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Multidisciplinary approach to the craniovertebral junction. Historical insights, current and future perspectives in the neurosurgical and otorhinolaryngological alliance

Approccio multidisciplinare alla giunzione cranio vertebrale. Cenni storici, attuali orientamenti e prospettive future nell'alleanza tra neurochirurgia ed otorinolaringoiatria

Massimiliano Visocchi[1,2,4], Francesco Signorelli[1], Claudio Parrilla[3], Gaetano Paludetti[3,4], Mario Rigante[3,4]

[1] Institute of Neurosurgery, Fondazione Policlinico Universitario A. Gemelli IRCCS, Rome, Italy; [2] Institute of Neurosurgery, Catholic University School of Medicine, Rome, Italy; [3] Otorhinolaryngology, Head and Neck Surgery, "A. Gemelli" Hospital Foundation IRCCS, Catholic University of the Sacred Heart, Rome, Italy; [4] Craniovertebral Junction Operative Unit, Master II Degree, Cadaver Lab and Research Center on Craniocervical Junction Surgery, Catholic University School of Medicine, Rome, Italy

SUMMARY

Historically considered as a nobody's land, craniovertebral junction (CVJ) surgery or specialty recently gained high consideration as symbol of challenging surgery as well as selective top level qualifying surgery. The alliance between Neurosurgeons and Otorhinolaringologists has become stronger in the time. CVJ has unique anatomical bone and neurovascular structures architecture. It not only separates from the subaxial cervical spine but it also provides a special cranial flexion, extension, and axial rotation pattern. Stability is provided by a complex combination of osseous and ligamentous supports which allows a large degree of motion. The perfect knowledge of CVJ anatomy and physiology allows to better understand surgical procedures of the occiput, atlas and axis and the specific diseases that affect the region. Although many years passed since the beginning of this pioneering surgery, managing lesions situated in the anterior aspect of the CVJ still remains a challenging neurosurgical problem. Many studies are available in the literature so far aiming to examine the microsurgical anatomy of both the anterior and posterior extradural and intradural aspects of the CVJ as well as the differences in all the possible surgical exposures obtained by 360° approach philosophy. Herein we provide a short but quite complete at glance tour across the personal experience and publications and the more recent literature available in order to highlight where this alliance between Neurosurgeon and Otorhinolaringologist is mandatory, strongly advisable or unnecessary.

KEY WORDS: instrumentation and fusion, endoscopy, transnasal approach, transoral approach, extreme lateral approach, far lateral approach, submandibular retropharyngeal approach, craniovertebral junction

RIASSUNTO

Storicamente considerata "terra di nessuno", la regione della giunzione cranio-vertebrale, così come la chirurgia di questa sede, hanno guadagnato altissima considerazione negli anni recenti per la complessità anatomica, funzionale e tecnica. La gestione multidisciplinare e l'alleanza tra neurochirurghi e otorinolaringoiatri anche in questo ambito è diventata sempre più forte negli anni. La giunzione cranio-vertebrale ha una architettura anatomica ossea, neurovascolare e muscolare unica e complessa in quanto non solo separa e congiunge il cranio con il rachide cervicale, ma presenta pattern speciali di flessione, estensione e rotazione assiale. La stabilità è garantita da una complessa combinazione di supporti ossei e ligamentosi, che consentono ampi gradi di motilità. La conoscenza dell'anatomia e della fisiologia della giunzione cranio-vertebrale consente di comprendere meglio le procedure chirurgiche e le patologie specifiche che interessano questa regione anatomica. Sebbene siano passati anni dell'inizio della chirurgia pioneristica di questa regione, le lesioni situate nella porzione anteriore della giunzione cranio-vertebrale riman-

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Correspondence
Francesco Signorelli
Institute of Neurosurgery, Fondazione Policlinico Universitario A. Gemelli IRCCS,
largo F. Vito 1, 00168 Rome, Italy
E-mail: francesco.signorelli@policlinicogemelli.it

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gono ancora una stimolante sfida per il neurochirurgo. Molti studi sono presenti in letteratura con l'intento di esaminare l'anatomia microchirurgica delle porzioni anteriori, posteriori, extra e intradurali della giunzione cranio-vertebrale, così come le differenze e tutte le possibili vie di approccio a 360° per esporre al meglio e trattare patologie di questa regione. In questa revisione verrà effettuata una disamina sullo stato dell'arte in tale chirurgia, partendo dall'esperienza personale, dalle pubblicazioni e dalla letteratura più recente, al fine di sottolineare quando la collaborazione multidisciplinare sia fondamentale, altamente consigliata o non strettamente necessaria nella gestione delle patologie della regione cranio-vertebrale.

PAROLE CHIAVE: *fusione e procedure strumentate, endoscopia, approccio endoscopic transanasali, approcci transorali, approcci laterali al basicranio, approccio sottomandibolare retrofaringeo, giunzione cranio-vertebrale*

Introduction

Despite the continuous evolution and refinements of operating techniques, the disposability of dedicated surgical instruments along with the growing awareness and experience of the dedicated surgeons, treatment of craniovertebral junction (CVJ) pathologies still is a complex challenge. The tricky combination of bony, muscular and neurovascular vital structures crowded in a deep and narrow space makes surgical approaches to the CVJ hard and risky. Depending on the location of the lesion, surgical approaches have traditionally been directed toward ventral, dorsal and lateral aspect of the cervico-medullary junction. The anterior aspect of CVJ can be approached by the transoral approach (TOA), simple or extended, the endoscopic endonasal approach (EEA), introduced by Kassam [1], and the submandibular approach (SMA), i.e. retropharyngeal approach, which is indicated only in selected cases.

Posterior suboccipital approach (SOA) intra-extradural approaches along with instrumentation procedures has been traditionally considered for inferior craniectomy with or without C1-C2 laminectomy for CVJ lesions. Through the same route it is possible to perform C0-C1-C2 instrumentation procedures with titanium cables, wires, screws and rods in order to fix and stabilize the CVJ.

Intradural lesions located at the ventrolateral aspect of CVJ can be approached by means of a postero-lateral or far lateral approach (FLA), an extension of the suboccipital approach with removal of a variable amount of occipital bone. Extradural lesions of the same region may require an antero-lateral or extreme lateral approach (ELA), which allows a better control of the entire length of the vertebral artery (VA), the jugular foramen, the lowest cranial nerves, and the jugular-sigmoid complex. Finally, the posterior midline approach is the most popular in the neurosurgical culture both for extra and intradural surgical control of the CVJ and mainly for instrumentation and fusion techniques.

Moving from a comparative analysis of the CVJ approaches, and in the wake of our surgical experience [2-7] consisting of more than 40 anterior surgical procedures including TOA and EEA, more than ten comprising ELA, FLA, SMA and more than hundred posterior instrumentation and fusion procedures, we herein outline the experience matured in our department including an equipped Cranio-Vertebral Junction Laboratory for anatomical dissection [8-10], a II Degree Master Course on Surgical Approaches on CVJ and a University Research Center on CVJ, all mastered and directed by the Senior Authors (MV and GP) along with the Junior Authors (MR and FS) and referring to the Surgical Department / Pole of Medical Interest of our Catholic University of Rome Medical School.

In this review we will try to identify and objectivate the coworking potential of Neurosurgeons and Othorhinolaringologists in the common CVJ surgery field of interest.

Where alliance between neurosurgeons and otorhinolaringologists is mandatory?

Submandibular anterior Approach (SMA)

Terms like anterolateral [11], submandibular [12], anterior high cervical [13], and retropharyngeal pre-vascular [14] have been used to describe a surgical approach between carotid sheath laterally and pharyngeal constrictor muscles medially to high cervical spine. Cloward [15] and Robinson and Smith [16] are generally acknowledged as establishing the anterior approach to the cervical spine for the management of disk herniation. McAfee et al. [14] described the retropharyngeal pre-vascular approach using the same fascial plane described by Southwick and Robinson [17]. Submandibular retropharyngeal approach provides a direct, perpendicular trajectory to the C2-3 interspace through a "natural" corridor above the superior laryngeal nerve (SLN) and below the hypoglossal nerve. The approach requires a very little retraction and, comparing to other approaches (especially ELA) is associated with a lower risk of hypoglossal, glossopharyngeal and superior laryngeal nerves injury. These risks can be further limited using an endoscope-assisted retropharyngeal approach, mainly indicated for lesions involving the clivus. Nevertheless, care must be taken when using the approach in the setting of prior neck dissection. On the other hand, this route can be burdened by some complications as respiratory dysfunctions; pharyngeal fistula; transient hoarseness and dysphagia; dural leakage; hypoglossal and facial nerves paresis and salivary fistula.

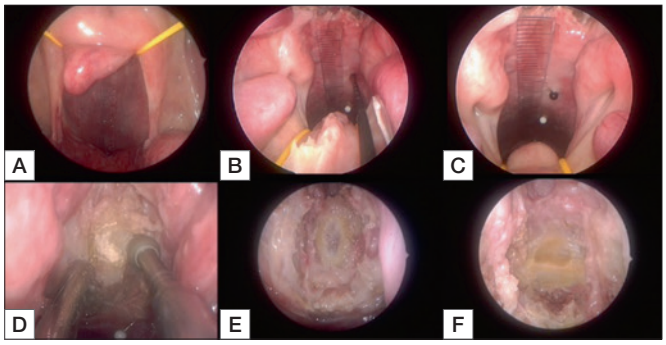

Figure 1. Anatomical studies comparing the exposure of transoral (**A**) and endoscopic transnasal approach (**B, C**) followed by exposure of the anterior arch of C1 (**D**), odontoid (**E**) and its removal (**F**) through a combined transoral transnasal approach.

This surgical field appear more consistent with the oncologic Otorhinolaringological background up to the anterior profile of C1 and C2.

Where alliance between neurosurgeons and otorhinolaringologists is strongly advisable?

TOA and EEA

TOA still represents the "gold standard" for the surgical treatment of several conditions resulting in anterior CVJ compression and myelopathy [18]. Refinements of the approach have been introduced during the late 1970s by Menezes who outlined several issues that now represent pivotal steps of the approach [19]. Nevertheless, some concerns, such as the need of temporary tracheostomy and postoperative nasogastric tube [20], soft palate morbility, overall led in 2005 to the introduction by Kassam et al. [1] of the EEA (Fig. 1).

EEA

Although this approach, conceived in order to overcome these surgical complications, rapidly gained wide attention, a clear predominance over the TOA in the treatment of CVJ pathologies, is still matter of discussion. In recent years, several papers have reported anatomical studies and surgical experiences in EEA to target different areas of the midline skull base, including the CVJ [20-28]. Starting from these preliminary experiences, further anatomical studies defined the theoretical (radiological) and practical (surgical) craniocaudal limits of the endonasal route (Fig. 2) [29-31]. Our group, on the basis of the clinical experience gained after 30 anterior procedures, both transoral and transnasal, did the same for the transoral approach [32,33] and compared the reliability of the radiological and surgical lines of the two different approaches. Very recently, a cadaveric study tried to define,

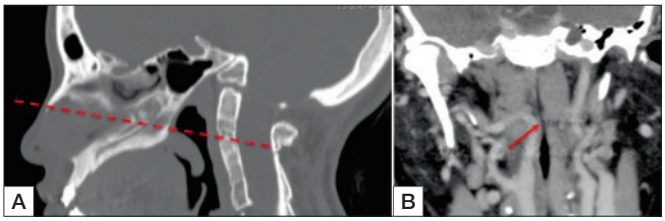

Figure 2. Importance of accurate preoperative radiological evaluation in order to choose the best corridor of approach. (**A**) CT scan of children with impression basilaris in which we preferred a transnasal corridor. (**B**) Angio CT showing an internal carotid kinking in the pharyngeal wall that exclude the anterior route to the CVJ.

with the aid of Neuronavigation (Fig. 3), the upper and lower limits of the endoscopic transoral approach [34].

This approach appears more consistent with the global rhinological endoscopic experience of the Othorhinolaringologist up to C1-C2.

TOA is a ventrally directed approach from the inferior third of clivus to C2-C3 interspace. It allows the shortest, wider and most direct access to the CVJ, among the other approaches to the CVJ [35]. Extensions of the approach, sometimes necessary to expose more rostrally located pathologies, carry the risk of numerous permanent comorbidities expecially on the soft palate and the need for temporary tracheostomy and nasogastric feeding tube [20]. The need to overcome the impact and significance of these comorbidities has led to the development of potentially less invasive techniques, such as the EEA. As widely demonstrated by numerous comparative anatomic and clinical studies, the

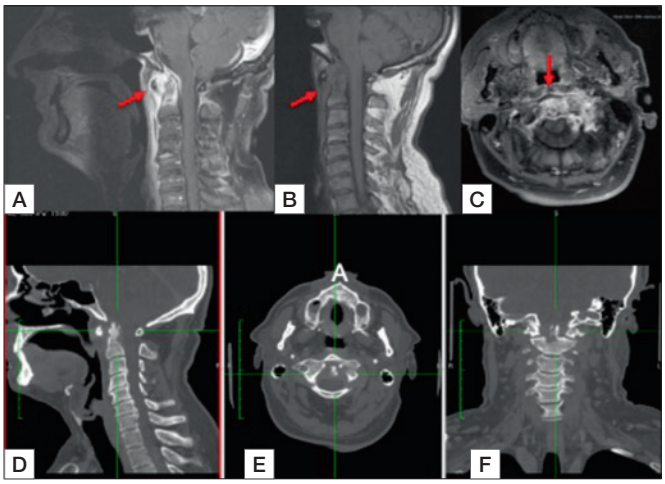

Figure 3. Use of navigation system to perform a biopsy of a lesion (arrow) of the odontoid on sagittal and axial MRI (**A, B, C**). Intraoperative view with CT scan (**D, E, F**) The cross-air revealed a correct target reached through a minimally invasive EEA. The biopsy revealed a localization of myeloma.

endoscope provides also an improved rostral exposure, brighter illumination and closer visualization of the surgical target [35] and can be also used during a TOA, as a valid complement tool in a combined procedure. Nevertheless, a recent systematic review and meta-analysis [37], while demonstrating a statistically significant increased risk of postoperative tracheostomy after TOA comparing with EEA, showed a slight, although not statistically significant, tendency toward a morbidity/mortality prevalence of EEA on TOA (Fig. 4).

In order to clearly define the limits of the TOA, our research group devised a radiologic "theoretical" line, the Palatine Inferior dental Arch line (PIA), as a reliable predictor of the maximal superior extension of the transoral approach and then compared the reliability of the radiological and surgical lines of the two different approaches [33]. Very recently, a cadaveric study tried to define, with the aid of Neuronavigation, the upper and lower limits of the endoscopic TOA [34]. Starting from our previous experimental volumetric studies [32,33] and other recent contributions, we tried to experimentally exploit the accuracy provided by Neuronavigation, to further compare operative sagittal and axial extensions of the transnasal and transoral corridors. Our observations were consistent with a relevant advantage of TOA over EEA in all the specimens. According to other clinical and experimental studies reported in literature, we found several advantages of TOA over EEA: wide working area in terms of both craniocaudal and lateral extension, a more familial anatomy for neurosurgeons, a safer top-down drilling of the clivus and odontoid with a better

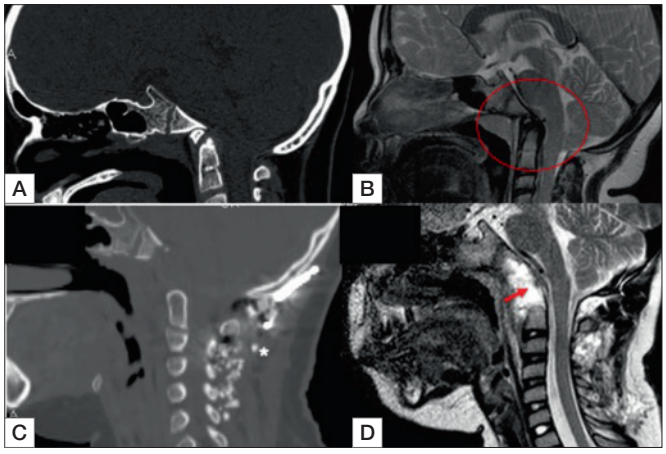

Figure 4. Axial CT scan (**A**) and T2 weighted MRI (**B**) of platybasia and impressio basilaris with bulbo-medullar compression (rounded area) treated through a pure transnasal endoscopic approach. In the inferior line post-operative CT scan (**C**) and MRI (**D**) showing a decompression of the bulbopontine (arrow) angle and the posterior stabilization (*) the absence of tracheostomy can be also observed.

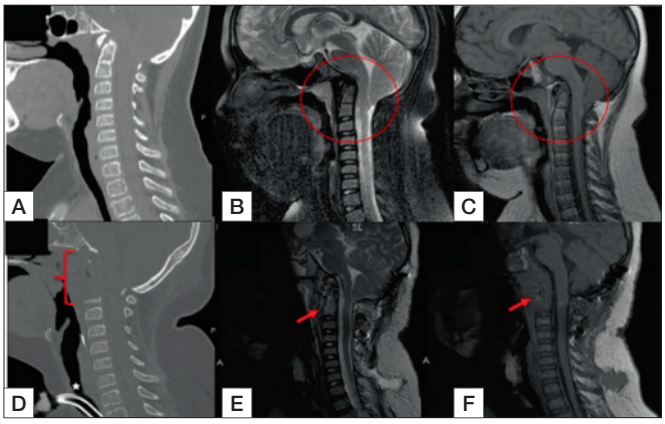

Figure 5. CT scan (**A**) and T2 (**B**) and T1(**C**) weighted MRI of a case of impressio basilaris and platybasia + bulbo-medullar compression (rounded area) This lesion was treated with transoral approach that allowed a wide exposure and resection from clivus to C1(⦚) and decompression (arrow) as showed in postoperative CT (**D**) and T2 (**E**) and T1(**F**) MRI, in which you can observe the presence of tracheostomy (*).

detachment of the ligaments (Fig. 5). On the other hand, excluding some well-known disadvantages and predictable complications appreciable only in clinical setting, such as working in a contaminated field, CSF leak management, the airway swelling, the upper airway obstruction and the velopharyngeal insufficiency, our study confirms the relevance of fixed obstacles to the required retraction as the tongue and the teeth.

The management of TOA requires the role of the Otorhinolaryngologist for performing tracheostomy, cooperate in the surgical exposure and final reconstruction of the pharyngeal opening.

ELA

Starting from the 1970s, many surgeons developed and introduced new skull base approaches to the lesions of the anterolateral CVJ introducing several variations and modifications. Hammon in 1972 and thereafter Heros in 1986 described a true lateral suboccipital approach for vertebral and vertebrobasilar aneurysms [38,39]. Heros described the combination of a lateral suboccipital craniotomy, C1 laminectomy and drilling of the occipital condyle (OC). George described a VA medial mobilization from C2 to its dural entrance point, with ligation of the sigmoid sinus and without condyle drilling. Spetzler, Bertalanffy, and Seeger mobilized the VA from C1 to dural entrance point, by drilling C1 facet, posterior C1 arch and posterior lateral third of the OC [39-43]. In recent years, extensive use of tools like neuroendoscope and neuronavigation, greatly implemented safety and efficacy of this and other skull base approaches, as demonstrated by several cadaveric studies [44-46]. ELA is

a direct lateral approach to the deep anterior portion of the SCM, behind the internal jugular vein and anterior to the VA. It is generally considered a more aggressive extension of far lateral approach. This term comes from 1990 when Sen and Sekhar described an alternative way to deal with meningiomas and schwannomas located anteriorly at the CVJ [47]. The rationale behind this procedure is to allow gross total resection of lesions with significant lateral extensions that would be otherwise inaccessible via anterior or classic FLA. ELA involves a greater extent of bony removal, skeletization of the jugular bulb along with the sigmoid sinus (in the transjugular variant), and more often VA transposition. These technical nuances overall widen the surgical corridor, but inherently are associated with a higher rate of morbidity and mortality [48,49].

ELA provides good access to the bone and extradural anterior and lateral space. It can be easily extended caudally to the cervical spine and it offers simultaneous control of the VA, cervical segment of the ICA, the lower cranial nerves, and the sigmoid-jugular complex [50].

In ELA, muscles are detached from their insertion on the transverse process of atlas. Great attention should be paid to avoid damage of VA, internal jugular vein, and spinal nerves, which are under these muscles. The key point for dissection and control of the VA is to preserve the periosteal sheath surrounding it. Our study further confirms that ELA allows exposure of the whole odontoid process, the inferior clivus, and the medial surface of the contralateral atlanto-occipital joint.

In this surgery the more confident knowledge of Otorhinolaryngologists of the superficial and middle and deep plane layers of the neck make this alliance absolutely advisable.

Where alliance between neurosurgeons and otorhinolaringologists is unnecessary?

Transcervical Anterior Approach (TCA)

Wolinsky described an endoscopic transcervical approach in order to perform odontoidectomy without traversing the oral cavity [51]. A recent cadaveric study exploited the feasibility of an endoscope-assisted retropharyngeal approach to the CVJ and clivus following submandibular gland resection [52].

The knowledge of the Neurosurgeons of this region gained by cervical spine surgery along with the skill obtained in spine traumatology aimed to screwing the odontoid fractures with biplanar fluoroscopy, make him confident and no surgical alliance seems to be required for this infrequent surgery.

FLA nowadays represents a mainstay for the surgical treatment of intradural pathologies at the ventral CVJ. Since the first description of Heros and George [53], extensive discussion and modifications of this approach have been reported in the literature. Several cadaver studies have demonstrated the use and benefits of the endoscope in the FLA. A study [54] has divided the surgical corridors for inserting the endoscope into upper, middle and lower. The cranial nerves VII and VIII, IX and X, and XII are respectively roof and floor of the three corridors and provide access and observation of the aspects of brainstem and posterior circulation by means of 0° lens (upper and middle corridor) and 30° lens (inferior corridor). Another cadaver study compared 3D endoscopic and microscopic vision in FLA after partial condilectomy and resection of jugular tubercle. The study concluded that the 3D endoscopic probe is too large and the surgical maneuverability is significantly hampered. Several authors have stated similar benefits of endoscope use in clinical series. These studies report a significant benefit in the endoscope's ability to identify any tumor adherent to brainstem or clivus amenable to resection [55].

For this approach the Neurosurgeon appears to be quite confident, since it can be considered an extension of the classic well known PIFP but in park bench position.

SOA Occipitocervical fusion (OCF) as well as C1-C2 is indicated for instability at the CVJ. Numerous surgical techniques, which evolved over 90 years, as well as unique anatomic and kinematic relationships of this region present a challenge to the neurosurgeon. The current standard involves internal rigid fixation by polyaxial screws in cervical spine, contoured rods and, eventually, occipital plate. Such approach precludes the need of postoperative external stabilization, lesser number of involved spinal segments, and provides 95-100% fusion rates. New surgical techniques such as occipital condyle screw or transarticular occipito-condylar screws address limitations of occipital fixation such as variable lateral occipital bone thickness and dural sinus anatomy. As the C0-C1-C2 complex is the most mobile portion of the cervical spine (40% of flexion-extension, 60% of rotation and 10% of lateral bending) stabilization leads to substantial reduction of neck movements. Preoperative assessment of vertebral artery anatomical variations and feasibility of screw insertion as well as visualization with intraoperative fluoroscopy are necessary. Placement of structural and supplemental bone graft around the decorticated bony elements is an essential step of every OCF procedure as the ultimate goal of stabilization with implants is to provide immobilization until bony fusion can develop.

This historical neurosurgical approach makes the Neurosurgeon absolutely confident, since it is required for conventional posterior cranial lesions approaches.

Future perspectives

In recent years, the surgical armamentarium has been enriched with *high-definition 4 K endoscope* [56] *as well as exoscope* [57] *systems*, which potentially provide a wide viewing angle as well as high-resolution image quality available with an endoscope with an optic resolution power equal or superior to the conventional Operating Microscope (OM) [57]. In particular, the exoscope is a new surgical tool recently conceived in order to overcome some limitations of OM and endoscope. Limitations of the first are mainly ergonomics: the size and weight, the ocular-dependent visualization, the continuous need of refocus because of the short field depth at high magnification and of continuously readjusting the OM and the body position in order to preserve a perfect stereoscopic picture. Limitations of the endoscope include a short focal distance and a limited field of view that requires an endoscope placement in the surgical field with the shaft reducing the available working space. Overall, these limitations are even more evident in complex and narrow anatomical corridors as those of the CVJ. Besides to the classic neuronavigation with preoperative neuroradiological assessment it's worth mentioning also *OArm neuronavigation and intraoperatory System.* Intraoperative imaging represents another important upgrade in neurosurgery.

For spinal surgery in particular, the introduction of the OArm system has made it possible to implement the safety of instrumentation procedures on the one hand, allowing much more accurate intraoperative neuronavigation than traditional techniques; secondly the setting with intraoperative imaging allows a real-time verification of the effectiveness of the procedure, such as in cases of medullary decompression or the correct positioning of arthrodesis systems [58].

OArm acquisition, comparing to fluoroscopy, not only should have the obvious advantage of a better definition with a resulting easier screws insertion, but, for sure, it permits an intraoperative direct and indirect assessment of bony and legamentous CVJ anterior decompression. In two of five cases, after OArm acquisition the cranio-caudal decompression was augmented because it proved to be suboptimal in an absolutely reliable and anatomically detailed way. Otherwise in our previous experience concerning fluoroscopic monitoring of TOA, the use of Iopamire, as contrast filler of the surgical cavity, allowed in a quite fair way to, indirectly, evaluate possible residual compression at the CVJ. Otherwise, it does not provide a real time visualization.

Finally, the possibility to convert the intraoperative neuronavigated 3D modality into 2 D real time OArm monitoring is very unconfortable due to the poor volume space available for the surgeon (also in the presence of EX) and the need of complex, time consuming and uneffective surgical manouvres required.

The spreading diffusion of such technologies seems to belong to the personal and institutional skill of both Neurosurgeons and Otorhinolaryngologists, always more devoted to share common objectives, operative tools for a common clinical and experimental final strategy.

Conclusions

The present paper confirms the irreplaceable role of interdisciplinary coworking in order to improve the difficult knowledge of the CVJ. Anatomical dissections in the training of surgeons, especially when approaching an anatomical region among the most complex such as the CVJ, is possible only with sharing experience and traditions and it is of paramount importance when dealing with this region. Accurate and multidisciplinary preoperative evaluation of the best corridor of approach, taking care also of all the possible intra, perioperative and postoperative problems are nowadays the mainstays for the best treatment of the patients affected of pathologies of CVJ.

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