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**Case Report**

# A rare presentation of systemic complication of hypoxic ischemic encephalopathy in post cochlear implant child

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## ABSTRACT

Cochlear implant is a small, complex electronic device to restore some hearing in profoundly deaf or severely hard-of hearing people when organ of corti is not developed or destroyed by injury to such an extent that no hearing can be obtained by hearing aids. Many classifications have been proposed for cochlear implant related problems .We report a case of post cochlear implant child who reported to OPD with complaint of progressive weakness of both upper and lower limb which developed 1 year after surgery ,which parents attributed it as a complication of surgery. On retrospective analysis all patient preop investigations were normal. After detailed examination of the child, neurophysician concluded that child is suffering from global developmental delay with dystonic cerebral palsy. MRI brain and cochlea which was done before the surgery which showed T1 hypointense,T2 and FLAIR hyperintense ,bilateral symmetrical foci with high ADC values are seen in bilateral lentiform nuclei .It has been reported that the bithalamic hyperdensities on CT and/or MRI in severely asphyxiated neonates, were suggested of a distinctive pattern of brain injury which might be useful as an early predictor of status marmoratus,afrequent pathological correlate of dyskinetic cerebral palsy. Approximately 1% of infants who survived perinatal asphyxial HIE would develop delayed-onset dystonia in the course of 7 to 13 years (median 10 years) after birth.

**Keywords:** Cochlear implant, Systemic complication, Hypoxic ischemic encephalopathy

## INTRODUCTION

Cochlear implantation has been established to restore the hearing in children or adults who are having bilateral severe to profound sensorineural hearing loss and had no benefit from hearing aid for at least 6 months.CI implant surgery is an elective procedure, so surgical complications are not justifiable. So surgeons performing CI surgery should have adequate knowledge regarding all the possible complications. Many classifications have been proposed for cochlear implant related problems like minor and major based on severity of symptoms and the need for revision surgery device related, surgical and medical complication, local and systemic complications. Here we present a case of postcochlear implant surgery patient reported with a systemic complication 1 year after cochlear implant surgery which is unrelated to surgery, but can develop coincidentally in any post cochlear implant child who had a hypoxic insult at birth.[1]

## CASE REPORT

A 6year old child came to ENT outpatient department with history of cochlear implant surgery 1 year back and she has received 6 cycles of speech therapy. Intraoperative ART (Auditory response threshold) and NRT (Neural response telemetry) showed value within normal range. C arm Xray showed electrode arrays implanted within cochlea. Post operative period was uneventful and the child was discharged. The child was brought 1 year after cochlear

implant surgery by his grandfather with c/o progressive weakness of both upper and lower limbs. On history of these symptoms developed gradually over a period of 1 year. Since the symptoms appeared coincidentally after the surgery, the patient's relatives were correlating that the symptoms developed due to the complications of the cochlear implant surgery.

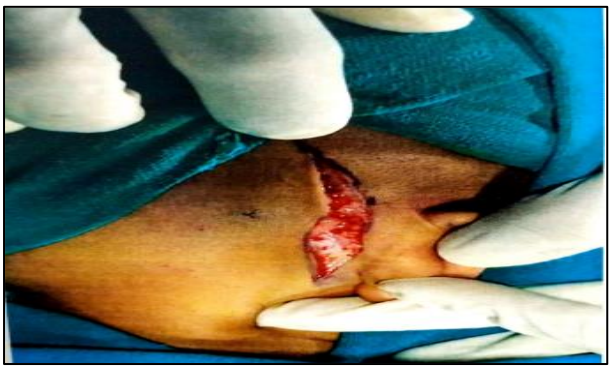

**Figure 1: Intraoperative picture of incision site.**

**Figure 2: Presentation of child 1 year after surgery.**

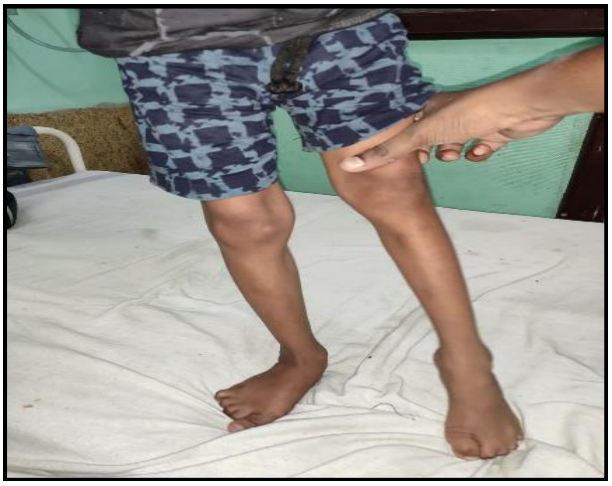

**Figure 3: Presentation of child 1 year after surgery.**

On examination, there was dystonia of both the upper and lower limbs. Neurophysician opinion was sorted. After detailed examination of the child they came to a conclusion that child is suffering from global developmental delay with dystonic cerebral palsy. They reevaluated the MRI brain & cochlea which was done before the surgery which showed T1 hypointense,T2 and FLAIR hyperintense ,bilateral symmetrical foci with high ADC values are seen in bilateral lentiform nuclei which were not seemed significant at the time of surgery. However they correlate it with the complication and suggested it as the late systemic complication of hypoxic ischemic encephalopathy. Preop Temporal bone CT showed no abnormalities. Preoperative file of the patient was reviewed in which child had developmental delay and history of suggested that the child didn't cry immediately after birth which closely supported the diagnosis.

## DISCUSSION

Cochlear implant surgery is a rewarding surgery with regaining of special senses to a child. Since it improve the quality of life of a child we can't deny the surgery to a child by anticipating the rare complications. However, we should always advice the parents that children with hypoxic history at birth with MRI suggestive of hypointense lesions in basal ganglia can develop dystonia at any point of their life. So, ENT surgeon should always be aware of this entity and planning of surgery is extremely important.

Numerous classifications have been made to describe the complications of cochlear implant surgery. On the basis of onset of complications, it has been defined as early (within 3 months postop) like wound gaping, surgical wound swelling, tinnitus, vertigo and late (more than 3 months postop) like cholesteatoma, device failure and electrode dislocation. On the basis of severity of complication and the need for revision surgery or medical treatment it has been described as major and minor respectively. Bathia and Venail1 cited death, meningitis, surgery without reimplantation due to large scalp necrosis, severe infection, ear drum perforation, receiver repositioning and cholesteatoma as major while transient facial palsy, scalp hematoma, infections that resolved without recourse to surgery, tinnitus, and facial stimulation as minor complications. Reviewing through the literature.[2] In a study by Halawani and et al in 1027 cochlear implant recipients post-operative complication were reported in 10.2%.[3] Minor complication were most often encountered (9.5%) in which swelling (wound seroma or hematoma) was the most common. The major complication was only 0.7% with meningitis in only 1 patient. In a study by Ciorba and et al in 432 consecutive cases of cochlear implant, overall rate of complication was 9.1% in which most of them were minor.[4] Wound swelling and infections represent the most common complications. 3% patients underwent explanation followed by reimplantation. Various online journals were reviewed but couldn't find even a single case report showing the systemic

complication of a congenital insult attached with cochlear implant surgery. So, this made me to share my case with all the cochlear implant surgeons who may come over a case like mine at any point of their career.

Dystonia and other movement disorders may arise: immediately after acute brain injury, as acute neurological deficits are improving, and after a prolonged period of apparent neurological stability.[5] Dystonia is a rare sequela of perinatal hypoxic-ischemic injury.[6-9] In 20 cases of delayed-onset dystonia reported after perinatal asphyxia, the interval between dystonia and perinatal asphyxia varied from 1 to 32 years (mean delay 12.3 years).[6,9] It has been reported that the bithalamic hyperdensities on CT and/or MRI in severely asphyxiated neonates, were suggested of a distinctive pattern of brain injury which might be useful as an early predictor of status marmoratus, a frequent pathological correlate of dyskinetic cerebral palsy.[10,11] It was hypothesized that a delay in onset of the movement disorder after asphyctic brain injury might reflect the time required for remyelination, inflammatory changes, oxidation reactions, maturational or aberrant synaptic reorganization, trans-synaptic neuronal degeneration, or denervation supersensitivity to occur.[5] Based on the study by Cerovac et al and others, approximately 1% of infants who survived perinatal asphyxial HIE would develop delayed-onset dystonia in the course of 7 to 13 years (median 10 years) after birth.[12] This has to be kept in mind by each one of us which can occur as a very rare complication following cochlear implant surgery even though it is unrelated to the surgery ,it can impair the post surgical performance of the child as well as create a panic button in the parents. The surgeon should always discuss with the parents about antenatal, natal and postnatal causes of sensorineural hearing loss and the post op complications that can occur related to surgery as well as the delayed systemic manifestation of the congenital insult as mentioned in my case.

## CONCLUSION

Late onset dystonia can occur as a very rare complication following cochlear implant surgery even though it is unrelated to the surgery, it can impair the post surgical performance of the child as well as create a panic button in the parents. The surgeon should always discuss with the parents about antenatal ,natal and postnatal causes of sensorineural hearing loss and the post op complications that can occur related to surgery as well as the delayed systemic manifestation of the congenital insult as mentioned in my case.

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