Trochlear Nerve Schwannoma: Case Report and Literature Review

Schwannoma do nervo troclear: revisão da literatura e relato de caso

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Abstract

Schwannomas arise from the Schwann cells of the peripheral and cranial nerves. They represent 8% of the primary cerebral neoplasms. Although schwannomas usually develop in sensory nerves, most often on the vestibular and trigeminal nerves, in very rare cases they can develop in motor nerves. We reported an unusual case of a 29-year-old woman with headache, nausea, vomiting, and blurred vision ongoing for 3 years. Magnetic resonance of the brain showed a solid-cystic expanded injury, heterogeneous, with limits partially defined and epicenter on the pineal gland. The lesion presented hyposignal in T1 and isosignal in T2. An intense enhancement of the solid part was observed after contrast injection. Foci of calcification and absence of diffusion restriction were also observed. The patient underwent microneurosurgery with supracerebellar infratentorial approach in a seated position. Subtotal resection was performed with maintenance of calcified tumor tissue adhered to the right Rosenthal basal vein. In the postoperative phase, the patient remained with diplopia when looking down; however, she reported improvement of headache and nausea.

Keywords
► neurilemmoma
► trochlear nerve
► diplopia

Resumo

Os schwannomas surgem das células de Schwann, dos nervos periféricos e cranianos e representam 8% das neoplasias cerebrais primárias. Apesar de os schwannomas se desenvolverem comumente em nervos sensoriais, mais frequentemente no nervo vestibular e trigêmeo, em casos muito raros ele pode se desenvolver em nervos motores. Relatamos um caso raro, de uma paciente do sexo feminino, 29 anos, com quadro de cefaleia, náuseas, vômitos e turvação visual há três anos. Ressonância magnética de encéfalo demonstrou lesão expansiva sólido-cística, heterogênea, com limites parcialmente definidos e epicentro na glândula pineal. Apresentou hipossinal em T1 e isossinal em T2. Houve intenso realce da parte sólida após injeção de contraste. Foram observados focos de calcificação e ausência de restrição à difusão. A paciente foi submetida a tratamento microneurocirúrgico com acesso infratentorial supracerebelar em posição sentada. Houve ressecção subtotal com manutenção de tecido tumoral calcificado aderido a veia basal de Rosenthal direita. No pós-operatório, a paciente permaneceu com diplopia na mirada ocular para baixo, entretanto apresentou melhora de cefaleia e náuseas.

Palavras-chave
► neurilemoma
► nervo troclear
► diplopia

Introduction

Schwannomas arise from the Schwann cells and represent 8% of the primary cerebral neoplasms.[1] They usually develop in sensory nerves, most often in vestibular nerve.[2,3] In very rare cases schwannomas can develop in motor nerves, including the trochlear nerve.[2] Trochlear nerve schwannomas are extremely rare and, according to our review, there are 37 surgical cases related in the literature, including ours.

Case Report

The case described is that of a female patient, 29 years-old, with headache associated with nausea, vomiting and blurred vision for 3 years. Imaging studies performed in another service showed an expansive lesion, causing mass effect, resulting in hydrocephalus. The patient was submitted to ventriculoperitoneal shunt, with improvement of the clinical condition. After the surgery, clinical follow-up was realized.

The patient was admitted in our service 2 years after the ventriculoperitoneal shunt surgery. She had diplopia when looking down, headache and nausea refractory to the clinical treatment. Magnetic resonance imaging (MRI) of the brain showed a solid-cystic expanded injury, heterogeneous, with limits partially defined and epicenter on the pineal gland. The lesion presented hyposignal in T1 and isosignal in T2. An intense enhancement of the solid part was observed after contrast injection (►Figs. 1, 2, 3). Foci of calcification and absence of diffusion restriction were observed as well.

Cerebrospinal fluid examination did not show tumor cells and the search for α-fetoprotein and human chorionic gonadotropin were negative. The patient underwent micro-neurosurgery with supracerebellar infratentorial approach in a seated position. The surgery was performed with ultrasonic aspiration and central enucleation of the tumor, allowing a better mobility of the tumor capsule and dissection of deep veins and mesencephalic roof. Subtotal resection was performed, with maintenance of calcified tumor tissue adhered to the right Rosenthal basal vein due to profuse
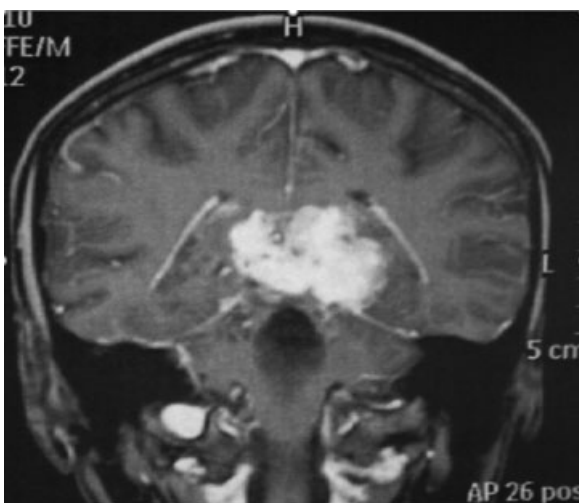
Fig. 1 Coronal cut in T1 after contrast injection with expansive lesion in the topography of the pineal gland.
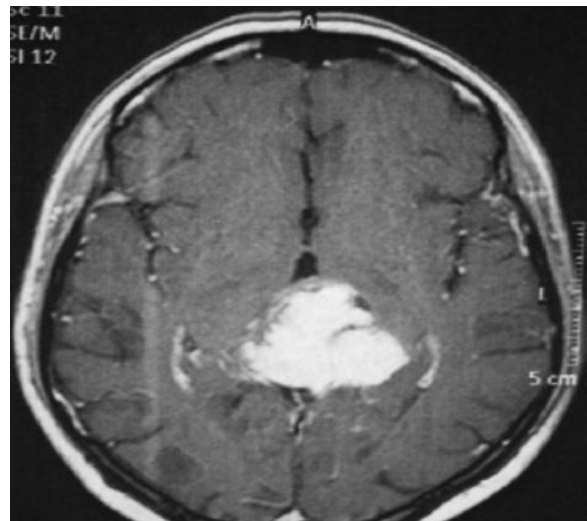
Fig. 2 Axial cut in T1 after injection of contrast with expansive lesion in the topography of the pineal gland.
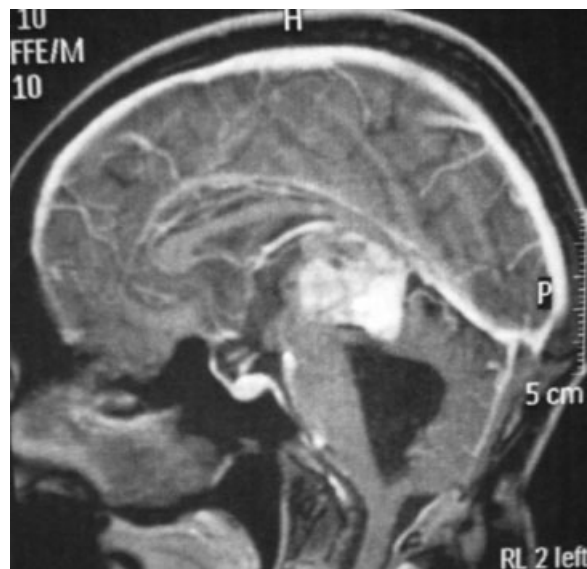
Fig. 3 Sagittal cut in T1 after injection of contrast with expansive lesion in the topography of the pineal gland.

bleeding when attempting resection of the tumor portion in this topography (►Fig. 4).

The fourth nerve schwannomas typically show predominance of Antoni B tissue, have frequent clusters of xanthomatous macrophages and are poor in Verocay bodies (these are often more common in spinal tumors). Reticulin is abundant and distinctly pericellular in schwannomas.

Schwannomas are strongly and uniformly reactive for S-100 protein; they may be positive for epithelial membrane antigen (EMA), but not frquently, and do not exhibit the membrane-characteristic pattern of meningioma. Microscopic patterns and immunohistochemical findings in our case (►Fig. 5). In the postoperative phase, the patient remained with diplopia when looking down, reporting improvement of headache and nausea.

Fig. 4 Axial cut of computed tomography with contrast confirming postoperative status.

Discussion

We did a literary review using the PubMed and COCHRANE platforms searching for the following terms: trochlear nerve schwannoma and schwannoma surgery.

We found the occurrence of 37 surgical cases of trochlear schwannoma. The average age at diagnosis was 44.8 years, with a standard deviation of 15.1. The female gender was predominant, with 21 cases (58.3%) (►Table 1).

Trochlear nerve schwannoma presents as initial symptoms, in most cases, diplopia (60%), hemiplegia (43%), headache (40%) and cerebellar symptoms (37%), such as ataxia, dysmetria and nystagmus.[29,33] In the present study, trochlear nerve palsy was evidenced in 17 (50%) of 34 cases
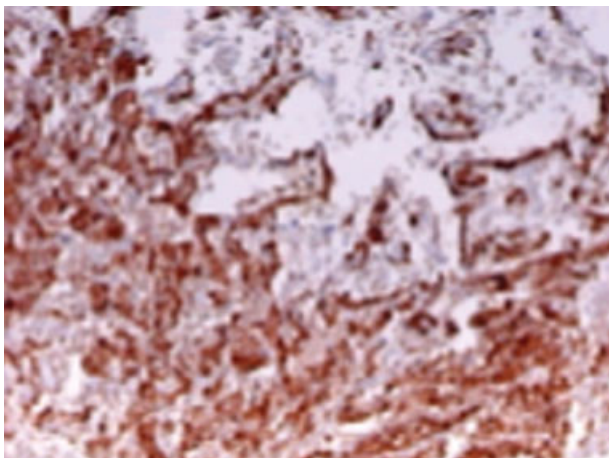
Fig. 5 Schwannoma strongly and uniformly reactive for S-100 protein by immunohistochemistry.

(►Table 1). In three case reports, intratumoral hemorrhage occurred as a complication.[7,26,33]

Macroscopically, schwannomas are encapsulated, well-delimited, lobulated, generally grayish masses, and may have areas of cystic alterations and xanthomatosis.[36,37]

Magnetic resonance imaging showed to be the most used imaging exam in the evaluation of schwannoma.[24,29,31,32,38] Computed tomography (CT) may also be used for the same purpose.[33] At CT, it tends to present as a single, well-circumscribed and solid-appearing mass, located mainly in the course of the IV cranial nerve.[32,38] At MRI, schwannomas present with hyposignal in T1, hypersignal in T2 and show an intense enhancement after intravenous administration of gadolinium contrast.[38]

Schwannomas can be diagnosed with conventional techniques when they are small and located in the cerebellum-pontine cistern. However, when they reach very large sizes and the intracanalicular part is not obvious, it may be difficult to discriminate them from meningiomas and metastases.[39] So, the use of advanced imaging techniques may be necessary. Studies with vestibular schwannomas by the cerebellar-angle have shown that, through diffusion-weighted imaging (DWI), it is possible to observe that the schwannomas solid component, usually isointense to brain parenchyma, presents an apparent diffusion coefficient (ADC) that ranges from 1.1 to $1.7 \times 10-3$ mm^2/S, which is considered elevated when compared with the normal brain parenchyma ($1.4 \times 10-3 mm^2/S$).[40,41] This increased coefficient may reflect the lower cell density of the Antoni B cells of schwannomas. However, the ADC alone cannot differentiate schwannoma from meningiomas. Even though schwannomas present a significantly higher average ADC value, there is considerable overlap among the values, which makes it difficult to differentiate meningiomas from schwannomas.[41]

Otherwise, it is possible to differentiate schwannomas from meningiomas through magnetic resonance (MR) perfusion. The schwannomas cerebral blood volume (CBV) is significantly smaller when compared with meningiomas.[42] Even if there is an overlap in the reason between the CBV of both entities, a threshold of 4.4 is the highest found in the schwannomas, while the meningiomas CBV average ranges from 6 to 9.[42–44]

Another way to differentiate meningiomas from schwannomas is through MR proton spectroscopy, where the schwannomas show a myo-inositol peak of 3.55 pm and absence of alanine.[45] Meningiomas are characterized by a high choline signal with very low signs of creatine and N-acetylaspartate, and alanine presence.[46]

The trochlear nerve schwannomas are classified in three subtypes: cisternal, cisternocavernous and cavernous.[13] In our review, 34 of the 37 cases evaluated were cystenal (91.8%), two cases (5.4%) were cisternocavernous and one case (2.7%) was cavernous.

Contrary to what can be found in the literature regarding trochlear schwannomas centered in the cisternal portion, the present case reports that the trochlear schwannoma was found with epicenter in the pineal gland. In our review, this only occurred in one other occasion, generating a second reported case with this approach for the referred pathology. The

Table 1 Literary review of trochlear nerve schwannomas.

Reference	Age/Sex	4th cranial nerve palsy	Location	Surgery approach	Follow-up
King[4]	55/F	No	Ambient Cistern	Subtemporal transtentorial to the right	Appearance of fourth cranial nerve palsy on the right side and paresis in the right leg
Boggan[5]	32/F	Yes	Ambient Cistern	Subtemporal transtentorial craniotomy to the right	Persistence of fourth cranial nerve palsy on the right side and absence of corneal reflex on the right side after 8 months.
Leunda[6]	54/M	Yes	Ambient Cistern	Subtempora ltranstentorial craniotomy to the right	Persistence of fourth cranial nerve palsy on the right side after 1 year.
Leunda[6]	16/F	Yes	Cisternocavernous	Subtemporal transtentorial craniotomy to the left	Persistence of fourth cranial nerve palsy on the left side after 6 months.
Yamamoto[7]	37/F	Yes	Ambient Cistern	Subtempora ltranstentorial craniotomy to the right	Persistence of fourth cranial nerve palsy on the right side after 1 year.
Garen[8]	18/F	No	Ambient Cistern	Subtemporal craniotomy to the left	Improvement of third cranial nerve palsy.
Tokuriki[9]	43/M	No	Ambient Cistern	Temporal craniotomy to the right	Appearance of fourth cranial nerve palsy on the right side after 1 month.
Murakawa[10]	23/F	No	Ambient Cistern	Pterional and suboccipital	Appearance of fourth cranial nerve palsy on the right side.
Maurice-Williams[11]	56/M	No	Ambient Cistern	Lateral suboccipital craniotomy to the left	Appearance of fourth cranial nerve palsy on the left side after 2 years.
Samii[12]	53/F	Yes	Ambient Cistern	Pterional craniotomy to the left	Persistence of fourth cranial nerve palsy on the left side.
Celli[13]	51/M	Yes	Ambient Cistern	Subtempora ltranstentorial craniotomy to the right	Persistence of fourth cranial nerve palsy on the right side after 5 years.
Lanotte[14]	NI	NI	Ambient Cistern	NI	NI
Jackwoski[15]	26/F	Yes	Ambient Cistern	Transtemporal craniotomy to the left	Persistence of light fourth cranial nerve palsy on the left side.
Abe[16]	60/M	No	Ambient Cistern	Lateral suboccipital craniotomy	Appearance of paralysis in the IV pair.
Abe[16]	57/M	No	Ambient Cistern	Subtemporal transtentorial craniotomy to the right	Appearance of fourth cranial nerve palsy after 4 months.
Dolenc and Coscia[17]	68/M	No	Ambient Cistern	Lateral suboccipital craniotomy to the right	Appearance of postoperative fourth cranial nerve palsy, but with improvement in 2 months. Improvement of the hemiparesis to the left within 2 months.
Beppu[18]	66/M	No	Ambient Cistern	Lateral suboccipital craniotomy to the right	Appearance of paralysis of the abducent nerve.
Santoreneos[19]	35/F	No	Ambient Cistern	Subtemporal transtentorial craniotomy	Appearance of fourth cranial nerve palsy on the left side.
Nadkarni and Goel[20]	48/F	No	Ambient Cistern	Subtemporal transtentorial craniotomy	Superior oblique muscle weakness

(Continued)

Table 1 (*Continued*)

Reference	Age/Sex	4th cranial nerve palsy	Location	Surgery approach	Follow-up
Matsui[21]	61/M	Yes	Ambient Cistern	Presigmoid transpetrosal craniotomy to the right	Persistence of fourth cranial nerve palsy on the right side.
Veshchev[22]	26/F	No	Cavernous Sinus	Pterional craniotomy to the left	Appearance of postoperative fourth cranial nerve palsy on the left side, with significant improvement in 4 months.
Türe[23]	31/M	Yes	Pineal Region	Infratentorial - Lateral supracerebellar	Persistence of fourth cranial nerve palsy on the left side.
Shenouda[24]	49/M	Yes	Cisternocavernous	Presigmoid combined transpetrosal craniotomy	Persistence of fourth cranial nerve palsy on the left side and deafness.
Du[2]	17/F	No	Ambient Cistern	Orbitozygomatic pterional	Asymptomatic
Shenoy and Raja[25]	54/F	No	Ambient Cistern	Subtemporal transtentorial	Appearance of hemianesthesia to the left after 1 year.
Ohba[26]	48/M	Yes	Ambient Cistern	Anterior transpetrosal	Permanence of the fourth cranial nerve palsy on the right side after 4 months.
Gerganov[27]	52/F	Yes	Ambient Cistern	Suboccipital	Permanence of dysfunction in the trochlear nerve.
Grigorian and Korobova[28]	47/F	NI	Ambient Cistern	Retromastoidal	NI
Grigorian and Korobova[28]	44/F	NI	Ambient Cistern	Paramedian subtentorial supracerebellar	NI
Kohama[29]	47/F	No	Ambient Cistern	Posterior transpetrosal craniotomy	Appearance of fourth cranial nerve palsy on the left side.
Bartalena[30]	50/F	Yes	Ambient Cistern	Subtemporal-transpetrosal	Persistence of diplopia with posterior surgical correction by myectomy.
Younes[31]	65/F	Yes	Ambient Cistern	Pterional	Improvement of fourth cranial nerve palsy after 2 years postoperative.
Boucher and Michael[32]	64/M	Yes	Ambient Cistern	Anterior transpetrosal	Persistence of fourth cranial nerve palsy on the right side.
Hatae[33]	44/M	Yes	Ambient Cistern	Zygomatic transpetrosal	Persistence of fourth cranial nerve palsy on the left side.
Samadian[34]	63/M	No	Ambient Cistern	Left retrossigmoid	Appearance of fourth cranial nerve palsy.
Chaudhry[35]	24/F	No	Ambient Cistern	Paraoccipital posterior interhemispheric transtentorial, from the right side	Appearance of postoperative fourth cranial nerve palsy without complete resolution after 6 months. Emergence of ataxia, with almost complete resolution after 6 months.
Cunha (present study)	29/F	Yes	Pineal Region	Supracerebellar – infratentorial	Fourth cranial nerve palsy with 3 months postoperative follow-up.

Abbreviations: F, female; M, male; NI, not informed.

incidence of tumors in the pineal region ranges from 0.4 to 1% of all primary brain tumors, with studies showing a higher percentage of tumors in this region in children and adolescents, with rates ranging from 3 to 8%.[47–51] As a differential diagnosis for the pineal region, we must consider the tumors divided in four categories: germ cell tumors, pineal parenchyma tumors, pineal gland support tissue tumors (for example, astrocytomas) and tumors originating from nearby structures (thalamic astrocytomas, tumors of Plexus choroid).[52]

The most common tumors of the pineal region are the germinative ones, responsible for 40 to 80% of the total number, with germinoma being the main representative.[47,53] Tumors of the pineal parenchyma have a prevalence of 11 to 40% of pineal tumors, and can be divided into pineocytes, pineoblastomas, papillary tumors of the pineal region (PTPRs) and pineal parenchymal tumor of intermediate differentiation (PPTID).[53,54]

Tumors derived from supporting tissues or structures around the pineal gland include astrocytomas, ependymomas, papillomas and carcinomas of the choroid plexus, lymphomas, gangliogliomas, dermoid and epidermoid cysts, meningiomas and metastases.[55] According to our literary review, our study is the second case of trochlear schwannoma that has its origin in the pineal region.

The therapeutic options for non-vestibular schwannomas include clinical observation through serial image analysis, microsurgery and stereotactic radiosurgery.[56] Usually, when treating asymptomatic tumors, clinical observation may be a reasonable alternative, especially in elderly patients or those with other comorbidities, since these tumors usually have slow growth.[56]

According to our literature review, the most used surgical approach in the cases of trochlear schwannoma is the transtemperal subtemporal, performed in 13 of 36 cases, followed by transpetrosal (7 cases), lateral suboccipital (7 cases), pterional (5 cases), supracerebellar infratentorial (2 cases) supracerebellar subentential (1 case), and paraoccipital trastentorial (1 case) (►Table 1). In one case, the surgical approach was not described.[54] In our case, we used the intratentorial supracerebellar approach, the same one used in the Ture study.[23] The surgical approach used in the previous case of trochlear schwannoma in the pineal gland was the posterior transtentorial interhemispheric paraoccipital.[35]

The postoperative of the previously reported cases demonstrated the difficulty and vulnerability in preserving the trochlear nerve during surgery (►Table 1). Among the 17 cases that did not present preoperative paralysis, 12 developed a new trochlear paralysis in the postoperative period, and in only 2 cases there were significant improvements in trochlear paralysis in the postoperative follow-up. From the 17 cases in which the patients had IV paresis, in only 1 trochlear function improvement happened, with diplopia improvement between 4 weeks and paralysis improvement after 2 years postoperatively. However, in this same case, the MRI 3 months after surgery showed a small remnant tumor.[31]

Conclusion

Trochlear nerve schwannomas are extremely rare; however, they generally have a good neurosurgical resectability. Our work presents an unusual trochlear nerve schwannoma, corresponding to the wide range of the possible pathological diagnoses when the region of the pineal gland is involved. In addition, our case also contrasts with the majority of the cases reported in the literature, which shows tumor epicenter furthest from nerve trajectory.

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