	0.757
Mechanism of injury			
Ground-level fall	0.49	0.15, 1.57	0.228
Assault	0.33	0.02, 5.57	0.443
Motor vehicle injury	1.29	0.40, 4.17	0.673
Bicycle injury	5.66	2.12, 15.10	0.001
Fall from height	0.34	0.06, 1.79	0.201
Fall from stairs	2.89	0.98, 8.55	0.055
Other/Unknown	1.01	0.26, 3.89	0.985
High-energy injury	0.75	0.28, 2.04	0.575
Alcohol involved	1.05	0.42, 2.65	0.910
Associated injuries			
Glasgow Coma Scale <6	1.04	0.27, 4.02	0.950
Cervical injury	35.88	13.30, 96.75	<0.001
Traumatic brain injury	0.39	0.16, 0.96	0.039
Blunt cerebrovascular injury	2.96	0.52, 16.77	0.220
Thoracic injury	3.22	1.30, 8.00	0.012
Facial fracture	14.69	2.76, 78.15	0.002
Cranial fractures			
Base of skull	0.75	0.30, 1.84	0.529
Other part of skull	0.04	0.002, 0.67	0.025
Combined skull fracture	1.65	0.51, 5.37	0.405
Death during hospital stay	1.89	0.58, 6.16	0.292

CI, confidence interval; OR, odds ratio.

Table 4
Multivariable Firth logistic regression.

Variable	Adjusted OR[a]	95% CI[b]	p-value
Bicycle injury	3.35	0.88, 12.74	0.076
Fall from stairs	3.40	0.84, 13.82	0.087
Cervical injury	18.66	5.52, 63.12	<0.001
Facial fracture	5.99	1.01, 35.45	0.049
Other part of skull	0.01	0.001, 0.25	0.004
Base of skull	0.19	0.06, 0.58	0.003

CI, confidence interval; OR, odds ratio.

Table 5
Occipital condyle fracture (OCF) classification, treatment, and outcome.

Fracture type	Mueller 1	Mueller 2	Mueller 3
Number	17	2	0
Treatment, cervical collar	15	1	0
6 weeks	5	0	0
8 weeks	0	1	0
12 weeks	10	0	0
Treatment, surgical	0	0	0
Treatment, not available[a]	2	1	0
Outcome			
No symptoms	4	1	0
Mild pain or stiffness at follow-up	6	0	0
Moderate or severe symptoms	0	0	0
Died before discharge	2	1	0
Died after discharge, before follow-up	2	0	0
No follow-up information	3	0	0

[a] Specific treatment of OCF could not be found in medical records or the patient had died before specific treatment was decided.

et al., 2009; Maserati et al., 2009; Krüger et al., 2013; West et al., 2018). Unexpectedly, TBIs seemed to have had a protective effect regarding OCFs in our study population, as patients with TBIs were 61% less likely to sustain an OCF. This can partly be explained by the nature of the cranial fracture study population, in which TBI was common in general. Still, 10 out of 19 OCFs were associated with TBIs, emphasizing the potential severity of OCFs, as TBIs have been reported to be the major cause of in-hospital deaths and the main determinant of outcome among OCF patients (Hanson et al., 2002; Aulino et al., 2005; Maserati et al., 2009; Mueller et al., 2012; West et al., 2018). Three of our patients died during hospital stay. All three patients had serious TBIs, highlighting their influence on survival rate of OCF patients. For two of these patients, TBI was determined to be the cause of death.

Associations between facial fractures and cranial fractures have been established, but current knowledge concerning the relationship between OCFs and facial fractures is almost solely based on individual, descriptive case reports (Leventhal et al., 1992; Olsson and Kunz, 1994; Evans et al., 2010). Reich et al. emphasized that maxillofacial surgeons should be aware of a low but serious risk of cervical spine injuries in patients injured in craniofacial area (Reich et al., 2016). In that study focusing on cervical injuries in particular, six out of 3782 patients with craniomaxillofacial injuries had an injury of C0, i.e. occipital condyle. Due to potential severe complications of OCFs, evaluation of the risk of OCF in patients with facial fractures is necessary. In the present study, 18 of the 19 OCF patients had concomitant facial fractures. It is quite interesting that in eight of these patients, OCFs occurred without another type of cranial fracture. Thus, this association supports the notion that OCF is a specific entity with distinct characteristics not present in other skull fractures. Moreover, all of these eight patients with isolated OCFs and associated facial fractures sustained their injuries from low-energy mechanisms. Typically, the mechanisms related to facial fractures subject patients to sudden rotational movements or extension of the neck, and this energy transmission can target the occipital condyle. These findings bring novel insight into the relationship between OCFs and facial fractures. Thus, clinicians treating facial fractures should be aware of OCFs and adhere to routine comprehensive examinations and screening protocols.

Bicycle injuries accounted for a higher proportion of OCFs than other cranial fractures without OCFs. High-energy injuries and motor vehicle accidents represented a minority in our patients, in contrast to previous studies conducted on OCFs, which can be a result of our relatively small sample size. This especially highlights the fact that OCFs should not be forgotten when examining a low-energy trauma patient, even though OCFs are primarily considered to result from high-energy trauma. In our sample, OCF patients were predominantly males (14 out of 19), consistent with earlier studies (Tuli et al., 1997; Hanson et al., 2002; Capuano et al., 2004; Aulino et al., 2005; Caroli et al., 2005; Maserati et al., 2009; Mueller et al., 2012; Krüger et al., 2013; Byström et al., 2017; Burks et al., 2018; West et al., 2018; Borowska-Solonynko et al., 2019). Although ages of OCF patients in general were quite evenly distributed, OCFs were not found in children or adolescents. In addition, there seemed to be polarization between the sexes. None of the female patients were under 50 years of age, while there were nine male patients in that age group. Ten patients were over 50 years of age, five of each sex. This might be explained by the higher incidence of traumas in general among younger male patients (NTDB Annual Report, 2016).

Despite the low occurrence of OCFs, they are their own entity, with several previously presented treatment schemes and classifications. Mueller et al. presented a classification in 2011, which divided OCFs into three categories (Mueller et al., 2012). Type 1 includes unilateral OCFs without atlanto-occipital dislocation (AOD) (Fig. 1 and Fig. 2), and type 2 bilateral OCFs without AOD. Types 1 and 2 are considered stable and can be treated non-operatively with a 6-week cervical collar immobilization. Type 3 fractures are OCFs with AOD regardless of whether they are uni- or bilateral and are considered unstable, thus requiring surgical stabilization. The Mueller et al. classification does not take into account displaced OCFs, as these fractures might require surgical decompression and stabilization due to brain stem compression by the dislocated fragment. The Mueller et al. classification has previously been proved to be more useful in clinical decision-making than Anderson and Montesano's or Tuli's classifications, which were considered to have a more academic role (Byström et al., 2017). Additionally, differentiation between Anderson and Montesano types 1, 2, and 3 has been reported to be unreliable (Hanson et al., 2002; Maserati et al., 2009). Mueller et al. also found that all three types of OCFs in the Anderson and Montesano classification can be treated the same way, and they do not differ with regard to Injury Severity Score (ISS), radiological and clinical outcome, or mortality. Thus, we decided to classify our OCFs according to the Mueller et al. classification. Seventeen OCFs were type 1 and two were type 2. Our sample did not include any type 3 OCFs with AOD. All of our patients were treated non-operatively, as suggested by the Mueller et al. treatment scheme. At follow-up, no dislocation of fractures was detected. Some patients experienced mild stiffness and limited cervical movements when the cervical collar was removed, which are, however, to be expected after several weeks' immobilization. Based on our results, the Mueller et al. classification seems suitable for guiding the treatment of OCFs, at least type 1 and 2 fractures.

Fig. 1. A 29-year-old man fell with his bicycle, resulting in occipital condyle fracture on the left. The patient had also a non-dislocated upper orbital rim fracture, wide facial skin lacerations, and a tooth fracture.

Given the retrospective nature of our study, there are some limitations. Data were collected from medical records, and all clinical information on symptoms and findings was not available for every patient. There was no standardized treatment protocol, and every patient was individually treated based on the clinician's decision. The results of the treatment are based on medical records of the follow-up visit, which is not the optimal way of evaluating long-term outcome, as stiffness and pain of the neck are likely to

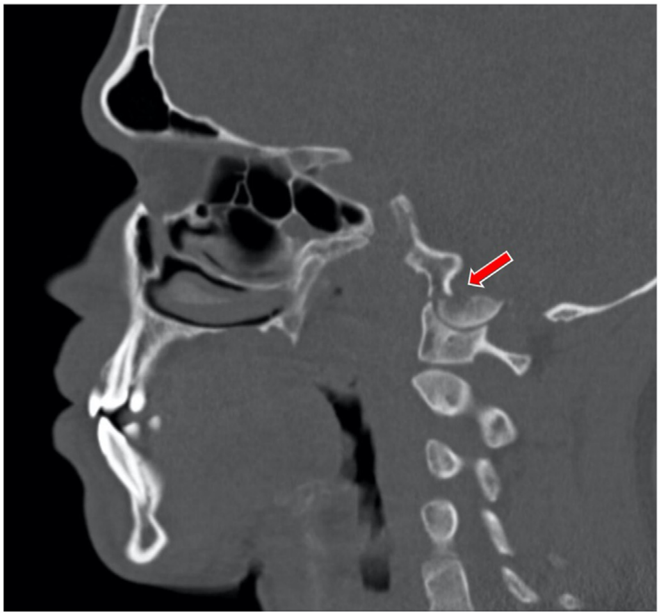

Fig. 2. The patient's left occipital condyle fracture, type 1 (Mueller et al. classification) was treated with a cervical collar for 12 weeks.

improve after removing the cervical collar, which enables evaluation of the actual long-term outcome. Additionally, there was no standardized evaluation method for the treatment result. Although our patient population included only 19 patients, our sample size was relatively good compared with those in other studies conducted on OCFs, due to the rare nature of this injury (Theodore et al., 2013). Also, as a strength, our study period was relatively long, and the CT images and medical records of all OCF patients of a large trauma center were retrospectively reviewed, thus representing well the OCF population of our hospital's catchment area during the 6-year study period.

5. Conclusion

OCF is a relatively rare injury, but it is associated with a high mortality rate due to the presence of related injuries. The classification scheme of Mueller et al. seems to be useful in guiding the treatment of OCFs. A significant proportion of OCFs are associated not only with cervical injuries, but also with facial fractures, and the possibility of OCF should be kept in mind in the presence of these injuries.

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CRediT author statement

Henri Lassila: Investigation, Data Curation, Writing - Original Draft, Writing - Review & Editing, Visualization.

Tero Puolakkainen: Conceptualization, Methodology, Investigation, Data Curation, Writing - Original Draft, Writing - Review & Editing.

Tuomas Brinck: Investigation, Writing - Original Draft, Writing - Review & Editing.

Michael Wilson: Formal analysis, Writing - Original Draft, Writing - Review & Editing.

Johanna Snäll: Conceptualization, Methodology, Investigation, Data Curation, Writing - Original Draft, Writing - Review & Editing, Supervision, Project administration.

Declaration of competing interest

The authors have no conflicts of interest to disclose.

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