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#### SURGICAL TREATMENT FOR SEVERE CONGENITAL TORTICOLLIS

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With the active movement, we could see a

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Key words: Congenital Torticolis

# **ANAMNESIS:**

A 14 year-old male patient was submitted from another Hospital because of an advanced congenital torticollis with fibrosis and severe shortening of the right sternocleidomastoid (SCM) muscle, due to a lack of follow-up in his reference Hospital. He was derived to our Hospital's Spine Team to make an assessment.

He had no Drug's Allergic Reactions or any important past illnesses.

# **PHYSICAL EXAM:**

In repose, we could see the right shoulder more elevated than the contralateral one, and an asymmetry because of a head displacement to the left. Moreover, he presented a head right tilt and a chin left tilt (figure 1.1). Also, a fibrous cord could be seen at the right SCM (figure 1.2). 35° restriction for the right rotation movement (figure 1.3), as well as a limitation for the complete neck extension at 25°. The patient referred pain when we touched the right Sternocleidomastoid.

Neurological and vascular exams did not present any complication.

# **COMPLEMENTARY TESTS:**

A head and neck TC was solicited in order to discard any vertebral malformation or any C1-C2 subluxation sign. They were discarded, because only an odontoid little asymmetry, smaller than 15mm was seen (figure 2.1).

In the RMI, we could discard the atloaxoid luxation. We saw that the atlo-axoid joint transverse ligament was entire. It also was appreciated a very small approximation between the odontoid and the atlas lateral mass, secondary to the torticollis. The vertebral bodies and the intervertebral discs were non-altered (figure 2.2).

EMG: a study from the sternocleidomastoid-depend muscles was done, but no patology events were found.

# **DIAGNOSIS:**

Congenital torticollis, due to the right sternocleidomastoid shortening.

# TREATMENT:

Due to the long evolution of the pathology, it was decided to perform a surgical intervention, starting with a medial supraclavicular approach on the distal tendons of the right sternocleidomastoid (figure 3). Then proceeded to dissect by planes until the transverse dissection of the platysma of the neck. After the release of adhesions and fibrosis, the sternal and tendons clavicular of the right sternocleidomastoid were identified, and a tenotomy was performed.

After checking, by manipulating the patient's head, that a high limitation degree persisted for neck mobility, a longitudinal approach was performed on the proximal and posterior border of the SCM. During the dissection by planes, the sensory branch (Greater Auricular nerve) of the Facial Nerve was identified, dissected and referenced (Figure 4) and the External Jugular vein was identified, too (figure 5). After the release of fibrosis and adhesions in the proximal insertion of the SCM, we proceeded to section the insertion of the SCM in the mastoid apophysis (Figure 6).

After this second approach, a clear improvement in head's mobility was observed, although there was still a partial limitation due to the fibrosis of the SCM's deep fascia, which led to its release.

Then, a final manipulation of the neck was done, in order to check that an adequate mobility had been achieved, and after checking the integrity of the neural potentials by the Neurophysiology team (figure 7), the wounds were closed by planes, leaving a drainage tube to avoid possible bruising.

The intervention was performed under general anesthesia, in the supine position and after administering antibiotic prophylaxis with Cefazolin 1 gram. After the surgery, the patient was carried to the Hospitalization floor.

# **EVOLUTIVE CARES:**

The patient presented an immediate postoperative period without complications, beginning the ambulatory and rehabilitation exercises on the 2nd postoperative day, and a bivalve corset was placed to keep the head tilted to the left and with a slight rotation of the chin to the right, which must be worn by the nights. After evaluating corset and rehabilitation exercises tolerance by the patient, he went home 7days postoperatively, and was referred to external consultations of the Spine Team after two weeks, where it was observed that the evolution of the scar was good, and the appearance and mobility of the neck had improved, but there was yet a residual deformity, very difficult to improve totally (Figure 8). At that moment the staples and stitches were removed, the scar was healed, and the patient was urged to continue with the prescribed rehabilitation exercises, until the next visit in a month.

# **DISCUSSION:**

Torticollis is a deformity of the patient's neck, which can have multiple causes as a substrate. To classify it, it is usually taken into account whether the deformity was present at birth (congenital) or is acquired, and whether or not it produces pain (1). The most frequent form is congenital muscular torticollis, due to the involvement of the SCM. (1,2,3)

Normally, the diagnosis of congenital torticollis is made by Physical Examination. since the congenital muscular torticollis produces some very striking appearance, consisting of the deformity of the neck, because there is a tilt of the head towards the involved side of the affected SCM, and a rotation of the chin towards the contralateral shoulder (1,2,3,4). Usually, this look is accompanied by a prominence or fibrous cord on the side of the neck, which goes from the proximal insertion of the SCM in the mastoid process to the insertions in the sternum and clavicle (1,2). When this pathology is diagnosed, it is necessary to include in the differential diagnosis bone abnormalities that can cause a similar appearance (3).

As usual, the favourite treatment for congenital muscular torticollis is conservative, because by means of massage and rehabilitation exercises, an acceptable correction of deformity and an adequate mobility range of  $\mathbf{is}$ achieved in approximately 90% of cases (2.5).Normally, the rehabilitation exercises consist of rotation movements of the chin towards the ipsilateral shoulder, and tilting the head towards the contralateral shoulder, as well as forcing the active rotation towards the affected side (1,2).

Due to the great effectiveness of conservative treatment, the surgical treatment option is reserved only for 10% of patients who do not achieve an adequate range of mobility (those in which 30 or more degrees are lacking to reach full rotation) or whose deformity is still excessive at the aesthetic level (1,5).

In addition, we must bear in mind that surgical treatment can have many disadvantages for young children, because the scar that occurs can cause new fibrosis, and that the functional results do not differ if surgery is performed in a bit older children, so which surgery usually takes place at school age (5-6 years) (1).However, it must be borne in mind that if it is not treated at this age, advanced torticollis can cause pain, neck deformities difficult to repair despite surgical treatment, as in the case we are occupied with, or complications susch as the loss of the SCM column or the presence of lateral bands (5).

When it is decided to perform the surgical treatment, the surgical technique must be carefully planned. There are many options, but the one preferred by many authors is the bipolar approach of the SCM, through which adhesions and fibrosis are released both in the distal end of the SCM and in the proximal insertion in the mastoid process, since a very satisfactory functional and aesthetic result is usually achieved (1,2,6). A variant of this approach is the Unipolar one, which is used for less severe cases, in which resolution is achieved by releasing only the fibrosis of the distal insertions of the SCM (1,3).

The main complications of this technique that have been seen in the literature are the involvement of the Auricular Nerve, sensitive branch of the facial, which crosses below the SCM, and the involvement of the veins of the neck, especially the External Jugular, which runs along its posterior border (1). Less important complications are the recurrence of fibrous bands in the clavicular space that can be seen in almost 50% of the patients, but fortunately without functional or aesthetic consequences, and the loss of the SCM muscle relief in the neck, which is observed in almost 80% of the patients (2,3).

# **CONCLUSION:**

To sum up, we can obtain two conclusions from the analysis of this clinical case. The first is that torticollis is a pathology that, if treated early, responds well to conservative treatment, but if there is a lack of follow-up and it becomes an evolved torticollis, it can be difficult to correct even with surgical treatment. So that, the best age range for the surgical treatment of the congenital torticollis is between 1 to 4 years of age. (4)

The second is that, within surgical treatment, for advanced torticollis the most appropriate approach is bipolar, releasing both the proximal and the distal insertion of the sternocleidomastoid (5).

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# **Cervical Tuberculosis Infection and its Treatment**

	Background: Extrapulmonar tuberculosis (TBC) frequently includes vertebral	
	affection but cervical levels are the less frequently affected. Its clinical	
	presentation is very variable depending on the age. Both the low frequency and the	
	clinical variations are the reason because its treatment in children is controversial	
Copete I,	and different depending on the hospitals.	
Ferràs J,		
Bas T.	Methods: We show a 3 years woman with an cervical abscess and spondylodiscitis	
	because extrapulmonar TBC which presented with medullar and tracheal	
Hospital	compression. Patient needed anterior and posterior approach during the surgery to	
Univeristari I	release of the compression and arthrodesis.	
Politècnic La Fe,		
Valencia	Results: Clinical symptoms resolved after surgery. No new signs neither symptoms	
Avda Fernando	after 18 months of fellowship and Magnetic Resonance Image (MRI) controls.	
Abril Martorell	Conclusional According with hiblig member and our own concernings are recommend	
106, Valencia,	to newconalize each treatment for each nations to the correct treatment of this	
Spain	disease event in the strange eases when surgery will be needed	
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Key words: Pott disease, Tuberculosis, Cervical Spine.

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# Introduction

Tuberculosis (TBC) is an infectious disease with an important prevalence in both the developed and underdeveloped countries. Axial skeleton affection (Pott Disease) is frequent when it has extrapulmonar dissemination. Although, cervical affectation is rare, and its clinical presentation is different depending on the age. Normally, in younger than 10 years, it is extensive, diffuse and creates big abscesses. Although, in older than 10 tears, it's normally more located and less purulent, but it normally associates a higher neurologic affection.

On the other hand, in very small children, diagnosis is difficult because of the difficult of anamnesis and exploration to associates a delay in the diagnosis and in the treatment too.

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We show a Tuberculous spondylodiscitis in a 3 years old girl treated surgically with debridement and cervical arthrodesis in two times surgery because the abscess formation that required surgical drainage.

# **Case Report**

Girl, 3 years old, no previous diseases. She comes to emergency department because of dorsal pain of 4 weeks of evolution, with worsening during the night and improvement with Non Steroidal Analgesia (NSAID). No previous trauma neither other significant events referred to initiate the pain. No fever and no other systemic signs neither symptoms. During clinical exploration, laterocervical adenopathies were found. Lhermitte sign negative, no neck rigidity neither other meningeal symptoms. Cranial Nerves were normal and sensibility and motor function was normal everywhere. Reflexes were normal and symmetric. No sphincter affection was saw.

Slight loss of strength was seen on the MMII with a 4/5 level of the MRC was diagnosed. A light inclination of the body was saw and a little tilt of the head was referred. Pain with cervical flexion, but no with other movements of the head.

She wasn't able to sustain its head in a sitting position. Muscular contraction in cervicothoracic level, where a muscular contracture was diagnosed.

Blood analysis was performed and inflammatory signs were saw with a VSG of 72 mm/h, LDH of 221 U/L). White blood cells (WBC) were normal. Cervical X-ray were performed and an anterior and right tracheal displacement was obvious, with maintenance of its diameter. Left hiliar adenopathies were saw.

MRI show a prevertebral mass with soft tissue characteristics from the superior vertebral plate of C7 until intervertebral space of T3-T4. Its dimensions were 42 x 20 x 32 mm in CC, AP and T respectively. T1 was destroyed. In MRI, trachea was stenotic 3 mm with medullar compression.

Mediastinal adenopathies, cervical bilateral and little abscesses in supra and infratentorial territories were saw.

Osseous gammagraphy was normal. CRL was normal, clear and transparent. No cells were obtained and glucose levels were of 58 mg/dl, 48,4 mg/dl proteins, what didn't suspect us infection of the meninges. Mantoux was positive in 48h and HBR + E were began 2 weeks before surgery.

Surgery was taken in two times. The first one, an anterior approach was performed in order to make the decompression using superior and inferior to T1 discectomy and corpectomy of T1. Abscess was cleaned and a Moss Miami cylinder was included. After surgery, the girl did an episode of desaturation, agitation and ocular deviation that needed intubation, which was maintained until second surgery. In the second surgery, a posterior approach liberation was developed with C7 and T1 laminectomy and C4-T3 arthrodesis with pedicle screws and laminar hooks. After second time surgery, no other complications were developed, and she was able to maintain a 100% saturation without other clinically important events. Normal sensibility and motor function was maintained. Fusion was helped by a Minerva-Indiana corset as external fixation.



Figure 1. MIR that shows soft tissue mass between C7 and T3 with anterior extension and medullar compression.



Figure 2. Cervical MRI, axial image that shows tracheal compression and stenosis.



Figure 3 X ray image after second surgery wearing a Minerva-Indiana corset.

# Discussion

TBC is a world prevalent disease with a higher number of cases in no developed countries. Osseous TBC takes place between a 1% to 6% of children with pulmonary TBC and the most frequent location of the extrapulmonar TBC is the axial skeleton, which has de own name of Pott's disease. Normally, dorsal and lumbar zones are the most frequently affected, but in a 7% of the Pott's disease, it affects to cervical spine, what represents a 0.03% of all the pulmonary TBC. Normally, it's presented as neck pain, but other symptoms can be developed as fever, weight loss, anorexia, edema, torticolis, adenopathies, kyphosis, respiratory restriction, dysphagia, trismus and hipoglossus affection.

Depending on the age, its clinical signs and symptoms can vary. While big abscesses and a big local extension of the disease is normal up to 10 years, in older than this age that's strange and there is a predominance of paraplegia.

Axial Skeleton X-ray, Thorax Radiographies, CT and MRI are necessary to study properly the local affection of the disease. MRI is the gold standard test because of the information about neural structures, paravertebral zone, neurological affection and extension, what is useful not just to the diagnosis, to the surgery planning too.

AntiTBC drugs have improved the results of the treatment and can treat successfully the most important part of the patients. Although, because of the resistance to this drugs or the bad treatment fellow by the patient, surgery can be necessary. With surgery, we obtain a fast decompression and we can remove the affected bone, getting a best access to the infection by the antibiotic drugs. Patients with a low neurologic affection and an early surgery have better functional results after the treatment than those with a worse neurologic situation and a later surgery treatment.

Overall, surgery can take place when it's pain, neurologic affection, osseous destruction, instability, dyspnea, dysphagia, dysphonia or axial deformity. Of course, surgery must be developed when medical treatment fails. Anterior debridement with vertebral fusion(Hong Kong method) is widely used in patients with vertebral tuberculosis. It has better results than medical treatment and better than anterior debridement without arthrodesis, overall when fusion and deformity correction are necessary. It reduces the time to reduce pain, the time needed to resolve the abscess, the neurologic symptomatology and axial deformity.

Because of the instability created with a radical anterior debridement, anterior osseous graft and posterior fusion are recommended, in order to prevent de deformity progression after surgery. Posterior fusion, complementary to posterior fixation and anterior fusion, improves the results but it increases the surgery time, the blood loss, the postoperative complications and the hospital stance.

In conclusion, the anterior approach with anterior radical debridement and anterior graft is the election treatment when the deformity is no very important. Double surgery approach in two times is used when there is a mutisegmentary affection with a big deformity related, because of the big instability created during the anterior release and the need of posterior fixation to prevent the deformity progression. propósito de un caso. Revista española de cirugía osteoarticular. 2011. 46 (248).

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# **Giant Cells Tumour**

#### Abstract

	Background: The first line of treatment of Giant Cells Tumor (GCT) consists in the curettage and coverage with bone graft. New treatments as Denosumab have shown great results as adjuvants in its treatment, and is gaining popularity in the treatment, especially because the high rates of local recurrence after surgery and the limitations of the technique in very aggressive tumors, or in those located in complex regions. This monoclonal antibody inhibits Receptor Activator of Nuclear Factor Kappa Beta Ligand (RANKL) and can only be administered in skeletally mature patients, so it is contraindicated during pregnancy. There are other adjuvant therapies to surgery, such as cryotherapy with liquid nitrogen or the phenolization, which can be equally effective.
Authors Copete I <sup>1</sup> , Ferràs J	Methods: We present the case of a pregnant woman of 28 years old diagnosed with an aggressive GCT of proximal tibia during the first trimester of pregnancy. On this occasion, surgical treatment was performed by curettage and high speed milling. Adjuvant treatment was added with liquid nitrogen and the cavity was filled in with demineralized bone matrix and hydroxyapatite. Based on our experience and on a revision of the literature, there are few cases described of the treatment of a GCT in a woman during the pregnancy.
Angulo MA.	Results: The patient's symptoms disappeared after the intervention. There have not
Hospital	been shown signs of recurrence after 10 months of clinical follow-up and simple
Univeristari I	radiology controls. The treatment did not produce adverse effects in the fetus during
Politècnic La Fe,	pregnancy neither breastfeeding.
Valencia	Conclusions: Indications of Denosumab in the treatment of GCT include unresectable
Avda Fernando	tumors or those in which its resection leads to high morbidity, as well as tumors in the
Abril Martorell	spine, sacrum, pelvis and challenging lesions located in the upper and lower extremities.
106, Valencia,	If we cannot opt for this drug for medical or other reasons, cryotherapy with liquid
Spain	nitrogen may be effective in its treatment.

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#### Introduction

Giant cell tumor (GCT) is a locally aggressive neoplasm characterized by a richly vascularized tissue with and multinucleated giant cells. It represents between 5% and 8.6% of primary bone tumors <sup>[1.3]</sup>. It usually appears after achieving skeletal maturity, when the growth plates have already been closed, between the ages of 20 and 40 years, with a predominance in females (2:1) <sup>[3]</sup>. The 60% of the GCT occur in long bones and it often extends to the articulation <sup>[1,2,3]</sup>. The most common sites are the distal femur, proximal tibia, distal radius, the proximal humerus and sacrum. Rarely, it can be multifocal and when it happens, it is very often associated with Paget's disease. The metastatic spread is rare <sup>[2,3,13]</sup>.

# **Clinical Presentation**

The most frequently symptoms are pain and local inflammation. There may be functional limitation or even pathological fractures. The clinical behavior of the CTG cannot be predicted with accuracy with histologic findings, clinical examination neither radiological images. Despite this, there have been numerous attempts to set the aggressiveness of a tumor and it seems that less aggressive tumors have a higher trabeculation and a smaller and softer cortical than more aggressive GCT.

Recurrences after surgical treatment by means of curettage and bone graft are frequent and they range between 12% and 50% <sup>[1.2]</sup>.

# **Radiological presentation**

GCT is presented in plain radiography as an osteolytic, expansive and located eccentrically lesion, with usually welldefined but not sclerosing edges. It may present trabeculation or internal pseudotrabeculation, which represents the non-uniform growth of the tumor, and it normally doesn't have periosteal reaction. It is important to make the differential diagnosis with a chondroblastoma, especially in those cases with signs of aggressiveness <sup>[2]</sup>.

The Computed Tomography (CT) allows to delimit the extent of the tumor and a better view of the areas of cortical destruction. Nuclear Magnetic Resonance (NMR) images of the GCT present low signal intensity on T1-weighted image (T1WI), and high signal on T2-weighted sequences (T2WI). NMRI are also effective to demonstrate the involvement of subchondral bone and the extent of the tumor to the joint.

The Positron Emission Tomography (PET) images, do not provide additional information, although they can be useful in the detection of multiple foci if clinically a GCT is suspected.

The malignant GCT does not have additional radiographic characteristics.

Numerous classifications have been developed to establish the better protocol to the management of the TCG, but neither prognosis nor the risk of recurrence are predictable on the basis of the clinical presentation, radiographic or histological.

The Enneking staging system for benign bone tumors can be used to determine the definitive treatment. Campanacci described a classification system for these lesions based on x-ray images (Table 1). Grade 1 lesions of Campanacci are rare, and the majority of GCT are defined as grade 2 <sup>[2]</sup>.

Degree/Stadium	Enneking	Campanacci
I	Benign, indolent and biologically	A well circumscribed radiolucent
	static	lesion, without aggressive features
		(periosteal reaction, mass of soft
		parts, disruption of the cortical).
II	Progressive growth, limited by	Relatively well-defined edges
	natural barriers	without a border radiopaque
III	Locally aggressive with soft tissue	Affected borders, with destruction
	mass	of the cortical bone and soft tissue
		mass

Table 1. Comparison between classification of Enneking and Campanacci

# **Histological presentation**

There is a dual population of cells: mononuclear stromal cells and giant cells, both kind of cells distributed throughout the tumor. These giant cells are morphologically similar to osteoclasts and they have an elevated activity of acid phosphatase. They are round, oval or fusiform and generally have a large core with little chromatin and few nucleoli. The histological study with silver staining reveals a dense network of reticulin fibers surrounding individually the cells. As has already been said, the prediction of the evolution of a TCG based on their histologic appearance is impossible, and the histologic features have not been shown to correlate with the degree of local recurrence neither with the appearance of lung metastases.

The secondary transformation to a malignant GCT histologically diagnosed, is exceptionally uncommon without any previous radiation therapy <sup>[13]</sup>.

# Treatment

The first-line treatment consists on curettage and filling in with bone graft  $^{[1,2,4,7,10]}$ , though it is true that its recurrence rate is between 20% and 45%, depending on the serie  $^{[2]}$ .

Medical therapies include bisphosphonates and Denosumab <sup>[2,3,4,5,6,10,13]</sup>. Other treatments that have been used are corticosteroids, interferon and chemotherapy, and new treatments are been developed, as Dasatinib, a new Tyrosine Kinase Inhibitor (TKI). After the approval of Denosumab to the treatment of GCT, the use of chemotherapy or interferon has been relegated to the metastatic malignant tumors <sup>[3]</sup>.

Phenolization, coagulation with Argon laser, Cryotherapy with liquid nitrogen, hydrogen peroxide, embolization <sup>[2.8]</sup> and bone cement<sup>[2.7]</sup> are adjuvant treatments to the surgery.

While as radiation therapy is not used routinely, it has been used as an alternative tool to the GCT located in difficult places such as the sacrum and the vertebral column, because its high risk of local recurrence after curettage. It has also been used interferon for local and systemic control of the disease, with variable results.

# **Case report**

We present a case of a 28-year-old pregnant woman of 12 weeks with a lesion in the proximal tibia epiphyseal region (Figure 1). Clinically, she referred pain on the medial compartment with the weight load. There was no instability or other symptoms neither signs of interest.



Image 3. Postoperative plain x-ray. AP and lateral projections.



Image 1. Nuclear Magnetic Resonance Images, coronal slices.



Image 2. . Nuclear Magnetic Resonance Images sagittal slices.



Image 4 . Control CT scan 9 months after surgery. Coronal slices.

A cylindrical piece of tissue was taken using incisional biopsy, and histological diagnosis of giant cell tumor of bone was done.

She didn't present other lesions, such as pulmonary metastases neither symptoms in other places.

The curettage was performed, including high-speed milling, adjuvant treatment with liquid nitrogen and filling in the defect with demineralized bone matrix and hydroxyapatite. After surgery and three days of hospitalization, body weight load was restricted and she was asked for to walk with the help of sticks, protecting the treated leg. During her hospital stance and the successive follow-up, the patient continued with pregnancy controls, occurring only a cytomegalovirus infection as complication. 10 months after surgery, the patient is asymptomatic and NRMI and Radiographic controls are satisfactory (Figure 6).



Image 5. control CT scan 9 months after surgery. Sagital slices.



Image 6. X-ray control 10 months after surgery

It is important to bear in mind that, as a pregnant woman, all tests were adjusted to the precise radiation doses, as well as the careful use of fluoroscopy during the procedure.

# **Surgical Technique**

In the first place, an incision medial to the tibial tuberosity is done, deepen until you reach the bone. A resection of the anterior cortex wall is performed and then curettage of the cavity with a little spoon and with high-speed ream is developed. Once this is done, the cavity is filled in partially with bone substitute. On this occasion, 10 ml were used at the level of the articular surface and the, carefully, we added liquid nitrogen. Finally, we filled the remnant cavity with 20ml of bone substitute, a piece of hydroxyapatite of 10x10x40mm and 30cc of hydroxyapatite granules. Finally, all the process was reviewed with fluoroscopy and a drain was also placed.

# Discussion

We often come across situations that we cannot deal with the standard procedures and in which we must consider other treatment alternatives. In this cases, it's necessary to know what tools do we have in order to get the better in our specific case.

Denosumab is a monoclonal antibody that binds with high affinity and specificity to RANKL <sup>[3,4,6 WCSD POLICY]</sup> by deleting the osteolytic activity. The GCT is characterized by stromal cells expressing RANKL and osteoclast-like giant cells that express RANK <sup>[3]</sup>. That's the reason because Denosumab delays the progression of this disease <sup>[3]</sup> Nonetheless, as other antiresorptive treatments do, it can lead to hypocalcemia and osteonecrosis of the jaw.

Actually, Denosumab has been approved along with surgery to the treatment of GCT in skeletally mature adults and adolescents on which, because its location, they are considered unresectable tumors or when its resection involves a high morbidity: spinal column, sacrum or the pelvis and challenging lesions in the upper and lower extremities [3,4,5,10,13] With Denosumab, it's possible to reduce the size of GCT preoperatively, providing relief of symptoms before surgery and facilitating the resection of the tumor <sup>[12]</sup> .Even so, when used for TCG bone, the long-term effects of Denosumab are unknown <sup>[10]</sup> .

Bisphosphonates are used on the basis of their ability to induce apoptosis in stromal cells and osteoclasts in vitro. The clinical evidence of bisphosphonates in GCT is limited to retrospective series and case reports, and more studies are needed to use it securely<sup>[2]</sup>.

It has been shown that cryosurgery reduces the rate of local recurrence up to 8% <sup>[2]</sup>. The freeze-thaw cycle kills cells away from the surface, which further extends the depth of the curettage. Cryosurgery involves the direct application of liquid nitrogen in the cavity of the tumor and has been shown to be an effective adjuvant therapy to tumor resection <sup>[2]</sup>. In spite of this, it has been associated with a significant incidence of pathologic fracture and vascular injury, the first being the most frequent complication <sup>[2]</sup>.

External beam radiation with argon laser has been used to complement the surgical treatment in patients who are medically inoperable or in those who have tumors that are technically difficult to remove or unresectable, due to its location <sup>[2]</sup>. Although radiation therapy is not used routinely, it has been used a great tool to treat the GCT in difficult places, such as the sacrum and the vertebral column. It has also been used interferon for local and systemic control and, with diverse results depending on the series <sup>[2]</sup>.

Other adjuvant treatments that have been used traditionally are the fenolization , hydrogen peroxide and the embolization and the bone cement .

The use of chemotherapy or interferon has been relegated to the metastatic malignant GCTs <sup>[3]</sup>.

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# Shoulder Septic Arthritis

#### Abstract

	Background: Septic arthritis is an emergency in orthopedics. Several mechanisms have been described: hematogenous spread, extension from an adjacent focus and direct inoculation, being the first one the most frequent mechanism. If not handled properly neither early, what can lead to the destruction of the articular cartilage and the production of sequelae. In an incipient case, good results can be got with conservative treatment, but usually surgical management is necessary for the resolution of the process.
Authors	Material and Methods: We present a case of a 5 year old child who suffered an episode of arthritis after the administration of vaccine against serogroup B Neisseria Meningitidis. Symptoms began 2 hours after vaccination with pain and fever of up to 39°C. At this moment, arthrocentesis was performed and intravenous antimicrobial therapy was initiated.
Copete I <sup>1</sup> , Ferràs J Angulo MA.	Results: The evolution was favorable with no need of surgical treatment. Symptoms continued going down after the management with arthrocentesis and intravenous antibiotic treatment, until disappearing completely in a few days. During the follow-up, no signs of recurrence have
Hospital Univeristari I Politècnic La Fe, Valencia Avda Fernando Abril Martorell 106, Valencia, Spain	appeared after 12 months. Conclusions: despite the fact that the hematogenous spread is the most frequent mechanism of establishment of arthritis septic, direct inoculation can justify a case of arthritis after vaccination in the deltoid region. We must be careful during vaccination with bacteria toxoids in the deltoid region, due to the risk of reactive arthritis and its differential diagnosis with septic arthritis. More studies are needed to clarify the diagnoses in the borderline cases, being molecular biology techniques as protein chain reaction a fast and useful tool.

1. Corresponding Author: 1. Corresponding Author: ivan.copete01@gmail.com Key words: septic arthritis, post-vaccination arthritis, inflammatory arthritis.

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# Introduction

Septic arthritis (SA) is a joint inflammation caused by microorganisms that leads to the destruction of the articular cartilage. It appears preferably in neonates and children between the ages of 2 and 3 years [1,5,10], and even though it can

affect every joint, the most common are the hip [1,2,4,5], the knee and elbow [2].Bacteria can penetrate to the joint for one of the three tracks: hematogenous, direct extension from an adjacent focus as a osteomyelitis and by direct inoculation of pathogens. The causative microorganism tend to be Staphylococcus aureus [1,2,12,14], Streptococcus haemolyticus, Haemophilus influenzae, pneumococcus and gonococcus. In recent years there has been an increase of Candida albicans due to prolonged intravenous catheterization in infants in critical condition [1]. In underdeveloped countries, it is frequently isolated non-typhoidal Salmonella, even being the most common pathogen in some regions [6]. In children older than 3 years, the germs are similar to adults: S. aureus, Streptococcus and N.gonorrheae. There are between a 20% and a 40% of septic arthritis in which the microorganism is unknown.

Regarding to the pathology, the synovial membrane becomes swollen, edematous and hyperemic, and increases the volume of synovial fluid which distends the joint. This may be fluid and cloudy and contain polymorphonuclear leukocytes. In addition, decreases the glucose content of the synovial fluid and increases the proteins. Bacteria can be identified in the joint through the implementation of smears and Gram staining. The persistence of the infection may accumulate pus in the articular cavity quickly and produce destructive changes in the hyaline cartilage of the joint. In addition, the infection can spread the underlying bone, causing an osteomyelitis

The onset of symptoms is usually acute, posing as joint pain, limp in case of lower limb involvement, functional limitation and bad general condition. Irritability, lack of appetite and fever are frequent, although in neonates systemic symptoms may not appear. In the clinical exploration, the joint is distended and hot because of the joint effusion. The joint is presented in the position in which pressure is the less, generally with a certain degree of flexion at the hip, knee, ankle, elbow; but in the neutral position on the wrist, and in slight flexion and abduction in the shoulder.

To carry out the diagnosis, clinical suspicion is mandatory, and it must be supplemented with image tests. On plain x-ray we can observe indirect signs of intraarticular effusion, as distension of the joint capsule, explained by an increase of the synovial fluid, which will be displayed easily and measured with the ultrasonography. In certain cases it may also be useful scintigraphy with Tc99m, which will show increased uptake.

In the laboratory data are useful parameters of sepsis, the elevation of acute phase reactants in blood analysis (Complete Blood Count, C reactive **Protein and Globular Sedimentation** Velocity) and the analysis of joint fluid. The biochemistry will show data previously announced, and increased polymorphonuclear leukocytes. It is important to order cultures and Gram Stain, because it can guide the empirical treatment. The certainty diagnosis is given by the positivity of the crops but sometimes, it doesn't proliferate pathogenic microorganisms. In these cases, diagnosis is done indirectly with the results of microscopic examination of arthrocentesis, histological signs compatible with an acute inflammatory process, irritability and

effusion, increase of the local temperature, ultrasound or radiographic data compatible, systemic response, fever, elevated inflammatory parameters in the blood test, improvement of the symptoms and signs with empirical antibiotic therapy and surgical drainage, as well as the absence of other possible diagnoses.

To evacuate the bacterial products and the remains of the infectious material, surgical drainage of the joint must be done in almost all patients. Sometimes, however, the infection is diagnosed early and the micro-organism is very sensitive to treatment, and in these cases the improvement with conservative measures can be impressive with immediate pain relief, improvement of mobility, the normalization of the temperature and the disappearance of local symptoms of effusion and synovial thickening. In these cases there would be no need for surgical drainage [1].

The synovial fluid culture remains the gold standard for diagnosis, but their results require time, so that the diagnosis in the emergency room, where normally are presented these cases, is based on the combination of clinical and laboratory parameters, being in many cases uncertain at the initial time.

#### Case Report

We present the case of a child of 5 year old without relevant clinical background, who went to the emergency department two days after having received the dose of vaccination against Neisseria meningitidis type B. Two hours after administration of the vaccine began with pain in the ipsilateral arm to the puncture site and fever of up to 39°C the night of the event. In the first assessment, he received treatment with antihistamines (Hydroxyzine dihydrochloride) and antipyretics, without improvement of the symptomatology.

No other infection neither inflammation focus were detected in the general anamnesis and exploration. He only presented swelling of the shoulder and increment of the local temperature, with pain on passive abduction from 90° and the active abduction of 20°.

On the complementary tests, the results of ultrasonography showed abundant liquid (7.4cc intraarticular.) with increased echogenicity without evidence of cellulitis or abscess of the soft parts; on the analytical study, it highlights a CRP of 48 mg/L, procalcitonin (PCT) of 0.27 ng/mL, 14,200 leukocytes with a 78% of Neutrophils/µL and an erythrocyte sedimentation rate (ESR) of 44 mm/h. The joint fluid taken by arthrocentesis was eco-guided and a total of 4 cc. were obtained. It was cloudy, with a white blood cell count >150,000/µL and a 94% of neutrophils. Biochemistry, gram stain and culture were performed. Gram staining show leukocytes but not bacteria. The crops were negatives after four days.

During the period of hospitalization, treatment with cloxacillin iv was prescribed . After four days, objective clinical improvement was seen, and decrease of acute phase reactants were obtained, changing at this time to a vancomycin therapy.

One year after the episode, physical examination, laboratory and ultrasound images, were completely normal.

#### Discussion

Although the direct inoculation of the germ has been attributed classically to an invasive procedure that violate the joint, sometimes it can be produced by accidental injection of pathogens after a vaccination or by the invasion of a close infectious process as a cellulitis. It's difficult to differentiate between an inflammatory reactive arthritis and an incipient septic arthritis, but it's important because its influence in the initial treatment.

Due to relatively frequent septic arthritis with negative cultures (till 78% depending on the series [15]), there are authors who defend the use of polymerase chain reaction (PCR) in medical centers to improve the detection of bacteria in the joint fluid, [9] predominantly in those cases in which the criteria of septic arthritis is not met the strict. However, it is a method that requires of the aspiration of the fluid to be carried out and that it takes an average of 14 days [9] to obtain the result. The combination of blood culture joint fluid culture and PCR improves the rate of detection of microorganisms. Nevertheless the PCR provides additional information for the diagnosis confirmation and has a higher rate of detection of bacteria than joint fluid cultures, the delay of the results and the inability to provide sensitivity to antibiotics are factors that currently limit its clinical utility [9].

With regard to the therapeutic management, the traditional treatment consists in a protracted course of intravenous antibiotics combined with aggressive debridement surgery. However, this approach is questioned by the tests that show satisfactory results with a shorter treatment and less invasive surgery. We consider too dogmatic to assert that all septic arthritis should be dealt with by arthrotomy and cannot be treated with intravenous antibiotic therapy and washing with arthroscopy. Thus, arthrotomy allows full debridement with the elimination of remains and the rupture of the loculations, but carries the morbidity of an open surgery and general anesthesia.

An alternative to arthrotomy is the single joint aspiration, which carries a minimal morbidity and may not require general anesthesia in older children, but it can provide a less satisfactory drainage of the joint. The arthroscopic wash is another option with some potential advantages and is increasingly used, particularly in the knee [6]. The majority of authors argue that in the septic hip deep, difficult to aspire and in which the consequences of sepsis can be devastating, the arthrotomy and flushing is the best method of treatment. Many others, however, discuss the best way to drain the shoulder in children: while some recommend the aspiration of all the shoulders septic joints, others defend the realization of arthrotomy. Despite all of this, the role of the various therapies remains very discussed [7].

If it seems clear that antibiotics should be administered parenterally initially and then orally by once the signs and symptoms of infection begin to be resolved [6]. The recommended duration of the antibacterial treatment varies from two weeks to more than three months, although the majority of the authors use a six week treatment[6]. Many authors argue that if the signs and symptoms subside in a few days, and the level of Creactive protein in the serum drops below 20 mg/l, the antibiotic can usually be safely suspended [10].

#### Conclusion

The diagnosis of arthritis post-vaccination is an important diagnostic challenge due to its synovial inflammatory response. The relatively low specificity of indirect diagnosis methods worsens the therapeutic decision between an inflammatory arthritis and a septic arthritis. Cultures are considered the gold standard for its diagnosis, but it requires more time than we can wait before treating it. The use of molecular biology techniques such as PCR allows the diagnosis in a shorter time than cultures, being it very useful in the cases of arthritis post-vaccination with bacteria toxins.

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