Occipitocervical fusion – An epidemiological drift experienced in an Irish tertiary spinal referral center: Twenty-year follow-up study

ABSTRACT

Background: Occipitocervical disease is common in the elderly population, and is on the rise due to an increasingly aging population.

Methods: We retrospectively reviewed all patients who underwent occipitocervical fusion in our institution over a 20 year period (1996-2016) at a tertiary spinal referral centre. Patients were divided in 2 groups. Group A included all patient who underwent OCF in the first decade between 1996 and 2005. Group B was all patients who underwent OCF in the second decade between 2006 and 2016.

Results: A total of 23 patients underwent occipitocervical fusion between 1996 until 2016 at our institution. Instability secondary to Rheumatoid arthritis was the leading factor in group A, responsible for 43 percent of cases. In group B, trauma was the leading burden accounting for 44 percent of the cases. In contrast to Group A however, only 19 % of OCFs occurred secondary to RA in group B. Our fusion rate was 96 percent with a survival rate of 67 percent.

Conclusion: We noticed a clear epidemiological drift in the cervical spine pathologies requiring OCF during the first and second decade of study period with an increase in prevalence of pathological fractures secondary to metastatic disease. In addition, a drop in rheumatoid cervical disease requiring OCF has been noted.

Keywords: Occipitocervical disease, Occipitocervical fixation, occipitocervical fusion

INTRODUCTION

Occipitocervical disease is common in the elderly population. The Republic of Ireland is one of many countries projected to have an increasing aging population. At the 2006 census, in the Republic of Ireland, there were 468,000 people aged 65+ (11% of the population). By 2041, it is estimated, there will be 1.4 million aged 65 and over (22% of the population). This will certainly have an increased medical impact as the incidence of occipitocervical disease is bound to increase with such an aging population. This will ultimately lead to an increase in the number of occipitocervical fusion surgeries performed each year.[1]

The are many indications for occipitocervical fusion (OCF) but by far, the most common include occipitocervical instability secondary to rheumatoid arthritis, congenital, bony/ligamentous abnormalities, trauma, neoplasm, or degenerative bone disease. In addition, it is also indicated in certain cases of atlantoaxial instability (AAI).[2] Rheumatoid disease is the most commonly seen inflammatory arthropathy affecting the high cervical spine.[3] In patients with RA, the prevalence of cervical involvement has been reported to range from 43 to 86%.[4,5] One of the earliest indicators of cervical spine involvement in RA is neck pain, and as many as 40%–88% of RA patients report complaints of this symptom.[4] Chronic inflammation
of the cervical spine due to RA initially leads to a proliferation of fibrovascular tissue and pannus formation resulting in bony erosion and ligamentous laxity. This cascade can lead to cervical spinal instability in the form of AAI, cranial settling, and subaxial subluxation (SAS) or a combination of the three.[3]

Considering that the majority of patients with occipitocervical instability are elderly with multiple comorbidities, there have been multiple studies evaluating the most effective treatment option. Clarke et al. have demonstrated that complex upper cervical spine decompression and fixation can be safely and effectively accomplished.[6] Moreover, Matsunaga et al. retrospectively analyzed the results of 16 patients with a longer than 10-year follow-up with upper cervical lesions due to rheumatoid arthritis who underwent OCF, to determine the final outcome of such patients. It was concluded that surgical stabilization is the treatment of choice in patients with occipitocervical instability secondary to RA, as surgical stabilization is shown to provide more than a 2-fold increase in the 5-year survival rate along with pain reduction and improvement in myelopathy.[7] More recently Guppy et al. prospectively reviewed 47 patients above the age of 65. They concluded that the high mortality rates were due to the advanced ages with comorbidities and in some cases of metastasis to the spine due to systemic spread, but not due to the surgical procedure itself.[9]

Deutsch et al.[9] reviewed long-term outcomes of OCF in 58 patients who underwent OCF from 1997 to 2001. Out of 58 patients, 34 (58.6%) patients had OCF due to sequel of rheumatoid arthritis and basilar invagination which comprises the majority of cases. Remaining had surgery for trauma and metastatic tumors. Similarly, at our institution in the first decade from 1996 to 2006, the majority of OCF was performed due to rheumatoid arthritis; however, in the second decade from 2006 to 2016, the majority of OCF was performed due to trauma and metastatic tumors. The goal of our study is to assess the epidemiological change in the surgical indications of OCF in an Irish tertiary referral center over the past two decades.

METHODS

All patients who underwent OCF in our institution over a 20 year period, from January 1996 to January 2016 were included in the study. These patients were identified from the surgical theater database and a retrospective medical chart review was performed. Each patient’s medical record was reviewed for demographics, clinical presentation, surgical indication, ASA grade, follow-up period, and need for revision surgery. The operative notes were also reviewed to assess levels of cervical fixation and implants. In addition, the follow-up outpatient department notes along with postoperative X-rays/computed tomography (CT) scans were also reviewed to evaluate rates of fusion, complications, and follow-up period. Once the data was obtained, we divided the patients into two groups. Group A included all patients who underwent OCF in the first decade between 1996 and 2005. Group B was all patients who underwent OCF in the second decade between 2006 and 2016. Distributing these patients in the above two groups, allowed us to compare the surgical indications of patients that underwent OCF in these two decades. Following chart review, we classified the surgical indications into five groups: rheumatoid Disease, metastatic disease, trauma, degenerative, and congenital.

RESULTS

A total of 23 patients underwent OCF between 1996 and 2016 at our institution. About 57% were female (13 patients) and 43% were male (10 patients). The mean age was 68 at time of surgery (ranging between 39 and 90). Table 1 summaries the surgical indications for each group.

Group A (1996–2005)

A total of 7 patients underwent OCF between 1996 and 2005. Instability secondary to rheumatoid arthritis was the leading factor in this group, responsible for 43% of cases. However, trauma was responsible for only 14.2% of cases. No patient required revision surgery within this group.

Group B (2006–2016)

A total of 16 patients underwent OCF between 2006 and 2016. In this group, trauma was the leading burden accounting for 44% of the cases (7 patients). In contrast to Group A, however, only 19% of OCFs occurred secondary to RA in this group (3 patients). Pathological fractures were responsible for 25% of the cases (4 patients). One of these patients had a pathological fracture in C1, two patients had pathological fractures in C2, and one patient with both vertebrae involved. Multiple myeloma was the responsible primary in two of these patients, whereas breast cancer and nonsmall cell lung cancer were each accountable for one case.

Table 1: Demonstrates the surgical indications of occipitocervical fusion in Groups A and B

| Surgical indications | Group A (7 patients) | Group B (16 patients) |
|----------------------|----------------------|-----------------------|
| Rheumatoid disease   | 3                    | 3                     |
| Metastatic disease   | 2                    | 4                     |
| Trauma               | 1                    | 7                     |
| Degenerative         | 1                    | 1                     |
| Congenital (Os odontoideum) | 0            | 1                     |
| Total                | 7                    | 16                    |
One patient in this group required revision surgery following OCF indicated for a traumatic C2 fracture. He presented with ongoing pain and stiffness, and underwent revision OCF at 1 year following his initial surgery.

The overall fusion rate of all patients (Group A + B) was 96% (22/23).

The survival rate for the 23 patients included in our study was 67%.

This is an example of a 39-year-old woman presented with acute-onset severe right side occipital headache. The patient had no history of malignancy or trauma to her neck, before the presentation. No myelopathic signs on the clinical examination were noted. Further investigations, which included a CT of the patients C-spine, this revealed a pathological fracture of the lateral mass of C1 with the evidence of a lytic lesion [Figure 1]. Clinical examination detected a breast mass. The MRI of C-Spine revealed a significant lesion of the lateral mass of C1 [Figure 2], with multiple other spinal metastases evident throughout her spine. Based on the Spinal Instability Neoplastic Score, the patient was determined to have a score of eight, based on the location of lesion and presence of pain. With this SIN score, this was classified as a potentially unstable cervical spine. This patient underwent occipital cervical fusion to stabilize the lesion. Figure 3 shows postoperative anterioposterior, lateral, and open mouth peg views of the cervical spine.

DISCUSSION

The management of pathological conditions involving the craniovertebral junction is known to be challenging due to the complex anatomic and biomechanical characteristics of this region. Thankfully, however, OCF surgery has significantly evolved over the past few years. The development of modern screw-based constructs has revolutionized the outcomes of OCF. They have been shown to provide better stability and a higher rate of fusion than previously used constructs. In fact, Deutsch et al. reported a 94% fusion rate in 58 patients who underwent OCF with a variety of constructs, most of which were screw based. Clarke et al. further evaluated the safety of modern techniques of OCF in the elderly by performing a retrospective review of twenty consecutive patients over the age of 65 who underwent OCF between 1995 and 2005. They concluded that although many elderly patients have multiple medical comorbidities complicating their care, with careful medical management, and control of perioperative complications, these patients can make a significant neurological recovery with OCF.

Apart from significant changes in the technique of OCF over the past decade, there has been a substantial change in the indication for surgery. For a long period of time, rheumatoid arthritis had been considered to be a major indication of OCF. In fact, this correlation has been first recognized in 1890 by Garrod after he reported 178 patients with cervical spine involvement in a series of 500 patients with RA. However, due to the therapeutic advances and the widespread use of disease-modifying antirheumatic drugs (DMARDS) and TNF-α inhibitors that significantly slow the progression of the disease, the true prevalence of cervical spine abnormalities in rheumatoid arthritis is not known. Even though cervical spine disease is considered to be common in patients with rheumatoid arthritis, there is no doubt that the prevalence has decreased with contemporary medical management using DMARDS and TNF-inhibitors. Most recently, Campo et al. reviewed a
case series of 120 patients who underwent OCF between 2004 and 2013. In their case series, trauma was the most common etiology of occipitocervical instability affecting 47%, with only 10.8% of patients undergoing OCF secondary to rheumatoid arthritis.\textsuperscript{[16]} Moreover, Zhang et al. performed a meta-analysis to analyze the trend of prevalence and progression of anterior atlantoaxial subluxations (aAASs), vertical subluxations, SASs, and associated cervical myelopathy in RA over the past 50 years. The prevalence of aAAS was 36% in the 1970s and earlier, 36% in the 1980s, 32% in the 1990s, and 24% in the 2000s \( (P = 0.04) \). VS and SAS had comparable prevalence rates.\textsuperscript{[17]} This clearly demonstrates a dramatic gradual decrease in aAASs over the years following the introduction of DMARDs. Therefore, it is not surprising that our results have shown a similar trend with a decrease in number of OCF surgeries being performed secondary to RA.

Another aspect that has been noted is the increase in the prevalence of pathological fractures secondary metastatic disease requiring OCF. It is reported that the metastatic spine tumors occur in 5%–10% of all cancer patients, with cervical spine involvement being relatively uncommon accounting for <10% of all spinal metastasis.\textsuperscript{[18]} However, the incidence of symptomatic spinal metastases has increased given the cancer patients longer life expectancy.

**CONCLUSION**

We noticed a clear epidemiological drift in the cervical spine pathologies requiring OCF during the first and second decade of the study period. This change in the surgical indication is attributed to early introduction of DMARDs in rheumatoid arthritis and also longer survival of cancer patients even with multiple metastatic lesions in the spine. At 20 years of follow-up survival rate was 67%. Only one patient needed revision surgery.

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**Conflicts of interest**

There are no conflicts of interest.

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