

OS ODONTOIDEUM – CASE REPORT

OS ODONTOIDEUM – RELATO DE CASO

OS ODONTOIDEUM – REPORTE DE CASO

ANDRÉ RODRIGUES PINHO¹, VITORINO VELUDO MOUTINHO¹, NUNO PAULO ALEGRETE SILVA¹, ANTÓNIO FRANCISCO MARTINGO SERDOURA¹, JOANA MANUEL FERREIRA FREITAS¹, RUI ALEXANDRE PEIXOTO PINTO¹, ABEL VITORINO TRIGO CABRAL²

ABSTRACT

Objective: To report the difficulties in managing a case of os odontoideum. **Methods:** Female patient, 12 years old who developed a quadriparesis after minor cervical trauma in October 2005. In the emergency department a congenital cervical anomaly was identified. The patient was placed in a Stryker® frame and, few days later, in a halo bracing. After 3 months, an infection around the pins emerged and the halo vest had to be removed. A severe C1-2 instability persisted and a C1-C2 Gallie procedure was attempted. In the following weeks the bone disappeared and another procedure was attempted in June 2006 - C1 laminectomy and occiput-C3 fusion. In the following months the neurological status of the patient improved and a complete mass of occiput-C3 fusion was observed. **Results:** We choose a posterior cervical arthrodesis of C1-C2 using the Gallie technique. Since the condition was not resolved we performed a second surgery, C1 laminectomy (determined by SAC of 8, 3 mm in MRI) followed by posterior occiput-C3 fusion. In our case, until now, there is no evidence of axial decompensation, but a more prolonged follow-up is needed. **Conclusions:** The treatment of os odontoideum has many considerations but the essential that in the presence of instability and neurological deficit a solid fusion is achieved. In case of failure of posterior atlantoaxial wiring, the occiput-C2 or C3 fusion with rods seems to be an excellent option with a high rate of success, avoiding the need for additional support.

Keywords: Odontoid process/surgery; Odontoid process/radiography; Cervical vertebrae; Spinal injuries.

RESUMO

Objetivo: Relatar as dificuldades no tratamento de um caso de os odontoideum. **Métodos:** Paciente do sexo feminino com 12 anos de idade que desenvolveu tetraparesia após trauma cervical discreto em outubro de 2005. No departamento de emergência, constatou-se uma anomalia cervical congênita. A paciente foi colocada em cama Stryker® e alguns dias depois, em colete Halo Vest. Depois de 3 meses, ocorreu infecção em torno dos pinos e foi preciso remover o colete. Houve persistência de instabilidade C1-C2 grave, tentando-se a resolução com a técnica de Gallie C1-C2. Nas semanas seguintes, o enxerto desapareceu e outro procedimento foi tentado em junho de 2006 – laminectomia de C1 e fusão occipital-C3. Nos meses posteriores, o estado neurológico da paciente melhorou e verificou-se massa de fusão completa occipital-C3. **Resultados:** Escolhemos a artrodese cervical posterior C1-C2 usando a técnica de Gallie. Uma vez que o problema não se resolveu, realizamos uma segunda cirurgia, laminectomia de C1 (determinada por SAC de 8, 3 mm na RM) seguida de fusão occipital-C3. Em nosso caso, até agora, não há evidência de descompensação axial, mas é necessário um período maior de acompanhamento. **Conclusões:** o tratamento de os odontoideum tem muitas considerações, mas é essencial, na presença de instabilidade e déficit neurológico, obter fusão sólida. Em caso de falha de amarrilho atlanto-axial posterior, a fusão occipital-C2 ou C3 com hastes parece ser uma opção excelente, com alta taxa de sucesso, evitando a necessidade de suporte complementar.

Descritores: Processo odontoide/cirurgia; Processo odontoide/radiografia; Vértebras cervicais; Traumatismos da coluna vertebral.

RESUMEN

Objetivo: Informe sobre las dificultades para tratar un caso de os odontoideum. **Métodos:** Paciente, una adolescente, de 12 años de edad, que desarrolló una cuadriparesia después de un traumatismo cervical leve en octubre de 2005. En el Departamento de Primeros Auxilios, se identificó una anomalía cervical congénita. La paciente fue colocada en un marco de Stryker®, unos días después, en un braguero de halo. Cuando transcurrieron 3 meses, surgió una infección alrededor de los pernos y se tuvo que retirar el chaleco de halo. Persistió una grave inestabilidad C1-2 y se intentó un procedimiento de Gallie C1-C2. En las semanas siguientes, el alambre desapareció y, en junio de 2006, se intentó otro procedimiento - laminectomía C1 y fusión de occipucio-C3. En los meses siguientes, el estado neurológico de la paciente mejoró y se observó una masa completa de fusión occipucio-C3. **Resultados:** Elegimos una artrodosis cervical posterior de C1-C2 usando la técnica de Gallie. Como la condición no se solucionó, realizamos una segunda cirugía, laminectomía C1 (determinada por SAC de 8, 3 mm en Imágenes de Resonancia Magnética), seguida por una fusión posterior occipucio-C3. En nuestro caso, hasta la fecha, no hay evidencia de descompensación axial, pero se necesita de un seguimiento más prolongado. **Conclusiones:** El tratamiento de os odontoideum tiene muchas consideraciones, pero lo esencial es que, en la presencia de inestabilidad y de déficit neurológico, se obtiene una fusión sólida. En el caso de falla del sistema de alambres posteriores atloidoaxoideos, la fusión occipucio-C2 o C3, con bastoncillos, parece ser una opción excelente que tiene una tasa alta de éxito, evitando la necesidad de apoyo adicional.

Descriptores: Apófisis odontoides/cirugía; Apófisis odontoides/radiografía; Vértebras cervicales; Traumatismos vertebrales.

1. Doutor em Ortopedia do Departamento de Ortopedia e Traumatologia do Hospital S. João – Porto, Portugal.
2. Professor Doutor do Departamento de Ortopedia e Traumatologia do Hospital S. João – Porto, Portugal.

Trabalho realizado no Departamento de Ortopedia e Traumatologia do Hospital S. João – Porto, Portugal.

Correspondência: Departamento de Ortopedia e Traumatologia do Hospital S. João - Alameda Professor Hernâni Monteiro Serviço Ortopedia e Traumatologia H. S. João 4200 Porto - Portugal.
Email: arpcinco@hotmail.com

INTRODUCTION

In 1863, separation of the odontoid process from the body of the axis was described in a postmortem specimen. In 1886, Giacomini coined the term *os odontoideum*^{1,2}.

Os odontoideum is rare, but the exact frequency is unknown. Known cases are either incidentally detected or are diagnosed when patients become symptomatic. No large-scale screening studies have been performed¹.

Based on the position of the dens tip *os odontoideum* is described as orthotopic or dystopic. In an orthotopic *os odontoideum*, the *dens* is in anatomic position. A dystopic *os odontoideum* is present when the *dens* is in any other position^{2,3} (Figure 1).



Figure 1. Orthotopic *Os Odontoideum*.

CASE REPORT

The authors report a case of a twelve year old female that developed a tetraparesia after a minor cervical trauma in October of 2005. In the emergency department a congenital cervical anomaly was identified. The patient was placed in a Stryker® frame and, few days later, in a halo bracing. After 3 months an infection around the pins emerged and the halo had to be removed. A severe C1-2 instability persisted and a C1-C2 Gallie procedure was attempted in January of 2006, 3 months after trauma. In the following weeks the graft disappeared and a severe C1-C2 instability persisted with the space available for the spinal cord was 10,8 mm in the X-ray and 8,3 in the MRI (Figures 2 and 3). Another procedure was attempted, in June 2006, that was a C1 laminectomy and fusion Occipital-C3 with Vertex System® (Medtronic) five months after the first procedure (Figure 4 e 5). In the following months the patient neurological status improved, allowing her to walk without assistance and to make her normal daily life activities. In July, 2 months after Occipital – C3 fusion, the fusion was evident in the X-ray. In the next one and half year of follow up she gained a good function – with only a functional grade 3 of 5 in intrinsic muscle of the left hand (C7/C8) as residual deficit - and a complete mass fusion Occipital – C3 was visible in July 2007 (Figure 6).

DISCUSSION

The primary factor that causes post-traumatic tetraparesia, frequently secondary to a slight trauma, is atlantoaxial instability. The stability of the atlantoaxial articulation depends fundamentally upon integrity of the odontoid process and the ligaments. When the odontoid process is disrupted, from traumatic or nontraumatic causes, the atlas will displace along the odontoid process, leading to instability². The differential diagnosis of injuries of the upper cervical spine in children includes fractures through the base of the



Figure 2. In the X – Ray the SAC is 10,8mm.

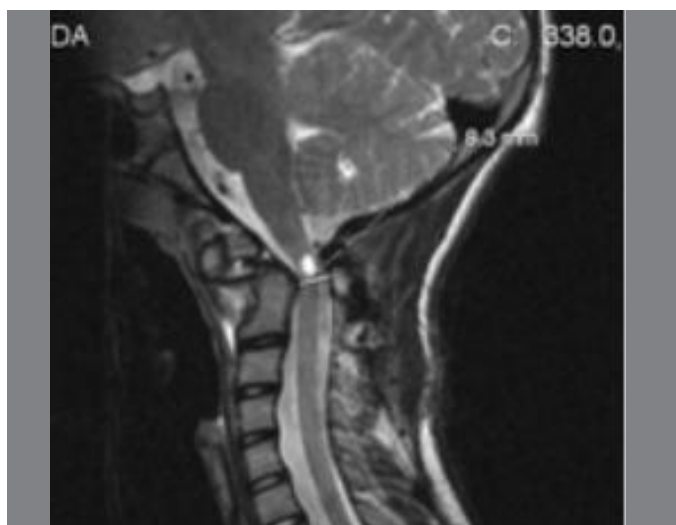


Figure 3. In the MRI the SAC is 8,3mm.



Figure 4. Intra – Operative C1 laminectomy and C0 – C3 fusion.



Figure 5. Lateral cervical spine showing C0 – C3 fusion in reduced position.



Figure 6. Lateral cervical spine showing C0 – C3 fusion in reduced position after one and half year

odontoid process at the vestigial disc between the odontoid and the body of C2 and atlantoaxial rotatory subluxation². Good radiological evaluation of the patients is mandatory with anterior and lateral radiographs of the cervical spine and open-mouth anterior-posterior⁴.

Os odontoideum appears as a round or oval ossicle with a smooth uniform cortex separated from the base of the axis by a wide gap^{4,5}. The ossicle border does not directly match up with the axis body. The wide gap separating the os odontoideum and the axis should lie above the level of the superior articular facets². The critical space available for the spinal cord (SAC) was proposed as 13 mm⁶. At the level of atlantoaxial joint, the spinal cord has a diameter

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of 8 – 11 mm, and the transverse ligament has a diameter of 4 mm⁴. In this case remains a 10, 8 mm space in Rx and 8,3 mm in MRI. A SAC of 11 mm is related with clinical improvement after a solid arthrodesis. It is important to check instability index since patient with more than 40 % have a high risk of myelopathy. Another factor of high risk is sagittal plane rotation angle with more than 20 degrees associated with a high risk of developing myelopathy⁷.

Patients with posttraumatic progressive neurological deficit should be treated surgically^{5,7}. Different treatment methods have been advocated by different authors^{2,6}. The standard surgical technique is posterior atlantoaxial wiring with either a Gallie or a Brooks fusion. The Magerl like procedure was not an option because of the patient age. We have chosen a posterior cervical arthrodesis of C1 – C2 using a Gallie technique. In patients with os odontoideum, posterior subluxation can occur with the Gallie procedure. This problem is related with over-tightening of wiring that causes posterior translation of the C1 ring and brings the ossicle into the canal and against the spinal cord. It is important that radiographs are obtained intraoperatively assuring that the patient is fused in a neutral position⁸.

Another major complication is nonunion, reaching 30 % when a fusion C1 – C2 with posterior atlantoaxial wiring is attempted. Post-operative management is important to avoid this complication. This includes a cervical bracing, as in our patient, or even a halo - bracing. Serial radiographs should be obtained to ensure progression to fusion and maintenance of stability. Nakagawa has presented a series of occipitocervical fusion in children, using rectangular rods that eliminated the complications associated with the halo and wire techniques for C1 – C2 fusion⁹ Wudbhav reported a series of 15 patients that were fused C1-2 or C0-C2 with or without wiring, with 4 non unions, 2 of them occurring in the 3 patients that were not placed in a halo jacket¹.

On the second surgery the option for a C1 laminectomy was determined by a SAC in MRI of 8, 3 mm. In presence of a myelopathy it is safer to relieve the compression of the spinal cord between the os odontoideum and the posterior arch of the atlas avoiding unexpected cord injury during the surgical procedure. The C1 laminectomy was followed by posterior occipito – C3 fusion.

We have to limit the posterior exposure of the cervical spine to prevent extension of the fusion to subjacent levels¹⁰. This has been reported in 42% of the children in the Parisini reports¹¹.

At this cervical level the optimum C1-C2 fusion angle should average 20° and in our case it has reached 38°. This alteration may implicate a change in sub axial alignment that may be resolved with the follow up. The cases with compensatory sub axial changes were found in patients whose fusion angle exceeded 30°¹¹. In our case, until now, it is not evident sub axial decompensation but a more prolonged follow-up is needed.

CONCLUSION

The treatment of os odontoideum has many considerations but the essential in the presence of instability and neurological deficit is to obtain a solid fusion. The halo bracing is the key factor of the treatment. It should be used to achieve a more anatomical position before and during surgery and, after surgery, to ensure good immobilization until a solid fusion is achieved. In the case of a failure of a posterior atlantoaxial wiring a C0 – C2 or C3 fusion with rods appear to be an excellent option with a high rate of success, avoiding the need of a supplementary support.

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